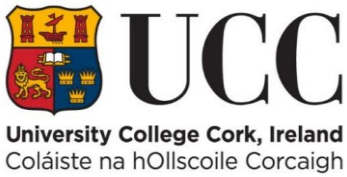
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Rethinking stillbirth through behaviour change

Thesis presented by

Ms. Tamara Escañuela Sánchez

or the degree of

Doctor of Philosophy

University College Cork, School of Medicine and Health

Research conducted at the INFANT Centre and the Pregnancy Loss Research Group,
Department of Obstetrics and Gynaecology, Cork University Maternity Hospital, Cork.

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Dr. Sarah Meaney, Prof. Molly Byrne.

30th December 2022

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List of Abbreviations

ACCODS	Authority, Accuracy, Coverage, Objectivity, Date, Significance
ACOG	American College of Obstetricians and Gynaecologists
aOR	Adjusted Odds Ratio
APEASE	Affordability, Practicability, Effectiveness and Cost-Effectiveness, Acceptability, Safety and Equity
aRR	Adjusted Risk Ratio
BCT	Behaviour Change Technique
BCTs	Behaviour Change Techniques
BCTTv1	Behaviour Change Technique Taxonomy (version 1)
BCW	The Behaviour Change Wheel
BPPs	Biophysical Profiles
CASP	Critical Appraisal Skills Program
CBT	Cognitive Behavioural Theory
CC4SP	Canadian Collaborative for Stillbirth Prevention
CI	Confidence Interval
COM-B	Capability, Opportunity, Motivation, Behaviour
COVID-19	Coronavirus Disease
CS	Caesarean Section
CUMH	Cork University Maternity Hospital
DC	Dichorionic
EPOC	The Cochrane Effective Practice and Organisation of Care
GP	General Practitioners
GRADE-CERQual	Confidence In the Evidence from Reviews Of Qualitative Research
GWG	Gestational Weight Gain
HCPs	Healthcare Professionals
HIC	High-Income Countries
HR	Hazard Ratio
HSE	Health Service Executive
ICD	International Statistical Classification of Diseases And Related Health Problems
ICD-10	International Classification of Diseases, Version 10
ICD-PM	International Statistical Classification of Diseases And Related Health Problems Perinatal Mortality
ICSI	Intracytoplasmic Sperm Injection
IMPROVE	Improving Perinatal Mortality Review and Outcomes Via Education

ISA	International Stillbirth Alliance
IUGR	Intrauterine Growth Restriction
IVF	In Vitro Fertilisation
JBI	Joanna Briggs Institute Critical Appraisal Checklist
KPIs	Key Performance Indicators
MBRRACE-UK	Mothers And Babies: Reducing Risk Through Audits and Confidential Enquiries Across The UK
MC	Monochorionic
MCDA	Monochorionic-Diamniotic
MCQIC	Maternity And Children Quality Improvement Collaboration
MECC	Make Every Contact Count
MHCP	Maternity Health Care Providers
MiNESS	The Midland and North of England Stillbirth Study
NCEC	National Clinical Effectiveness Committee
NHS	National Health Service
NICE	The National Institute for Health and Care Excellence
NMSP	National Maternity Strategy Plan
NPEC	National Perinatal Epidemiology Centre
NST	Non-Stress Tests
NWIHP	National Women and Infants Health Programme
OR	Odds Ratio
PAN	Perinatal Audit Netherlands
PAPP-A	Pregnancy-Related Plasma Protein A
PGDM	Pre-Gestational Diabetes Mellitus
PHN	Public Health Nurse
PIGF	Placental Growth Factor
PM	Perinatal Mortality
PMMRC	Perinatal & Maternal Mortality Review Committee
PMR	Perinatal Mortality Rates
PPI	Public And Patient Involvement
PreWG	Prevention Working Group
PRN	Perinatal Registry Netherlands
RCOG	Royal College of Obstetricians and Gynaecologists
RCPI	Royal College of Physicians of Ireland
RFM	Reduced Fetal Movements
ROI	Republic Of Ireland
RR	Risk Ratio
RSE	Relationships And Sex Education
SBB	Safer Baby Bundle

SBLCB	Saving Babies' Lives Care Bundle
SCT	Social Cognitive Theory
SGA	Small-For-Gestational-Age
SIDS	Sudden Infant Death Syndrome
SPSP	Scottish Patient and Safety Programme
sRR	Standardised Rate Ratio
SSRIs	Selective Serotonin Reuptake Inhibitors
TDF	Theoretical Domains Framework
THCA	Tetrahydrocannabinolic Acid
TPB	Theory Of Planned Behaviour
TTM	Transtheoretical Model of Health Behaviour Change
WHO	World Health Organisation

Declaration

This is to certify that the work I am submitting is my own and has not been submitted for another degree, either at University College Cork or elsewhere. All external references and sources are clearly acknowledged and identified within the contents. The work, upon which this Thesis is based, was carried out in collaboration with a team of researchers and supervisors who are duly acknowledged in the text of the Thesis. I have read and understood the regulations of University College Cork concerning plagiarism.

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Research Article	Conference/Event
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Escañuela Sánchez, T., Meaney, S. & O'Donoghue, K. Stillbirth and risk factors: an evaluation of Irish and UK websites.	*Poster Presentations:* ▪ New Horizons Conference, 5th December 2019, UCC, Cork. ▪ British Maternal & Fetal Medicine Society BMFMS Perinatal Medicine Conference 2020. Cancelled due to COVID-19. ▪ 17th Annual Psychology, Health and Medicine Conference. Psychological Society of Ireland, 22nd May, 2020. Cork. Changed format due to COVID-19. ▪ Stillbirth Summit 2021. Star Legacy Foundation. 23rd -24th June 2021. Online. ▪ 18th Annual Psychology, Health and Medicine Conference, Division of Health Psychology of the Psychological Society of Ireland. 12th - 14th May 2021. Online
Escañuela Sánchez, T., Matvienko-sikar, K., Linehan, L., Donoghue, K. O., Byrne, M., & Meaney, S. Facilitators and barriers to substance-free pregnancies in high-income countries: A meta-synthesis of qualitative research	*Oral Presentations:* ▪ New Horizons Conference, 3rd December 2020, UCC. Cork. ▪ Stillbirth Summit 2021. Star Legacy Foundation. 23rd -24th June 2021. Online. ▪ Annual Conference of the European Health Psychology Society. 23rd - 27th August 2021. Online.

	▪ 18th Annual Psychology, Health and Medicine Conference, Division of Health Psychology of the Psychological Society of Ireland. 12-14 May 2021. Online. ▪ Society for Reproductive and Infant Psychology (SRIP). Annual conference. 8th – 10th September 2021. Online. ▪ International Stillbirth Alliance (ISA) Conference. 11th - 13th November 2021. Online. *Poster Presentation:* ▪ International Society for Prenatal Diagnosis (ISPD) 25th International Conference on Prenatal Diagnosis and Therapy, 6th – 8th June 2021.
Escañuela Sánchez, T, Linehan, L., O'Donoghue, K., Byrne, M., & Meaney, S. Facilitators and barriers to seeking and engaging with antenatal care in high-income countries: A meta-synthesis of qualitative research.	*Oral Presentation:* ▪ Stillbirth Summit 2021. Star Legacy Foundation. 23-24 June 2021. ▪ International Stillbirth Alliance (ISA) Conference. 11-13 November 2021. Online *Poster Presentation:* ▪ New Horizons Conference, 3rd December 2020, UCC. Cork. ▪ Society for Maternal and Fetal Medicine SMFM's Annual Pregnancy Meeting. 25th-30th January 2021. Online. ▪ NPEC Study Day 2021. ▪ International Society for Prenatal Diagnosis (ISPD) 25th International Conference on Prenatal Diagnosis and Therapy, 6th – 8th June 2021. ▪ NPEC Study Day, 21st January 2022. Online.

Escañuela Sánchez, T, Meaney, S., O'Connor, C., Linehan, L., O'Donoghue, K., Byrne, M., & Matvienko-Sikar, K. Facilitators and barriers influencing weight management behaviours during pregnancy: a meta-synthesis of qualitative research.	*Oral Presentation:* ▪ International Stillbirth Alliance (ISA)/ International Society for the Study and Prevention of Perinatal and Infant Death (ISPID) Conference. 11-13 November 2021. Online. ▪ Society for Reproductive and Infant Psychology (SRIP). Annual conference. 15th - 16th September 2022. Online. *Poster presentations:* ▪ Annual Conference of the European Health Psychology Society. August 2022. Online. ▪ International Society for Prenatal Diagnosis (ISPD) 26th International Conference on Prenatal Diagnosis and Therapy, 19th – 23rd June 2022, Online. ▪ International Stillbirth Alliance Conference. 15th – 17th September 2022. Salt Lake City, Utah (USA).
Escañuela Sanchez, T., Matvienko-Sikar, K., Meaney, S., Byrne, M., O'Donoghue, K. Exploring first time mothers' experiences and knowledge about modifiable risk factors for stillbirth: a qualitative study.	*Oral presentations:* ▪ Society for Reproductive and Infant Psychology (SRIP). Annual conference. 15th - 16th September 2022. Online. ▪ Psychological Society of Ireland Annual Conference, 9th - 11th November 2022. Portlaoise, Ireland. *Poster presentations:* ▪ Annual Conference of the European Health Psychology Society. 23rd – 27th August 2022. Online. ▪ International Society for Prenatal Diagnosis (ISPD) 26th International Conference on

	Prenatal Diagnosis and Therapy, 19th – 23rd June 2022, Online. ▪ College of Medicine and Health Research Conference, "from Molecules to People". 22nd September 2022. UCC ▪ International Stillbirth Alliance Conference. 15th – 17th September 2022. Salt Lake City, Utah (USA). ▪ BMFMS. British Maternal and Fetal Medicine Society Annual Conference. 17th – 18th November 2022. Birmingham (UK).
Rutherford, E., **Escañuela Sánchez, T.,** O'Donoghue, K (2022). Exploring healthcare professionals' experiences when communicating with pregnant women about stillbirth	*Oral presentation:* ▪ Psychological Society of Ireland Annual Conference, 2022. Portlaoise, Ireland. *Poster presentations:* ▪ International Stillbirth Alliance Conference. 15th – 17th September 2022. Salt Lake City, Utah (USA).
Escañuela Sanchez, T., Matvienko-Sikar, K., Meaney, S., Byrne, M., O'Donoghue, K. The RELEVANT Study: Rethinking stillbirth through behaviour change.	*Poster presentation:* ▪ International Stillbirth Alliance Conference. 15th – 17th September 2022. Salt Lake City, Utah (USA).

Education, training, and events

Year	Course	Provider
2018	Webinar: Core Outcome Sets in Randomised Trials.	Prof. Declan Devane, Health Research Board, NUI Galway
2018	Study day: The Rule rather than the Exception: Embedding Implementation Science in Population and Health Services Research to Promote Effective Translation.	University College Cork
2019	Designing Effective Interventions for Health Behaviour Change: An Introduction.	Prof. Molly Byrne, NUIG Galway.
2019	PG6001 STEPS - Scientific Training for Enhanced Postgraduate Studies	UCC Postgraduate Module
2019	PG6008 - Qualitative Data Analysis and Computer Assisted Qualitative Data Analysis Software for the Social Sciences and Humanities.	UCC Postgraduate Module
2019	PG6009 Graduate Information Literacy Skills.	UCC Postgraduate Module
2019	Introduction to Qualitative Research Methods.	University of Bristol
2020	PG6024 Qualitative Research Inquiry.	UCC Postgraduate Module
2020	Webinar: Covid Realities: Interviewing at a distance: navigating practical, emotional, and methodological challenges.	Nuffield Foundation, University of York.

2020	Webinar: An introduction to the practical applications of the Theory Domains Framework.	Dr. Andrea Patey. Health Behaviour Change Research Group, NUI Galway.
2020	Webinar: Theoretical conceptualisation of pregnancy & motherhood: opportunity for health behaviour change.	The British Psychological Society.
2020	6th Annual Digital Health Conference. Behaviour Change for Health: New and emerging science and technologies.	
2021	Introduction to Qualitative Evidence Synthesis.	Evidence Synthesis Ireland.
2021	Using a GRADE-CERQual Approach in Qualitative Evidence Synthesis Online Training	Evidence Synthesis Ireland.

Abstract

Background

Worldwide, two million babies are stillborn every year. While the majority of stillbirths occur in low and middle-income countries, stillbirth is still one of the most common adverse pregnancy outcomes in high-income countries. In Ireland, the latest National Perinatal Mortality Clinical Audit report states a stillbirth rate of 4.20 per 1000 births for the year 2020, showing an increase compared to previous years. The belief that reduced stillbirth rates in high-income countries cannot be achieved is refuted by differences in stillbirth rates across different countries.

Although not all stillbirths are preventable, there has been a call made in high-income countries to focus on risk factors for stillbirth, in order to reduce stillbirth rates. These risk factors include sociodemographic factors, medical factors, obstetric history related factors, placental and fetal related factors as well as behavioural and lifestyle related factors. Some of these factors are modifiable through medical management or through behaviour change modification. This Thesis focuses on risk factors that have potential to be modified through maternal behaviour change interventions: substance use (smoking, alcohol, and illicit drug use), high BMI, sleep position, and attendance at antenatal care.

Strategies have been successfully implemented internationally to reduce stillbirth rates by designing and implementing care bundles that, amongst other elements, take into consideration the modifiable/behavioural risk factors for stillbirth. However, in Ireland, no such initiatives have been developed, although recommendations have been made that support their development. For behaviour change interventions or public health initiatives to have the best possible success in reducing the rates of stillbirth, they need to be designed with a solid evidence base. Hence, the overall objective of this Thesis was to build the evidence base to enhance the understanding of the modifiable behavioural risk factors for stillbirth and pregnancy. Further, this evidence base is needed to inform future development of a

behaviour change intervention that could be part of a care bundle with the objective of reducing stillbirth rates in Ireland.

Methodology

To address the Thesis´s aims, both qualitative and quantitative methods were utilised. Applying multiple methods to explore a phenomenon provides flexibility to analyse different aspects of it in the different studies. Initially, a non-systematic review of the literature was conducted to identify the target behavioural risk factors that this project was going to focus on (Chapter 2). A website quantitative content analysis was conducted to assess the availability of information related to stillbirth and behavioural risk factors for stillbirth in Irish and UK websites (Chapter 3). For this study, descriptive and inferential statistics were utilised. Further, three systematic qualitative meta-synthesis were conducted to identify facilitators and barriers to modify identified behavioural risk factors according to the pregnant women's experience (Chapters 4-6). A meta-ethnographic approach as described by Noblit and Hare was adopted to conduct these qualitative meta-syntheses. Reflexive Thematic Analysis as described by Braun and Clarke, with a constructivist approach, was used to conduct a qualitative semi-structured interview study with postpartum women about their experiences of stillbirth information provision and behaviour change during their antenatal care (Chapter 7). Finally, a systematic review of interventions designed in the context of stillbirth prevention that targeted behavioural risk factors was conducted (Chapter 8). This systematic review had the objective of identifying which behaviour change techniques (BCTs) have been used to date.

Results

The findings of the literature review (Chapter 2) showed that the modifiable behavioural risk factors with the strongest evidence of associations with stillbirth were substance use, smoking, heavy drinking and illicit drug use, lack of attendance and compliance with antenatal care, weight-related risks, and sleep position. The quantitative content analysis of websites (Chapter 3) revealed that information

about stillbirth and behavioural risk factors for stillbirths was scarce on websites directed at the pregnant population, with only one website containing all the information sought. Five main areas of concern were identified across the three meta-synthesis of qualitative research of facilitators and barriers influencing women's prenatal health behaviours (Chapters 4-6), regardless of the behaviour explored: 1) health literacy, awareness of risks and benefits; 2) insufficient and overwhelming sources of information; 3) lack of opportunities and healthcare professionals attitudes interfering with communication & discussion; 4) social influence of environment, and 5) social judgement, stigmatisation of women and silence around stillbirth. Further, the qualitative study with postpartum women (Chapter 7) revealed that women perceived behaviour change during pregnancy as easy and natural, as they were focused on obtaining the best outcomes for their babies. Although women had high levels of awareness regarding health advice, their awareness about stillbirth was very limited. Women reported a lack of discussion about stillbirth and behavioural risk factors during their antenatal care; however, most women showed a positive disposition towards receiving this information because "knowledge is key", as long as it is done in a "sensible manner". The systematic review of interventions designed in the context of stillbirth prevention identified nine relevant interventions. From the BCT coding, it was established that the most common BCT used was "information about health consequences", followed by "adding objects to the environment" (Chapter 8).

Conclusion

This research makes a valuable contribution to the understanding of the maternal behaviours associated with an increased risk of stillbirth, and it provides a necessary evidence-base to inform future prevention strategies to reduce rates of stillbirth in Ireland and in similar healthcare settings. This research sought to incorporate women's voices and use research methods to produce high-quality results that meet the research objectives. The findings from the studies in this Thesis support four overarching topics and highlight issues related to 1) health literacy and sources of

information, 2) relationships with healthcare professionals (HCPs), 3) healthcare systems and structural barriers, and 4) interpersonal, social and structural factors. In response to the research findings, several recommendations are made in relation to policy, practice and research which are grounded on women's experiences during pregnancy. Regarding policy, these recommendations include improving education and information sources for women and HCPs, providing pregnancy-specific supports, utilising community services to support women with behaviour change, and developing a care bundle to tackle the behavioural risk factors for stillbirth. Furthermore, the work practice recommendations made include developing clinical guidelines to support HCPs in providing care to pregnant women, and prioritising health promotion during antenatal care. These priorities might also serve to help funders and researchers to design and conduct policy-relevant research. The key future research areas identified by this Thesis are in relation to the involvement of PPI representatives, the assessment of the quality of the available sources of information and the further exploration of potential facilitators and barriers to modify pregnant women's sleeping position from a qualitative perspective. In addition, this Thesis proposes a detailed process to continue building on the work set out in the different studies to develop a pregnancy-specific behaviour change intervention for the modifiable behavioural risk factors for stillbirth in the future.

Chapter 1 - Introduction

Chapter 1 – Introduction

1.1 Introduction

Stillbirth is one of the most devastating pregnancy outcomes that parents can face, and unfortunately, it is also one of the most common[1]. Stillbirth is a global health burden that affects 2 million babies every year[2]. The rates of stillbirth vary across different high-income countries, with a rate of 4.20 per 1000 births in Ireland in 2020[3]. The idea that no stillbirth is preventable is a myth[4,5], and previous research has highlighted the importance of focusing stillbirth preventive efforts on tackling risk factors for stillbirth[6].

Some of the risk factors that have been associated with stillbirth are related to medical issues affecting the mother and/or the foetus, while others include sociodemographic factors and behaviours that are associated with an increased risk[7]. The behavioural risk factors for stillbirth such as substance use, lack of attendance at antenatal care, sleep position and weight related risks have the potential to be modified. Thus, increasing our understanding of these behaviours is essential to inform the development of strategies to address them[7].

This chapter provides an overview of the different issues that are limiting stillbirth prevention efforts. Starting with differences in definitions which have an important influence on the reporting of rates in different countries (Section 1.1 and 1.2) and an overview of the main causes for stillbirth (Section 1.3). It then provides an overview of the impact that experiencing a stillbirth has on affected families at different levels (personal, couples, siblings, and in terms of future pregnancies), on healthcare staff (Section 1.4), as well as the economic burden associated with stillbirth (Section 1.5). This is followed by the differences in the challenges that low-, middle- and high-income countries face when trying to reduce their rates of stillbirth (Section 1.6). Section 1.7 will outline the different risk factors for stillbirth, ending with a section on behavioural risk factors (Section 1.7.5), which are the main focus of this Thesis.

Further, an overview of the previous efforts that have been conducted to date internationally to reduce the rates of stillbirth will be outlined (Section 1.8), followed by a brief explanation as to why this is relevant in the Irish context (Section 1.8.3). Lastly, and considering that the main focus of this project is the behavioural risk factors for stillbirth, a section is dedicated to behaviour change during pregnancy (Section 1.9.1), followed by behaviour theory and behaviour intervention design relevant to the context of stillbirth prevention (Sections 1.9.2 and 1.9.3).

1.2 Stillbirth definition

In Ireland, stillbirth is defined as a baby born with no signs of life at 24 weeks gestation or later, or with a birthweight of 500 grammes or more[8]. However, stillbirth definitions differ depending on the country. The World Health Organisation (WHO) proposes different definitions for stillbirth depending on what purpose that definition is going to be used for. For general statistics and registration, the International Classification of Diseases, 10th revision (ICD-10) defines stillbirth as "*the death of a fetus that has reached a birth weight of 500 g, or if birth weight is unavailable, gestational age of 22 weeks or crown-to-heel length of 25 cm. Within this category, ICD classifies late fetal deaths (greater than 1000 g or after 28 weeks) and early fetal deaths (500–1000 g or 22–28 weeks)"*[9]. The WHO also recommends that all babies born of at least 500 grammes should be included in statistics about perinatal death. For international comparison and reporting, the WHO recommends using a gestational week threshold of ≥28 weeks and a birthweight of ≥1000 grames[10]. However, 35% to 50% of stillbirths in high-income countries occur below the WHO recommended cut-offs of ≥28 weeks gestation and birthweight of ≥1000 grames[6,11]. The use of the WHO definition leads to an under-reporting of stillbirths that occur at less than 28 weeks, which represent those showing the least improvements in reductions in high-income countries (HIC)[6]. Lawn et al. (2016) propose that the definition should be based only on a gestational age threshold, as this factor is a better predictor of viability than birthweight[1].

Examples of international differences in the definition of stillbirth can be found in the literature[10,12]. Kelly et al. recently conducted a systematic review of the literature to explore the different definitions used in high-income countries around the world, stillbirth and mortality rates and how these rates are affected by standards of care, especially around resuscitation[12]. The review findings concluded that within the EU, the legal classification of stillbirth registration differs from ≥22-28 weeks gestation and ≥500-1000 g[12]. Additionally, the US, Australia, New Zealand, Azerbaijan and Canada use 20 weeks gestation as their gestational age lower limit, and Norway uses 12 weeks. The authors also concluded that some of the variations in survival rates can be a consequence of the application of full treatment versus comfort care in very premature babies; these decisions in care are based on the perception of the viability of the baby[12]. Full treatment care has curative intent, whereas the objective of comfort care is pain and symptom relief, without curative intent. Kelly et al. propose in their review that Ireland should redefine its definition of stillbirth and adopt the definition recommended by multiple professional organisations of ≥22 weeks gestational age or ≥400 g as a birthweight[12].

The survival rates of very premature babies (under 25 weeks gestation) have been increasing over time thanks to improvements in medical care[12,13], however, recommendations regarding the baby's care also vary across countries. Guillén et al. conducted a systematic review of guidelines for the management of very premature deliveries (22-25 weeks gestation). The authors concluded that there is apparent consensus regarding most guidelines recommending comforting care for babies born at 22 weeks gestation and active treatment for babies born at 25 weeks gestation. The recommendations are much more variable when it comes to babies born at 23 and 24 gestational weeks[13]. These differences and inconsistencies in recommendations about care influence the stillbirth definition used.

Using different definitions is problematic because it causes discrepancies and inaccuracies in international reporting and comparison, and potentially excludes some cases of stillbirths from the epidemiological data. Furthermore, the differences

in definition also have implications for bereaved families, as their rights might differ depending on the gestation of stillbirth. In Ireland, for example, women who have a stillborn child at >24 week gestation or a birthweight of ≥500 g currently can avail of 26 weeks of paid maternity leave[14].

1.3 Stillbirth rates

Stillbirth is one of the most common adverse pregnancy outcomes, with rates similar to neonatal deaths, childhood deaths (1 to 5 years old) and adult deaths associated with AIDS[15], and much higher rates than Sudden Infant Death Syndrome (SIDS)[16]. The rates of stillbirth are influenced by the different definitions used in each country, and hence international comparison is difficult. According to a systematic assessment conducted in 2019, globally, approx. 2 million babies were born still in 2019 (at 28 weeks gestation or more), with the global stillbirth rate being 13.9 per 1,000 total births[17]. The highest rates of stillbirth in 2019 were estimated for west and central Africa at 22.8 stillbirths per 1,000 births and followed by eastern and southern Africa at 20.5 per 1,000 births and south Asia at 18.2 stillbirths per 1,000 births[17]. This assessment concluded that 83.6% of all stillbirths occurred in low-income and lower-middle-income countries, while HICs accounted for 1.9% of the global rates of stillbirths[17].

In high-income countries, the estimated average rate of stillbirth at 28 weeks gestation is 3.5 per 1,000 births. Data published in the Lancet showed that these rates vary from 1.3 in Iceland to 8.8 in Ukraine[6]. The latest Euro-Peristat report published in 2018 provides data on the different European countries in 2015, according to this report, the European stillbirth rate in 2015 was 2.7 per 1000 total births[18]. In 2015, stillbirth rates at or after 24 weeks gestation were the highest in Bulgaria (6.9 per 1000 births), followed by Hungary (4.3 per 1000 births), Croatia (4.3 per 1000 births) and England and Wales (4.2 per 1000 births), and the lowest rates were found in Cyprus (1.8 per 1000 births), followed by Iceland (2.0 per 1000 births), Finland (2.5 per 1000 births), Denmark (2.5 per 1000 births), and Austria and the Netherlands

(both at 2.9 per 1000 births)[18]. Most European countries had rates between 2.5 and 4.5 stillbirths per 1000 births at 24 weeks gestation or after[18].

Most recent data shows that, in Ireland, according to the latest data from the National Perinatal Mortality Clinical Audit conducted by the National Perinatal Epidemiology Centre (NPEC), the rate of stillbirth (at ≥24 weeks gestation or with a birthweight of ≥500 g) was 4.20 per 1,000 births in 2020[8] which is similar to the UK's rate in 2015[19]. The report states that since 2012, the perinatal mortality rates have levelled off, and since 2018, there has been an increase in perinatal mortality rates in Ireland, which are also observed in the stillbirth rates (See figure 1.1)[8].

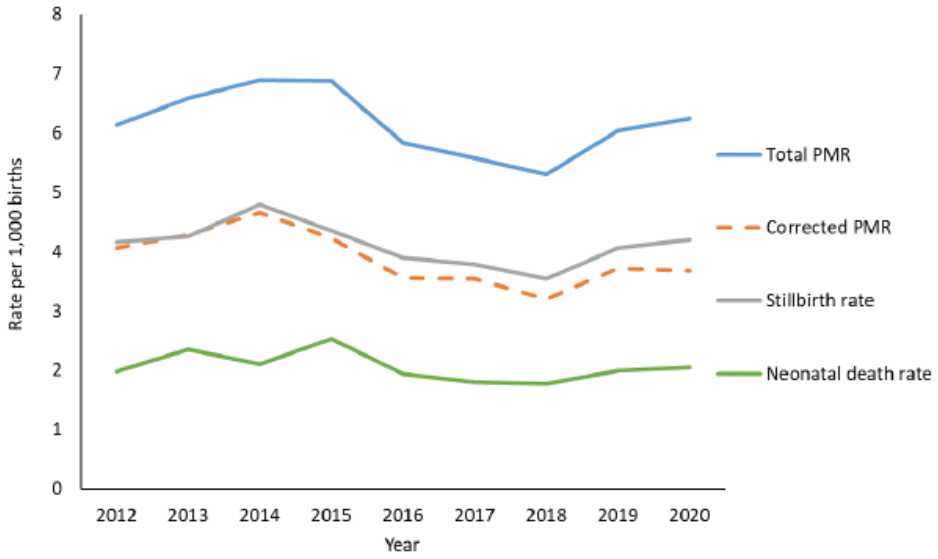


Figure 1. 1: Obtained with permission from the Perinatal Mortality National Clinical Audit in Ireland Annual Report 2020[8].

In the UK, the Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK (MBRRACE-UK) latest Perinatal Mortality Surveillance Report, reported a stillbirth rate of 3.33 per 1000 births in 2020, which is 21% lower than in 2013 (4.20 stillbirths per 1000 births)[19]. In the Netherlands, the national data on perinatal deaths (from ≥24 weeks or if missing ≥500 grams and up to 7 days post

birth) was 5 per 1000 births in 2020, no specific data on stillbirth rates is available[20]. The Netherlands had reported a reduction in their perinatal death rates of 6.2% per year from 2000 to 2015, whereas the Irish reduction was 3.5% per year for the same period[3].

The rates of stillbirth globally decreased by over 35% in the last 20 years (21.4 per 1000 births in 2000 to 13.9 in 2019), mostly in east Asia and the Pacific, followed by Eastern Europe and Central Asia[17]. However, this rate reduction is slow when compared with the decreases in mortality rates of children younger than 5 and also compared with progress in reducing neonatal mortality rates[17]. In high-income countries, the rates of stillbirth have remained steady, showing very small reductions in the last decade[21]. The differences in stillbirth rates between the different high-income countries and within the same high-income country show that there is a possibility to reduce the rates of stillbirth even further[21].

1.4 Main causes of stillbirth

Some challenges are associated with establishing the most common causes of stillbirth. Currently, there is no global system for reporting causes of death of stillbirth[22]. The WHO suggests using the International Statistical Classification of Diseases and Related Health Problems (ICD) for the classification of perinatal deaths[9]. In 2016, the WHO developed the ICD Perinatal Mortality (ICD-PM), which is an adaptation of the ICD to better capture perinatal mortality causes[5]. The ICD-PM differs from the ICD in that it captures the timing of death (antepartum, intrapartum or postpartum); it provides a multi-layered approach for the classification of the cause of death and it links the maternal factors contributing to the perinatal death[23]. Reinebrant et al. (2017) conducted a systematic review to identify the causes of stillbirth reported globally using the ICD-PM. The authors identified 85 reports from 50 different countries. High-income countries were more likely to define stillbirth using a lower gestational age of 20-24 weeks, whereas low-income countries tended to use 28 weeks[22]. The authors were able to identify 21 clinical classification systems in 41 of the reports, with the ICD classification system being the most commonly used

in high-income countries and middle-income countries[22]. Another 20 reports provided information on the cause of death without referring to any existing classification system. According to these authors, in high-income countries the category most commonly reported in the causes of death for stillbirth was 'unexplained' (32.1%), followed by 'all other causes' (22.7%), 'antepartum haemorrhage' (14.4%), 'other unspecified condition' (14%), 'placental conditions' (9.3%) and 'congenital anomalies' (8.4%)[22].

According to the latest NPEC Irish Perinatal Mortality report, in 2020, the most common cause of death in stillbirths was congenital anomalies (n=79 of 240, 32.9%), which included multiple anomalies, anomalies of the cardiovascular system, central nervous system, musculoskeletal and gastrointestinal systems, respiratory system and urinary tract[3]. The second most common cause of death for stillbirths in Ireland according to the 2020 data was specific placental conditions (n = 73, 30.4%), the most commonly occurring were placental malperfusion (26%) followed by fetal vascular malperfusion (19.2%). The third most common causes of death were specific mechanical issues. Antepartum or intrapartum haemorrhage accounted for 5.4% of stillbirths, and infection accounted for 4.2% of stillbirths[3]. The cause of death was unexplained for 10.8% of the stillborn babies; however, for more than half of these cases, post-mortem information was still not available[3]. The latest MBRRACE-UK report also reports findings on the cause of death of stillbirths[19]. In the UK in 2020, 33% of stillbirths were unexplained. Additionally, the authors report that 35% of stillbirths were attributable to placental causes[19].

1.5 Impact of stillbirth

Stillbirth is a tragic event that affects mothers, partners, families, siblings, wider family, healthcare professionals and the whole of society[24]. In this section, the impact of stillbirth on the different aspects of the parents' lives, as well as its influence over the healthcare professionals involved and the care system is discussed, as well as stillbirth's related economic cost at the individual level, and system level.

1.5.1 Psychological impact of stillbirth on bereaved parents

Previous research has explored the psychological impact of stillbirth on bereaved parents. In a state of acute grief, mothers and fathers report feelings of guilt and blame, regret, stigma, and even suicidal ideation[24]. Campbell-Jackson et al., (2014) conducted a systematic review of the literature to explore the impact of stillbirth after 20 weeks gestation, specifically on women, by exploring the quantitative and qualitative research. The authors concluded that stillbirth can be associated with different psychological symptoms such as anxiety, depression, distress and negative well-being, especially during the first few months post-stillbirth[25]. These findings are similar to those reported in another systematic review of the literature published in 2016 looking at the psychological impact of stillbirth on families[26]. The study findings reported that bereaved parents have an increased number of psychological and emotional disorders such as depression, general anxiety disorder, social phobia, agoraphobia, anger, negative cognitive appraisals such as sense of failure, long-term guilt, post-traumatic stress disorder and suicidal ideation[26]. Stillbirth may result in social isolation due to bereaved parents avoiding activities where they might have to confront families with babies or explain their experience[27]. This might be explained by a phenomenon commonly named disfranchised grief[26]. Parents report in the literature that they do not feel allowed or supported to mourn the death of their baby due to stigma and societal taboo and it not being culturally acceptable[27]. This stigma experienced by bereaved parents can lead them to feel more isolated, blamed, silenced and not recognised as parents[26,27]. Nuzum et al., (2018) concluded in their qualitative study looking at the impact of stillbirth on bereaved parents, that good communication and empathetic care have a great impact on improving the experience of bereaved parents and their grieving process[28].

While the majority of research has focused on women's experiences of stillbirth given the impact that having a stillborn baby has in their physical and mental health[25], and the importance of prioritizing their needs, some research has also focused on the experience of fathers after stillbirth[25]. Jones et al., (2019) conducted a scoping review to report on research on men's experiences of grief and loss following stillbirth and

neonatal death. The authors concluded that men tend to feel like they need to adopt a supportive role for their partner, sometimes to the detriment of their own mental health and well-being[29]. The loss of identity as a father and the lack of social recognition of fatherhood in the case of stillbirth can also lead men to feel disfranchised grief. Although this review's findings seem to show that the feelings associated with the loss are similar in men and women, their behavioural externalisation differs, probably because of social-cultural norms and beliefs about communicating and expressing emotions[29]. The authors highlight the importance of healthcare professionals acknowledging the parental loss and treating fathers as more than "support" for the woman, and that they not be assigned the task of informing family and friends of the death of their baby[29].

1.5.2 Impact of stillbirth on couples, siblings, and future pregnancies

Having a stillborn baby has a great impact on the relationship between parents. Divorce and relationship difficulties are frequently reported among bereaved parents[26]. As stated above, women and men might experience and express grief in different ways. These discrepancies in the grieving process amongst women and their partners might cause disputes, infidelity, difficulties in sexual interactions, and a lack of understanding of each other's experience[26]. However, some studies have also reported that experiencing such a tragedy together helped some couples get closer and feel an increase in their emotional bonding and intimacy, as they were relying upon each other for support[30]. The evidence supports that the different grieving styles that partners might adopt can act as opportunities to grow closer together or a risk for the relationship, depending on the other person's reaction towards it. For some, establishing boundaries around their grieving process created conflict, whereas couples that understood and allowed the other person to mourn in their own way with tolerance and respect, tended to create a stronger bond[30].

Experiencing a stillbirth also has consequences on future pregnancies for a couple. Women with a previous stillbirth have a higher risk of pregnancy complications and stillbirth recurrence[31]. Hence having a previous stillbirth will have implications on the

woman's care during her subsequent pregnancy. Understanding the circumstances of the previous stillbirth will have an impact on the potential risk of recurrence and the couple's counselling regarding future pregnancies[32]. Meaney et al., (2016) conducted a study exploring bereaved parents' experiences of future pregnancies and found that parents start thinking about future pregnancies only a few days after the loss of their baby. The parents stated that their perception of risk was altered, even reaching a fatalistic approach since they had not anticipated the possibility of having a stillborn baby before. According to the author's findings, women were more driven to plan another pregnancy, and fathers were more reluctant. Both mothers and fathers had concerns about the emotional implications of a new pregnancy and the expected level of care during that subsequent pregnancy[33]. These findings are similar to those of Murphy et al. (2021), in their qualitative study. Couples in this study also reported struggling to make decisions about future pregnancies, and it was women who had a stronger desire to get pregnant soon after the stillbirth, whereas men were more cautious and protective of their grieving time[34].

Couples who have experienced a stillbirth before will have different expectations regarding their care in future pregnancies than couples who have not experienced loss[33]. Mills et al., (2013) conducted a meta-synthesis of the literature intending to improve the understanding of parents' experiences of maternity care in pregnancy after stillbirth or neonatal death[35]. The authors reported that parents tend to adopt a more assertive attitude towards healthcare professionals in an attempt to gain control over the situation in a pregnancy following stillbirth. Parents also tend to seek an alternative care provider to the one they had in their previous pregnancy which resulted in stillbirth and remain more vigilant and alert towards potential signs related to the baby's distress. Parents reported the need to encounter healthcare professionals that are aware of their history, and are empathetic and supportive, without showing dismissive attitudes towards their fears and concerns. Women sought extra reassurance from healthcare professionals by demanding additional monitoring and scans in their following pregnancy after stillbirth. Additionally, the study also showed that parents highly valued a targeted specialised support system

throughout their subsequent pregnancies[35]. These qualitative findings concur with those obtained by Roseingrave et al. (2022) who found in their Irish retrospective cohort study of pregnancies after stillbirth, that the average number of antenatal appointments was twice as high as expected, and the number of ultrasound scans was 5 times higher than expected[36].

In addition to the impact on the couple's relationship, stillbirth affects the whole family and wider social context. For instance, research on the impact of stillbirth on siblings has reported the negative implications of the death of a sibling during childhood on the individual's future mental health as an adult[3]. This impact is often associated with intense feelings of survival guilt, but also with the family's potentially difficult adaptative response to the stillbirth[26]. Some parents have reported difficulties coping with their own grief and parenting their surviving children, whereas others found comfort in having existing children[26]. Other authors have studied the potential psychological effects of stillbirth on siblings born after a stillbirth had occurred. Turton et al., (2009) concluded from their study that children born after a stillborn baby and assessed at 6-8 years old did not present increased psychosocial vulnerabilities compared to their peers[37]. However, they were perceived by their mothers as having more overall difficulties and peer problems. The authors report that mothers who experienced a stillbirth perceive and act differently towards their next child, compared to non-bereaved mothers[37]. They hypothesised that these differences could be associated with the fact that mothers compare their living children's behaviour with the ideal imagined behaviour of the lost child, or that mothers perceive their children as fragile or vulnerable, and hence their need to be hypervigilant[26]. However, there is no evidence of the clinical significance of the effect of these differences on the children, so further research in this regard is necessary to examine potential long-term sequalae[37].

1.5.3 Impact of stillbirth on healthcare staff

Consultant obstetricians have previously reported that stillbirth is one of the most complicated experiences they have to endure during their professional practice[35]. Nuzum et al (2014), conducted a study exploring consultant obstetricians' experiences of stillbirth. The consultants interviewed expressed that caring for bereaved parents affected them personally and professionally. They spoke about the importance of providing human, sensible care, and about the professional responsibility they felt as main carers, but also related to the potential medico-legal concerns in stillbirth scenarios[35]. This study highlights the importance of providing healthcare professionals with professional and personal support structures, and that self-care is included in medical training and further professional development[35]. It also highlights the need to improve training of healthcare professionals in perinatal bereavement care to facilitate continuity of care for parents, as well as clear care pathways[35,38].

Intrapartum stillbirth experience amongst obstetricians is another area that has been explored previously in the literature. Similarly, to the findings discussed above, being exposed to an intrapartum stillbirth also has a huge emotional impact on obstetricians. Obstetricians and midwives participating in a study conducted by McNamara et al (2018), found the experience of intrapartum stillbirth to be upsetting and stressful, with sadness, shock and guilt being commonly reported emotions[39]. McNamara et al reported that obstetricians are also hugely concerned about the media attention related to intrapartum death, as these were commonly sensationalised and inaccurately reported[39]. Obstetricians also felt unsupported and isolated after caring for a woman who experienced an intrapartum death and highlighted the importance of providing support to each other and breaking the "blame culture"[39].

1.5.4 Economic burden of stillbirth for families and the healthcare services

Stillbirth also has a financial impact on the health services[40]. Gold et al., (2012) concluded in their study that stillbirths with maternal complications had higher

hospital costs and longer hospital stays in the US[41]. The cost increase of stillbirth is also associated with expenses related to investigating the cause of death, and the increase in the cost of a subsequent pregnancy[40]. These findings are similar to those reported by Callander et al., (2020) who concluded that after accounting for gestation at birth, the cost of stillbirth was 42% more than a livebirth[42]. In many cases, these additional costs are absorbed by governments or insurance companies, or within the health services, but sometimes the economic burden falls on the bereaved parents[40,42]. Further, the cost of the care of women that have a pregnancy following a stillbirth is also increased because, as mentioned above, women seek further visits and scan for reassurance[42]. Additionally, women after stillbirth had higher rates of caesarean section, induction of labour and preterm delivery compared to the general multiparous population[36], which are interventions with increased costs compared to expectant management[43]. The evidence also showed that intervention decision-making was often influenced by the previous history of stillbirth[36]. Heazell et al (2016) estimated costs of £3499 after a stillbirth of a non-recurrent cause to £4057 for a stillbirth of an unknown cause. According to the authors, pregnancy after stillbirth costs between £558–1735 more than if the previous pregnancy would have resulted in an uncomplicated livebirth[40].

Bereaved parents are also affected economically by stillbirth in many other ways. Previous research has reported that some parents have to assume the cost extends to $1179–11 605 for burial plots and $1410–4605 for memorials[40], although often these costs are covered by governments, hospitals or health insurance[40]. Some parents reported difficulties returning to full-time employment which affected their financial situation long term and costs associated with mental health care[40]. All of these additional costs can intensify the negative psychological symptoms of the grieving parents, which in some instances are not able to use their preferred choices of burial or care due to financial restrictions[40].

1.6 Different challenges to stillbirth prevention in low-, middle- and high-income countries

The challenges associated with stillbirth prevention are different amongst low-, middle- and high-income countries. The vast majority of stillbirths occur in low- and middle-income countries, especially in rural sub-Saharan Africa[2]. Some of these challenges can be attributed to health-system indicators. According to Lawn et al. (2011), in low-income countries with stillbirth rates of at least 25 per 1000 total births, half of the births occurred without skilled care attendants and the median number of nurses and midwives available per 1000 people was 0.5[2]; this is much lower than the WHO recommendation (2.5 per 1000 people)[44]. In low-income countries, labour and birth are of higher risk. In many countries of sub-Saharan Africa, over 50% of stillbirths are intrapartum. These babies are often born at term and their deaths are associated with suboptimal care, hence, they are preventable[2,15]. However, in high-income countries, intrapartum stillbirth rates are normally less than 0.5 per 1,000 total births[2] which marks a significant difference as to where preventive efforts should be focused depending on the context.

These differences in the challenges associated with stillbirth prevention have led some authors to study context-specific risk factors (See table 1.1). Aminu et al., (2014) concluded in their systematic review that the most common risk factors associated with stillbirth in developing countries were maternal age, gestational age at birth, parity (over 5), lack of adequate antenatal care, fetal sex, birthweight, multiple gestation, and maternal morbidity[45]. Di Mario et al., (2007) identified in their systematic review of risk factors in developing countries, that maternal syphilis, chorioamnionitis, maternal malnutrition, lack of antenatal care, and maternal socioeconomic disadvantage were significantly associated with an increased risk of stillbirth[46]. Similarly, McClure et al., (2006) reported that the most common risk factors in developing countries are obstructive/prolonged labour and associated asphyxia, infection, birth injury, and low availability of caesarean section (CS), congenitally acquired infections such as syphilis and gram-negative bacterial infections, hypertensive disease, poor management of preeclampsia/eclampsia,

poor nutritional status, previous stillbirth, congenital anomalies, malaria and sickle cell disease[45]. However, the most commonly reported risk factors in developed countries are congenital anomalies, growth restriction/placental thrombosis, medical diseases such as diabetes, hypertensive disease/pre-eclampsia, congenitally-acquired infections such as group B streptococcus and parvovirus 19, obesity and multiple gestation[2,45]. Additionally, in middle- and high-income countries, there is increasing concern about smoking and obesity[47].

Table 1. 1: Most common risk factors for stillbirth between low/middle- income countries and high-income countries.

Categories		**Main risk factors for stillbirth**[2,45,46,48]	
		Low/middle- income countries[45,46,48]**.**	**High-income countries**[2,45]
Sociodemographic factors		Maternal age Socioeconomic disadvantage Low access to C-Sections	
Medical factors	Maternal medical factors	Maternal morbidity Viral and bacterial Infections (malaria, syphilis) Poor management of pre-eclampsia Sickle Disease Lack of access to adequate antenatal care	Diabetes Hypertension disease Congenitally acquired infections
	Obstetric history	Parity >5	
	Pregnancy complications and fetal factors	Gestational age at birth Fetal sex Birthweight Multiple gestations Obstructed labour – asphyxia Birth injury	Congenital anomalies Growth restriction Placental thrombosis Multiple gestations
Behavioural and lifestyle factors		Maternal malnutrition	Smoking Obesity

Providing almost universal access to antepartum and intrapartum care with a focus on risk reduction and treatment of complications as they arise, in high-income countries, is what most likely produced the sharp decrease in stillbirth rates 50-60 years ago[15]. This has facilitated the decline in rates of fetal mortality associated with obstructed labour, asphyxia, hypertension, diabetes, Rh disease, placental abruption, post-term pregnancies and infections[15]. Hence, preventive efforts should focus on

improving intrapartum care and increasing accessibility of care in low- and middle-income countries, whereas improvements in antenatal care and risk identification including behavioural risk factors such as smoking and weight-related risks should be the focus in high-income countries[2].

1.7 Risk factors for stillbirth

A risk factor can be defined as "something that increases the chance of developing a disease"[49] or adverse health outcome. Establishing which risk factors influence a certain outcome can play an important role in the prediction and prevention of such outcomes[50]. Risk factors can be marked or variable depending on their ability to change either spontaneously or by external manipulation. Risk factors and their strength to predict a certain outcome may also vary depending on the population or the time of exposure, for example, depending on the stage of development of an illness.

Risk factors can be measured in populations where the risk factors and their correlated outcomes appear at variable frequencies amongst its individuals[50]. The study of the correlations between risk factors and their associated outcomes provides researchers with context-specific data that can be used to develop models to identify people at risk of health conditions efficiently[50]. This section discusses the evidence in the literature regarding the risk factors for stillbirth. These can be classified into different categories: sociodemographic factors, medical factors, and behavioural and lifestyle factors (See table 1.2).

Table 1. 2: Main risk factors associated with stillbirth.

Categories		**Main risk factors for stillbirth**
Sociodemographic factors		Maternal Age Ethnic minorities Socioeconomic status
Medical factors	Maternal medical factors	Hypertension Diabetes Thyroid disorders Bacterial and viral infections
	Mental health conditions	
	Obstetric history	Parity Previous history of pregnancy loss Multiple pregnancies History of Caesarean Section Use of assisted reproductive technologies
	Pregnancy complications and fetal factors	Fetal growth restriction Placental insufficiencies Pregnancy length Changes in fetal movements
Behavioural and lifestyle factors		Smoking Alcohol and illicit drug use Use of medical drugs and supplements Maternal weight Sleeping habits Antenatal care attendance

1.7.1 Sociodemographic factors

Maternal age

Over the past decades, women have intentionally delayed childbirth for different socioeconomic reasons. Some authors point out that this is due to an incompatibility between the optimal biological period to have children and the wish to obtain additional education and build a career[51]. Worldwide, 6.7% of stillbirths are attributable to a maternal age of 35 years old[1]. According to the latest NPEC data,

39.1% of the women who experienced a stillbirth in 2020 in Ireland were 35 years old or older (28.3% were 35-39 years old and 10.8% were over 40 years old)[3]. The authors found that there was a significant association between perinatal mortality and age, with women aged greater than 40 having 1.8 times the rate of perinatal mortality in 2020 ($p = 0.002$)[3].

Lean, Derricott, Jones, & Heazell, (2017) undertook a systematic review and meta-analysis that showed similar results regarding age and risk of stillbirth. The results of this meta-analysis demonstrated that there is an increased risk for stillbirth in advanced maternal age (Odds ratio (OR) 1.75) with high heterogeneity within the data ($I^2 = 95.6\%$; classification severe), and this risk augments as women get older ($R^2 = 0.61$)[52]. The definitions used in the studies included in this review were different in terms of the gestational age cut-off for each individual study which meant that the meta-analysis showed significant heterogeneity between the studies[52]. Huang et al. (2008) conducted a systematic review to analyse the effects of maternal age on the risk of stillbirth, however, it is not specified what definition of stillbirth they used[53]. The authors found that there was a significant association between maternal age over 35 years and stillbirth risk in all of the 31 retrospective cohort studies (77%) and in all 15 population-based studies they included in their review, with a variation in the relative risk of stillbirth from 1.20 to 4.53 for older (35 years or more) versus younger women (20 to 24 years old) [53]. Moreover, the authors reported that in 57% of the studies reviewed, the researchers adjusted their results for potential cofounders and the risk ratios (RR) did not change, suggesting that the association between increased maternal age and stillbirth risk was not influenced by any other factors[53].

In concordance with the studies mentioned above, Balayla, Assayag, & Abenhaim (2011) found in their population-based cohort study on 37 million births that occurred after 24 weeks gestation in the United States, that the risk of stillbirth seems to be lowest between the ages 15 and 35, and increases beyond that interval[54]. These authors also adjusted the results for demographic and obstetric

characteristics and the values of the OR oscillated between 1.02 and 2.22, incrementing with the different age intervals[54]. Similarly, Saccone et al. (2022) concluded in their meta-analysis that women aged >40 years had a significantly higher risk of stillbirth (RR 2.16) which increased as age progressed (women >50 years old RR 5.42)[55].

Hoffman et al. (2007) also agreed with the previous results in their retrospective study of all singleton pregnancies delivered between 1989 and 2004. The researchers divided their population into three different age groups: less than 35 years old; 35 – 39 years old and; 40 or more years old[56]. They concluded that the likelihood of suffering from fetal death (defined as the death of a fetus at or beyond 20 weeks gestation) was higher in the group of women at age 40 or older (OR between 1.10 and 2.93)[56]. In terms of gestational weeks, the authors found that the older group (more than 40 years old) had a higher risk of suffering from fetal death at 28 – 31 weeks (OR 2.93) and 40 – 41 weeks gestation (OR 2.28) [56]. However, the younger group (35 to 39 years old) had higher risks at 32 to 36 weeks gestation (OR 2.03;) [56]. They also concluded that age is an independent risk factor for fetal death. Similar to these results, Kenny et al. (2013) found the relative risk of stillbirth (considered after 24 weeks gestation) was significantly elevated in women aged 30-34 (RR 1.23) and higher in women aged 35-39 (OR 1.41)[57]. Women aged 40 years or older were twice more likely to have a stillborn infant than women under 30 years old[57].

One of the hypotheses that some authors use to establish the relationship between maternal age and stillbirth relates to the link between maternal age and disorders that are related to uteroplacental dysfunction such as fetal growth restriction, pre-eclampsia and placental abruption[58]. Miremberg et al (2020) found support for this hypothesis in their prospective case-control study. The authors found that the placentas from the advanced maternal age group (35 years or older) showed higher rates of maternal vascular lesions (39.1% vs. 24.5%, p=0.003). Maternal age was also associated with increased placental maternal vascular lesions independently of all other demographics (aOR 1.18)[59].

Ethnic minorities and immigrant women

It has been reported in the literature that there is an excess of fetal and infant mortality in ethnic minorities in different countries of Europe and the United States[60,61]. Fredsted Villadsen et al., (2009) conducted a study based on population-covering national registries to identify all live and stillbirths (defined as death after 28 weeks of gestation) of women from the five largest ethnicity groups in Denmark (Danish, Turkish, Lebanese, Pakistani, Former Yugoslavian, which includes Yugoslavia, Bosnia–Herzegovina, Serbia–Montenegro, Croatia, Macedonia and Slovenia, and Somali). They found that the relative risk for stillbirth among these ethnic minorities was higher than the risk in the Danish population (Relative risk (RR) between 1.04 and 2.11), especially in the Turkish, Pakistani and Somali mothers (RR 1.28, 1.62 and 2.11 respectively)[61]. The authors also adjusted the results depending on household income (RR from 0.83 to 1.72) and maternal education (RR from 0.91 to 1.85), and could not find interactions between ethnicity and those characteristics[61]. These findings are like those obtained by Matthews et al. (2022) in the UK. There, authors concluded that the rates of stillbirth at 24 weeks gestation or over were higher for Black African babies (3.83 per 1000 births), Black Caribbean babies (3.60 per 1000 births) and Pakistani babies (2.99 per 1000 births) compared to White ethnicities[62]. When it comes to the primary cause of death, the authors found that higher rates of congenital anomalies were present in Pakistani, Bangladeshi and black Africa ethnicities, whereas more placental causes were found in black ethnicities[62]. Similarly, the latest MBRRACE perinatal mortality report (2020) concluded that stillbirth rates are over twice as high for Black and Black British babies and around 60% higher for Asian and Asian British babies compared with babies of white ethnicity[63]. In Ireland, according to the latest NPEC perinatal mortality report, Irish Travellers, Asian and Black ethnicities were overrepresented in the total of mothers who experienced a perinatal death in 2020[3].

Further, it has been described that women from ethnic minorities are more likely to refuse caesarean sections due to fear, and also some women reduce their food intake to keep the fetus small to avoid an obstructed delivery. These practices arise from

the beliefs that caesarean section can put the woman's and baby's life at risk[64]. Another study conducted in Germany reported an insufficient use of antenatal services among migrant women (women from different regions of origin compared to German women); causal factors identified included difficulties in communication due to language barriers and lack of knowledge about pregnancy-related health issues[65].

Other researchers in the Netherlands compared the rates of stillbirth (defined as fetal death after 24 weeks gestation) and early neonatal mortality among mothers from minority ethnicities with those of Dutch mothers[60]. In this study, the Western ethnic groups included ethnic Dutch and other Western women with Caucasoid ancestry (Belgium, UK, Germany, etc) and the non-Western ethnic group was classified into five subgroups: the African or Creole, originating from Surinam (a former Dutch colony); the South Asian or Hindustani originating from Africa and India, respectively; Turkish/Moroccan women; East-Asian women originating mainly from Indonesia (a former Dutch colony) and other non-Western women. The authors found that teenage pregnancies, lower socio-economic status and late care-seeking were more prevalent among the minority ethnic groups[60]. The adjusted OR of the different ethnic groups regarding fetal mortality oscillated from 0.8 up to 1.8, being higher in the south Asian, African and Turkish/Moroccan groups (OR 1.8, 1.7 and 1.3 respectively)[60]. Hence, the risk associated with being part of an ethnic minority is a combination of other circumstances associated with social deprivation or economic disadvantages[66] rather than inherent differences amongst ethnicities in isolation.

Socioeconomic status

A number of studies have described an increase in the risk of stillbirth among women with lower socioeconomic status or women with lower education levels. A study conducted by Heazell et al. (2020) in the UK demonstrated that women living in the most deprived areas had a statistically significant increase in their risk of stillbirth (aOR 3.16) at 28 weeks gestation or more[62]. Further, unemployed women also had an increased risk (aOR 2.32). These findings are similar to those obtained in the

systematic review and meta-analysis conducted by Thomson et al. (2021) exploring the associations between socioeconomic factors and pregnancy outcomes in the UK and the Republic of Ireland. Thomson et al. found that lower levels of occupation/social class (OR 1.61) and unemployment in mothers (OR 1.24) and fathers (OR 2.26) were associated with a significantly increased risk of stillbirth[67].

Similarly, Stephansson et al. (2001) conducted a population-based case-control study of stillbirths in Sweden which concluded that when adjusting the OR for age, the risk of stillbirth after 28 weeks gestation was highest for blue-collar, unskilled workers (OR 2.6), and skilled blue-collar workers (OR 2.6) whereas white-collar, higher educated workers had an OR of 1.0[68]. In this study, the authors also adjusted the results for age, country of birth, maternal body mass, height, and smoking, and found that blue-collar workers, either unskilled or skilled, still had the highest OR (2.2 and OR 2.4 respectively). The researchers, however, concluded that blue-collar workers in this population had a higher tendency to engage in smoking or were more likely to have a higher BMI, which could be contributing to these differences among the groups[68]. The authors also speculated that the association between low socio-economic status and stillbirth can be due to a reduced level of education and access to information, differences in quality and quantity of medical care, increased emotional problems such as stress or depression and lowered self-esteem, as well as cognitive problems[68].

Besides employment, other authors have studied the level of education of women in association with the risk of stillbirth. A study conducted in Canada collected all the data of stillbirth cases recorded in Quebec, however, only intrauterine deaths of fetuses weighing 500 g or more are registered in that region. The results showed that the mean number of years in education was higher in the livebirths group of mothers than in the stillbirth one (13.1 years vs. 12.5). In addition, the authors concluded that women with the lowest education level were twice as likely to have a stillbirth than women with higher education levels (OR 1.98)[69].

1.7.2 Medical Factors

Maternal medical conditions

Some maternal diseases such as diabetes, hypertension, antiphospholipid syndrome, Systemic Lupus Erythematosus, chronic renal disease, thyroid disorders, cholestasis of pregnancy and heritable thrombophilia have been associated with a higher risk of stillbirth[70,71]. Below the most common co-morbidities with the strongest evidence are discussed in this section, although this list is not exhaustive.

Hypertensive disorders contribute to 9.2% of stillbirths[72], these disorders include chronic hypertension, pregnancy-induced hypertension and eclampsia. Xiong et al., (2018) undertook a study including almost 7 million pregnancies to establish the risk of stillbirth, at 28 weeks or later, associated with each type of hypertensive disorder in China. The authors concluded that the rate of stillbirth in singleton pregnancies was higher in women with hypertensive disorders, regardless of other risk factors (aRR 3.1) with a stillbirth rate of 21.9/1000 births[73]. Specifically for each hypertensive disorder, Xiong et al., (2018) reported the following results: chronic hypertension aRR 2.32; superimposed preeclampsia aRR 6.66; preeclampsia or eclampsia aRR 4.15; and, gestational hypertension aRR 1.21[73]. These results agree with those obtained by Allen and colleagues (2004) who concluded that women with any type of hypertension in pregnancy were 1.4 times more likely to have a stillborn baby (defined as fetal death at 20 weeks or ≥500 g.) than normotensive women. In cases of women with pre-existing hypertension, the risk was also increased (RR 3.2)[74].

Diabetes and gestational diabetes are medical conditions that have been widely studied concerning the risk of stillbirth. Ye et al. (2022) conducted a systematic review looking at the influence of gestational diabetes over different adverse pregnancy outcomes, the review did not find clear evidence for differences in the odds of stillbirth when comparing women with gestational diabetes versus women with no gestational diabetes (OR 1.08)[75]. The authors of this review acknowledged that the use of different definitions for stillbirth in the original studies might have caused potential bias in the analysis[75]. Similar findings were obtained by Lemieux et

al. (2022) in their systematic review exploring the association between gestational diabetes and stillbirth. Results did not show a significant association between gestational diabetes on its own and stillbirth, although their findings were also substantially heterogeneous[75]. A stratified analysis was conducted based on stillbirth definition by gestational age and found that when only including studies that used a 28-week gestational threshold to define stillbirth, gestational diabetes was associated with an increased risk of stillbirth (OR 1.27)[75]. The authors hypothesize that medical comorbidities such as obesity and advanced maternal age, which are independent risk factors for stillbirth and are associated with the development of gestational diabetes, might be biasing the association between gestational diabetes and stillbirth[75]. Regarding the differences found based on the stillbirth definition, the authors hypothesise that this might be associated with the fact that pregnancies are screened for gestational diabetes around 24-28 weeks gestation, and hence stillbirth occurring during that time cannot be attributed to gestational diabetes because this type of diabetes is commonly diagnosed after the 24th week of gestation[75]. Malaza et al. (2022) conducted a systematic review to compare the different pregnancy outcomes of women with pre-gestational diabetes (PGDM) and gestational diabetes. This review concluded that stillbirths are more common in women with type 1 diabetes mellitus and type 2 diabetes mellitus compared with women who develop gestational diabetes during pregnancy[75]. It seems that stillbirth might be associated with inappropriate glycemic control and placental insufficiency due to poor maternal vascular disease in PRGM.

Bacterial and viral infections can also have a huge impact on the risk of stillbirth[76]. Recent research has been looking at the influence of COVID-19 infection on pregnancy outcomes. Linehan et al. described a case study of third-trimester pregnancy complicated by COVID-19 named SARS-CoV-2 placentitis[77]. Allotey et al. (2020) conducted a systematic review to determine the clinical manifestations, risk factors and maternal and pregnancy outcomes of women with suspected or confirmed coronavirus disease. The authors conclude that the odds of stillbirth were higher for babies born to women with Covid-19 compared to babies born to women

with no Covid-19 infection (OR 1.81)[78]. These findings are similar to the ones obtained by Qin Wei et al, (2021) which concluded that people who had a SARS-CoV-2 infection in pregnancy were more likely to experience stillbirth compare to those who had not (OR 2.11)[79]. Schwartz et al. (2022) described that the lesions occurring during the course of a SARS-CoV-2 placentitis[77] hinder the placenta's ability to effectively perform its function of oxygenating the fetus, potentially leading to stillbirth and neonatal death via malperfusion and placenta insufficiency[80,81].

Mental health conditions

Mental health issues have also been associated with poor obstetric outcomes, including stillbirth[82]. Adane et al. (2021) conducted a systematic review and meta-analysis to explore the potential association between maternal prenatal mental health disorders and the risk of stillbirth and/or infant mortality[83]. The authors found that the odds of experiencing stillbirth or infant mortality for women with any maternal prenatal mental health disorder was increased (OR 1.59) compared with women without prenatal mental health disorders[83]. Women with depression had a significant increase in the risk of stillbirth (OR 1.51)[83]. Further, women with severe mental illness also had an increased risk (OR, 1.42)[83].

Wisbor et al (2008), conducted a prospective study to analyse the effect of psychological stress on the risk of stillbirth, defined as the delivery of a dead fetus at 28 weeks' gestation or later. The researchers concluded that women with high levels of stress accounted for a 4.9% of the total stillbirths and had an 80% increased risk of stillbirth (aOR 1.8)[84]. King-Hele et al. (2009) undertook a cohort study on 1.45 million live births and 7020 stillbirths which occurred at 28 weeks gestation or later, during 1973 – 1998 from the Danish national registers. The aim of the study was to determine whether the risk of stillbirth was higher when the babies were exposed to a history of any maternal psychiatric admission before birth[85]. The study also aimed to examine the relationship between the different causes of death of stillborn babies (antenatal complications, complications of delivery, congenital anomalies of fetus, all other causes of stillbirth), and different psychiatric disorders categories

(schizophrenia and related disorders, affective disorders, alcohol/drug-related disorders)[85]. The results of this study showed that the risks of stillbirth were raised for all of the causes of death of the stillborn baby that the authors analysed and for all of the maternal psychiatric diagnostics categories[85]. The risk in the category of history of alcohol/drug-related disorders was increased for stillbirth due to complications at delivery (RR 2.3), whereas in the affective disorders, the risk was higher for stillbirth due to fetal congenital anomalies (RR 2.4)[85]. The study did not find that the risk was higher in schizophrenia and related disorders in comparison with the other two disorder categories[85].

Although the reasons for the association between higher adverse perinatal outcomes and perinatal mental health is not fully understood, the fact that the risk was higher in all disorder categories can mean that these risks are associated with the psychiatric population itself, rather than with any specific diagnosis[85]. This can be because this population is more likely to engage in inappropriate antenatal habits like smoking or drinking and limit their attendance at antenatal care[85,86]. Hence, these associations are probably a complex array of direct illness effects, comorbidities and behavioural and lifestyle factors as well as an increased risk of pregnancy complications[83].

1.7.3 Obstetric history

Some factors that have been studied for their relationship to the risk of stillbirth relate to the obstetric/reproductive history of the woman. These risk factors include a history of previous pregnancy loss, either miscarriage or stillbirth, primiparity, multiple pregnancies, caesarean section in previous deliveries, interval between pregnancies and use of assisted reproductive technology[87].

Parity

Parity has been associated with the risk of stillbirth. Gardosi et al. (2013), conducted a population-based study to research different maternal risk factors, including parity, for stillbirth. The study included 92,218 singleton livebirths and 389 stillbirths from 24 weeks gestation onwards in their study. The results showed that nulliparous women had a higher risk of stillbirth than women who had had livebirths before (aOR

1.8). Further, the authors found that there was also a 60% increase in the risk of stillbirth for women who had given birth 3 times or more (aOR 1.6)[88]. Bai et al. (2002) also found this U-shaped relationship between parity and obstetric complications; the study concluded that the risk was higher for nulliparous women (OR 1.75) and women with high parity (≥ 4 previous pregnancies up to at least 20 weeks) (OR between 1.19 and 1.35)[89]. However, the risk of perinatal death for nulliparous women and women who had had 2 or 3 previous pregnancies was small (OR 1.17; OR 1.08 and 1.18 respectively), but for women who had 4 or more previous pregnancies, the OR was always above 1.5[89]. Aliyu et al. (2005), undertook a study selecting singleton live births and fetal deaths of 20 weeks or more to analyse the effect of high multiparity on the risk of stillbirth; the authors excluded nulliparous women for this study and focused on women of high multiparty (5 pregnancies or more). The results show a dose-response correlation, where the risk of stillbirth was consistently higher with increased parity (5-9 previous live births aOR 1.04; 10 – 14 previous livebirths aOR 1.96; ≥ 15 previous livebirths, aOR 2.23)[90]. The latest NPEC report on perinatal mortality also found that in Ireland, the number of women with at least three previous pregnancies was over-represented in the sample that experienced a perinatal loss in 2020 (n = 41, 11.5%)[3].

Previous history of pregnancy loss

Lamont et al. (2015), conducted a systematic review and meta-analysis including cohort and case-control studies to examine the association between stillbirth (defined as occurring at 20 weeks or more or a birthweight of ≥400 g) in a first pregnancy and the risk of recurrent stillbirth in subsequent pregnancies. The authors concluded that the odds of stillbirth for women who had experienced a previous stillbirth were five times higher than in women without such history (OR ranging from 1.00 to 23.75)[92]. Heinonen and Kirkinen (2000) conducted a cross-sectional study comparing the pregnancy outcomes of women with a history of stillbirth after 24 weeks gestation or fetal weight of ≥500 g compared with women with no history of pregnancy loss. The authors concluded that women with a previous stillbirth were more likely to have a placental abruption (5.4% vs. 0.7%, $p < 0.001$), delivery before

37 weeks (RR 2.25), to have a low birthweight infant (RR 2.70) and other obstetric outcomes that have been established as risk factors for stillbirth. Subsequent stillbirths were also more likely in the exposed group (1.4% vs. 0.5%), but the difference was not statistically significant (aOR 1.2)[93].

Quenby et al. (2021), concluded from their review that having a history of recurrent miscarriage (3 or more miscarriages) also increases the risk of stillbirth in future pregnancies[94]. The authors found that the adjusted OR for the risk of stillbirth was 1.00 after one miscarriage, 1.04 after two miscarriages, and 1.69 after three or more miscarriages[94]. These findings are in concordance with the latest NPEC perinatal mortality report, which concludes that experiencing three or more miscarriages was the second most common previous pregnancy problem identified in women who experienced a perinatal death in Ireland in 2020 (n=21, 8.3%)[3].

Multiple pregnancies

Multiple pregnancies have also been studied in relation to the risk of stillbirth, especially after the increase of the use of assisted reproductive technologies[95]. Cheong-See et al. (2016) conducted a systematic review to determine the risks of stillbirth by gestational age in uncomplicated dichorionic (DC) and monochorionic (MC) twin pregnancies. The authors concluded that for DC twin pregnancies, the risks of stillbirth starts increasing significantly after 37+0-6 weeks gestation (RR 3.4 for 37+0-6 weeks, RR 10.6 for 38+0-6 weeks and RR 9.3 at 39+0-6) [96]. In MC twin pregnancies, the risk of stillbirth increases significantly slightly earlier, at 36+0-6 weeks gestation (RR 4.5 at 36+0-6 weeks, RR 9.6 for 37+0-6 weeks and 7.6 at 39+0-6)[96]. Danon et al. (2013) conducted a systematic review to compare the risk of stillbirth in uncomplicated monochorionic-diamniotic (MCDA) twin pregnancies versus uncomplicated DC twin pregnancies[97]. The authors concluded that uncomplicated MCDA twin pregnancies are at increased risk of stillbirth throughout the third trimester compared to DC twin pregnancies (OR 3.9 at 37-31 weeks, OR 4.2 at 32-33 weeks, OR 3.7 at 34-35 weeks and OR 8.5 at 36-37 weeks) [97]. A recent meta-analysis studied the prognosis of the surviving co-twin following the spontaneous

intrauterine death of one of the fetuses[98]. The review concluded that for MC twins, the rate of co-twin intrauterine fetal death was significantly increased when the single twin death occurred before 28 weeks of gestation (OR 2.31) compared with the single intrauterine death occurring after 28 weeks gestation[98].

The latest perinatal mortality data in Ireland is also in concordance with these findings. The 2020 report states that multiple births are overrepresented amongst the perinatal death population (8.1% of all perinatal deaths, 6.2% of all stillbirths in 2020), which is double the proportion of multiple pregnancies in all births in 2020[3]. In the UK, it was decided to conduct a confidential enquiry into the stillbirths and neonatal deaths in twin pregnancies that occurred in 2017. The report concludes that in over half of the pregnancies improvements in care were identified that may have made a difference to the outcome of the baby. The confidential enquiry also identified major sub-optimal antenatal care in half of the pregnancies and major sub-optimal follow-up care for three-quarters of the woman[99].

History of Caesarean Section

Another obstetric outcome associated with stillbirth is the use of caesarean section in previous pregnancies. Keag et al. (2018) conducted a systematic review to explore the long-term risks and benefits of caesarean delivery for mother, baby and subsequent pregnancies. The authors concluded that women with previous caesarean section delivery had an increased risk of stillbirth (as defined by the original paper's authors) compared with women with previous vaginal deliveries (OR 1.27)[100]. Similar findings were obtained by O'Neill et al. (2013) who conducted a systematic review and meta-analysis to examine the association between previous caesarean section and the risk of subsequent stillbirth or miscarriage (the authors did not follow any strict definition for their review)[101]. The results showed that women with previous caesarean section delivery have 23% (OR 1.23) higher odds of suffering from stillbirth in their subsequent pregnancy compared with women with previous vaginal delivery[101]. Moraitis et al (2015), conducted a population-based retrospective cohort study and meta-analysis to assess whether caesarean section as the mode of

delivery in the first pregnancy was associated with unexplained intrapartum stillbirth in a second pregnancy[102]. This study also concludes that caesarean delivery at all gestational ages in the first pregnancy is an independent risk factor for stillbirth in a subsequent one (HR 1.47). According to the 2020 perinatal mortality data in Ireland, caesarean delivery was the most prevalent previous pregnancy-related problem amongst women who experienced a perinatal loss (23%, n = 58)[102].

Use of assisted reproductive technology (ART)

Some authors have also studied the association between the use of assisted reproductive technology (ART) and the risk of stillbirth. Sarmon et al., (2021) have recently published a systematic review and meta-analysis of ten studies to identify the risk of stillbirth from in vitro types of assisted reproductive technologies compared with spontaneous conception. The findings concluded that singleton pregnancies following IVF/IVF-ICSI were associated with an increased risk of stillbirth compared with pregnancies following spontaneous conception (OR 1.82)[77]. These findings are similar to those obtained by Pandey et al. (2012) who concluded from their meta-analysis that singleton pregnancies after IVF are associated with higher risks of obstetric and perinatal complications.

Some authors have hypothesised that the association between ART and the risk of stillbirth could be also due to the increased likelihood of multiple pregnancies when using these technologies[103]. Henningsen et al. (2014) examined a Nordic population-based cohort including 62,845 ART singletons and 29,793 twins. The study showed that ART singletons had a higher risk of stillbirth during gestational weeks 22 to 27 compared with spontaneous conception (aRR 2.08), whereas there was no significant difference beyond 28 weeks gestation (aRR between 0.79 and 1.28)[104]. The authors also concluded that the risk of stillbirth for ART twins was smaller than for spontaneously conceived twins, and they attributed this difference to the fact that the ART twins were less likely to be monozygotic[104]. Dhont et al. (1999) undertook a case-control study in Belgium on the same topic. The results of their study showed that the incidence of perinatal mortality among ART pregnancies was higher than in

spontaneously conceived pregnancies (1.8% vs. 0.8%)[105]. To study this hypothesis further, Geisler et al., (2014) conducted a study to assess the obstetric and perinatal outcomes of twin pregnancies depending on the mode of conception. Results concluded that there were no differences in perinatal mortality (intrauterine death, stillbirth and neonatal death) when comparing twins conceived naturally and twins conceived by ART (p = 0.182)[106]. It is possible that availing of good quality antenatal care and specialised clinics for fertility treatment and twin pregnancies in high-income countries that provide special considerations to these types of pregnancies might be contributing to risk reduction. The Irish Clinical Practice guideline for the management of twin pregnancy suggests that the *"care of multiple pregnancies should comprise a hospital-based model, to include frequent maternal and fetal surveillance and formulation of an individualised delivery plan*"[107], but does not make distinctions between twins conceived naturally and twins conceived as a result of the utilisation of assisted reproductive technologies[107].

Interval between pregnancies

The interval between pregnancies has also been studied regarding potential negative outcomes, including stillbirth[108]. Hegelund et al. (2018) conducted a register-based study in Denmark which aimed to investigate the relationship between inter-pregnancy intervals and the risk of adverse pregnancy outcomes. The results showed a relatively weak association between inter-pregnancy intervals and stillbirth, the authors found that inter-pregnancy intervals of 60 months or more had 1.7% increase risk of stillbirth after 22 weeks of gestation (Risk difference 1.7, 95%CI 0.4, 3.0) than women with inter-pregnancy intervals of 18 to 23 months (reference group), although this findings was not statistically significant[109]. Cofer et al. (2016) also conducted a study on the same topic in California. The results showed that women with short inter-pregnancy intervals had a higher prevalence of stillbirth compared with larger inter-pregnancy periods (0.34% at <6 months, 0.25% at 6 – 17 months, 0.29% at 18 to <51 months). However, the differences were not statistically significant when comparing shorter inter-pregnancy intervals with the

recommended one as per the study authors (<6 vs. 18 to <51 OR 1.20; 6 – 17 months vs. 18 to < 51 months OR 0.87)[110].

The inter-pregnancy interval has also been studied in the cases of previous pregnancy loss. Kangatharan, Labram, and Bhattacharya (2017) conducted a systematic review and meta-analysis to assess the effect of inter-pregnancy interval following a miscarriage over the following pregnancy outcomes. The authors found that the risk of stillbirth for women after miscarriage was not statistically different between the group of the inter-pregnancy interval of fewer than 6 months compared with that of more than 6 months (RR 0.88)[111]. Similarly, Regan et al., (2019) published an international cohort study exploring the association between interpregnancy interval after stillbirth and birth outcomes in the following pregnancy. The results showed that an interval between pregnancies shorter than 12 months was not associated with an increased odds of subsequent stillbirth (defined as fetal death at 22 weeks gestation or more) compared to a longer interpregnancy interval of 24-59 months[112].

1.7.4 Placental and fetal factors

There are some factors related to the pregnancy itself or the development of the fetus that have also been associated with a higher risk of stillbirth.

Placental causes

Placental conditions are amongst the main causes of stillbirth in high-income countries[22]. Ptacek et al. (2014) undertook a systematic review of the different placental pathologies related to stillbirth. The authors included 14 case-control studies, 26 cohort studies and one population survey in their review, and no specific definition of stillbirth was selected due to the variations used internationally. The study findings concluded that the proportion of stillbirths associated with a placental cause of death varied from 11.2% to 64.9%. According to the authors, these differences might be attributable to the variation in the definitions used in different studies and the different classification systems for "placental causes"[113]. Some of the studies included in the review identified abruption (5.3-12.9%) followed by placental insufficiency (2.6-8.1%) and placenta praevia (1.7%) as the most frequent conditions

associated with stillbirth. However, the authors concluded that even though their results suggested that placental abnormalities or dysfunction are associated with a large proportion of stillbirths, the precise role of those lesions is still unclear[113]. As advanced maternal age and smoking have been established as risk factors for fetal growth restriction and placental dysfunction, this could provide explanation for the association between those factors and the risk of stillbirth[58,114].

Placenta praevia is a condition in which the placenta grows in the lowest part of the uterus and covers all or part of the opening to the cervix[115] and occurs in about 0.3-0.5% of the pregnancies[116]. Crane and Van Den Hof (1999) undertook a population-based, retrospective cohort study to determine the neonatal outcomes of placenta praevia. The results show that perinatal death (including stillbirth and neonatal death) was present in 2.30% of the cases with placenta praevia vs. 0.78% in the cases without it. When adjusting the crude odds ratio for maternal age, gestational age and congenital anomalies, this risk of perinatal death associated with placenta praevia were not significantly increased[117]. Following these results, Yeniel et al. (2012) in their retrospective cohort study, did not find a significant association between the presence of placenta praevia and the risk of stillbirth defined as fetal death after 20 weeks of gestation (RR 2.13, 95%CI 0.84 – 5.06, p = 0.073) or fetal growth restriction (RR 0.59, 95%CI 0.28 – 1.21, p = 0.168), so the authors concluded that placenta praevia is not an independent risk factor for stillbirth[116].

Pre-eclampsia is a medical complication that is associated with induced hypertension during pregnancy. This complication has also been associated with a higher risk of stillbirth[118]. In a meta-analysis published by Flenady et al. (2011), three studies reported on pre-eclampsia. The authors concluded that there was a 60% increased risk of stillbirth in cases of pre-eclampsia (aOR 1.6)[7]. Similarly, Chappell et al. (2021) concluded that pre-eclampsia was significantly more prevalent amongst mothers of stillborn babies compared to mothers of liveborn babies (Pooled unadjusted RR 2.4, 95%CI 1.7 - 3.4)[118].

Placental abruption has been correlated with stillbirth extensively as stillbirths occur in most cases where more than half of the placenta separates from the uterus prematurely[119]. Approximately 0.4-1% of pregnancies are complicated by this condition[119]. Ananth et al. (1999) undertook a hospital-based retrospective cohort study to assess the association between placental abruption and stillbirth at 20 weeks gestation or more. The authors found a prevalence of this condition in 1% of participants and concluded that placental abruption has a huge impact on the risk of antepartum stillbirth (aRR 8.9) and intrapartum stillbirth (aRR 9.6)[120].

The latest perinatal mortality data shows that, in Ireland, specific placental conditions were the second most common cause of death in stillbirths in 2020 (n= 73 of 240, 30.4%), with vascular malperfusion being the most common placental condition (n=19 of 73, 26.0%), followed by fetal vascular malperfusion (n=14 of 73, 19.2%)[3]. Similarly, in the UK, over two-thirds of stillbirths in 2020 were associated with placental causes (35%)[99].

Fetal growth restriction

Fetal growth restriction and small-for-gestational-age are two fetal characteristics that have been strongly associated with fetal death. However, these two terms cannot be considered synonyms as being small for gestational age is not always pathological[121]. A study conducted by Frøen et al. (2004) on 291 cases of unexplained singleton antepartum stillbirth (defined as fetal death at age ≥22 weeks gestation or birthweight of ≥500 g) found that 52% of the cases were growth restricted. The authors also concluded that suboptimal growth was the most relevant factor influencing intrauterine unexplained death (OR 7.0)[122]. Gardosi et al. (2013) undertook a cohort study to determine the different risk factors associated with stillbirth after 24 weeks' gestation. The researchers concluded that the presence of fetal growth restriction (birthweight below 10th customised birthweight centiles) contributed the highest risk (aRR 7.8)[88]. A population-based study conducted in Sydney by Gordon et al. (2015) also agreed with this result. The study showed that stillbirths (defined as death in utero at ≥22 weeks gestation or ≥500 g) were more

likely in those women who had been followed up for suspected fetal growth restriction than in those who had not (aOR 5.5)[123].

These findings are also supported by the results published by the Irish National Perinatal Mortality Clinical Audit. According to this report, a diagnosis of fetal growth restriction (FGR) was reported in 18% (n = 42) of stillbirths. 25.3% (n = 21 of 83) of severely small for gestational age and 21.3% (n = 23 of 108) of small for gestational age babies were diagnosed with fetal growth restriction antenatally. Given these findings, the authors call for a *"Standardised approach to improved antenatal detection of fetal growth restriction (FGR) with timely delivery is a potential preventative strategy to reduce perinatal mortality"* [3].

Changes in fetal movements

Fetal movements are a commonly used measure to evaluate fetal well-being and they are perceived by most women at about 20 weeks of gestation[124]. The maternal perception of reduced fetal movements (RFM) has been associated with stillbirth[87,122,125]. Stacey et al. (2011) undertook a study to explore the relationship between late stillbirth (28 weeks gestation or later) and the perception of RFM by mothers. The authors concluded that women who had perceived a decrease in the strength of the fetal movements were more likely to have suffered from stillbirth than those who had not (aOR: 2.37)[126]. McCarthy et al. (2016) reviewed all women presenting with RFM over 28 weeks of pregnancy to a tertiary-level maternity hospital emergency room. The authors observed that the stillbirth rate of women presenting with RFM at 28 weeks gestation or over was 14.6 per 1000 births, which was significantly higher than the corrected stillbirth rate of the same year (2.9 per 1000 births in 2019)[127].

Koshida et al. (2017) undertook a population-based study in Japan to investigate the maternal reaction time after perception of the RFM of women who suffered a stillbirth after 22 weeks gestation. Results showed that more than one-third of the mothers with stillbirth perceived RFM but only 11% (n = 7 of 64) of them sought assistance within 24 hours; 23% (n = 15 of 64) sought care between 24 -48hrs after

perceiving RFM, 17% (n = 11 of 64) sought care 48-72hrs after perceiving RFM and 8% (n = 5 of 64) waited for more than 72hrs after perceiving RFM[124]. Data on the timing of care seeking after perception of RFM on the rest of stillbirth cases was not available in this study. The authors concluded that delay in maternal reporting of RFM is associated with stillbirth[124]. It is hypothesised that RFM might be an adaptive strategy of the fetus to suboptimal intrauterine conditions[124]. Utero-placental insufficiency can produce a redistribution of blood to the fetal brain and heart instead of the skeletal muscle due to a lack of oxygen in utero, which would consequently reduce the movements of the fetus to save resources[118,128]. Other authors have proposed that sudden episodes of excessive fetal activity might also indicate fetal compromise and can lead to stillbirth[129]. It was hypothesised that these sudden episodes of increased activity might be the consequence of fetal seizures, umbilical cord problems, infection, exposure to noxious stimuli or maternal anxiety[129]. However, the evidence that excessive fetal movement is a risk factor for stillbirth, and the understanding of the reasons for it, is limited.

1.7.5 Behaviour and lifestyle factors

Behavioural and lifestyle-related risk factors represent factors that have the potential to be modified and, as such, are the main focus of this Thesis. In this section different behavioural and lifestyle-related risk factors are presented individually, however it is crucial to acknowledge that these behaviours rarely occur in isolation[130–133]. By lifestyle-related risk factors we mean those risk factors that are modifiable through behaviour change, but that are not a behaviour in itself; for instance, weight-related risks that can be modified through nutritional and physical activity intervention. Behavioural and lifestyle-related factors often co-occur and are associated with other issues such as socioeconomic or sociodemographic factors[130–133]. Hence, although most of these behaviours or lifestyle factors have been reported as independent risk factors for stillbirth[134], they need to be understood taking into account all the potential interrelated influences over the risk of stillbirth[135], which will be discussed in the different chapters of this Thesis (Chapters 4-7,9).

Smoking

Cigarettes contain nicotine and other harmful elements that are developmental toxicants. These toxic components have a detrimental effect on the fetal brain at critical developmental periods, restrict the supply flow of oxygen and other essential nutrients and restrict fetal growth, and affect organ and neurological development[136]. In Ireland, according to the latest Perinatal Mortality Report, 13.7% of the 344 pregnant women included in the dataset were smokers at the first time of booking; of these women, only two stopped smoking during pregnancy[3].This figure is higher than the global estimates, a meta-analysis published in the Lancet concluded that the global prevalence of smoking during pregnancy was estimated to be 1.7% with the highest prevalence in the European Region (5.9%). The three countries with the highest prevalence of smoking during pregnancy were Ireland (38.4%), Uruguay (29.7%), and Bulgaria (29.4%), with a global prevalence of 72.5% of women smoking daily [137]. However, these data should be considered with caution, as the quality of some of these studies might be lower than desirable.

In a systematic review and meta-analysis of 25 studies published between 1998 and 2009 conducted by Marufu, Ahankari, Coleman, & Lewis (2015), the authors found that smoking during pregnancy was associated with a 47% increase in the odds of stillbirth (OR 1.47) when using the early definition (20 weeks gestation)[134]. Similarly, in a different meta-analysis, smoking during pregnancy was associated with a 36% increase in the odds of stillbirth at 22 weeks (aOR 1.36), and this risk was bigger in smoking pregnant women of 40 years or older[87]. Similarly, Qu et al. (2020) concluded from their national prospective cohort study conducted in China, that the rate of stillbirth was 0.61% for the maternal smoking group. Further, the authors found that maternal smoking (aOR 2.07) and passive smoking (aOR 1.22), were statistically significantly associated with an increased risk of stillbirth at 20 weeks gestation or more[138]. Varner et al., (2014), conducted a case-control study from 2006 to 2008 to assess the association between smoking during pregnancy and stillbirth defined as fetal death after 20 weeks of gestation. They concluded that increasing amounts of self-reported smoking were associated with an increase in the risk of stillbirth,

showing a dose-response effect. Women who reported smoking 1 to 9 cigarettes per day were 1.7 times more likely (OR 1.77) to have a stillbirth than women who did not smoke, whereas women who reported smoking 10 cigarettes or more had higher rates (OR 2.17)[139].

Some researchers have tried to study the effect of time of smoking exposure on stillbirth. Dodds et al. (2006) concluded in their study that the effect of smoking on stillbirth (defined as the death of a fetus weighing 500 g. or more) was similar in women who quit smoking before 16 weeks of pregnancy (HR 2.6) and women who continued to smoke after 16 weeks (HR 2.2)[140]. The authors reported that these findings give support to the hypothesis that the first trimester of a pregnancy might be more susceptible to the influence of smoking, and that cessation pre-pregnancy would be ideal[140].

The effects of smoking not only affect the pregnancy outcomes during the pregnancy itself but also it has been reported that smoking during a woman's reproductive years can also increase the odds of experiencing stillbirth. Hyland et al., (2014) found that the risk of stillbirth (defined as fetal death at 20 weeks gestation or more) for women who were ever-smokers was higher than for women who were never-smokers (OR 1. 44)[141]. These authors also concluded that never-smoking women exposed to passive smoking had higher odds of suffering from stillbirth than never-smoking women not exposed to passive smoking (OR 1.22)[141].

Alcohol and illicit drug use

Ethanol is the most commonly abused substance during pregnancy and it is a recognised public health problem[142]. Alcohol can cause damage to a developing foetus as it is a teratogen that can cross the placenta, causing brain damage and affecting other developing organs[143]. A systematic review and meta-analysis published by Popova et al., (2017) concluded that, globally, about 9.8% of women consumed alcohol during their pregnancy. These authors found that the countries with a higher incidence of alcohol consumption during pregnancy were Russia

(36.5%), the UK (41.3%), Denmark (45.8%), Belarus (46.6%; based on prediction), and Ireland (60.4%)[143].

Some researchers have tried to establish a correlation between the amount of alcohol consumed and the risk of stillbirth by studying the maximum amount of alcohol considered safe for consumption. Andersen et al., (2012) conducted a study where they collected data about the number of drinks (bottles of beer, glasses of wine, and glasses of spirits) that pregnant women were consuming weekly. The answers were coded as none, less than one bottle/glass per week or the number of bottles/glasses per week. The authors found in their study that the adjusted risk of stillbirth (fetal death after 22 completed weeks of gestation) was higher as the number of drinks consumed per week increased from ½ to 1 drink, 2 to 3½ and 4 or more drinks (HR 0.90; 0.88; 1.20 respectively)[144]. Cornman-Homonoff et al. (2012) conducted a study of 96 pregnant women who drank at least 4 drinks or more per day (48g of alcohol). Data were recorded in grams and standardised with one drink equalling 12 g. Study results showed a "*borderline significance*"[145] (p = 0.06) increase in the risk of stillbirth (defined as fetal death at 20 weeks) compared to the unexposed group[145]. However, the researchers also compared the stillbirth rate in the exposed group included in their study (3/96, 3.1%), with the stillbirth rate recorded in the Chilean National Data (7087/1.567.315, 0.45%), as there were no cases of stillbirth in their control group, concluding that the difference in the rates was statistically significant (p = 0.019)[145]. Further, a recent study conducted in South Africa explored the risk of stillbirth of pregnancies exposed to both alcohol and smoking consumption showed that the adjusted relative risk of late stillbirth (≥28 weeks gestation) was almost three times higher (aRR = 2.78) for dually exposed pregnancies. Additionally, the authors found that the risk of stillbirth at ≥20 weeks gestation is also almost two times higher than in the not exposed/quit early group (aRR 1.75)[138].

However, the evidence associating the risk of stillbirth with low-moderate prenatal alcohol consumption is less clear. Henderson et al. (2007) conducted a systematic

review to assess the effects of low-moderate levels of prenatal alcohol consumption (83g/week) compared with no consumption. The authors did not find evidence of an association between low-moderate levels of alcohol consumption and adverse outcomes, however, this does not mean that drinking at those levels during pregnancy is safe[146]. Given that no safe level of alcohol consumption during pregnancy has been identified, the Australian Guideline to reduce Health Risks from Drinking Alcohol and the Scottish Guideline for Pregnant Women and the National Institute of Health and Care Excellence (NICE) Quality standard [QS204] recommend that women who are pregnant or planning to become pregnant should not drink alcohol[147,148].

The drugs that have been more commonly found to be used during pregnancy besides tobacco and ethanol are cocaine, amphetamines, opioids, marijuana, hallucinogens and toluene-based solvents[149,150]. Establishing the prevalence of illicit drug use in pregnant women is very difficult given issues with self-report of use however[151]. The 2020 perinatal mortality data in Ireland showed that six women had a documented history of drug misuse before pregnancy and four had a history of drug misuse during pregnancy[3].

Illicit drug consumption and its impact on pregnancy outcomes is also represented in the literature on stillbirth risks. Varner et al. (2014), concluded in their study that a positive test for any drug in the umbilical cord homogenate was associated with an increase in the risk of stillbirth at 20 weeks gestation (OR 1.94), and this risk was even higher in women with a positive toxicology screen who also reported having used illicit drugs (OR 3.30)[139]. The most common drug that these authors found was tetrahydrocannabinolic acid (THCA), which is the active component of cannabis, and this was significantly associated with a higher risk of stillbirth (OR 2.34)[139]. Following THCA, the most commonly found drugs were hydrocodone and morphine[139].

Use of medical drugs and supplements

The evidence regarding the safety and risk associated with using medically prescribed drugs, over-the-counter drugs and food or herbal supplements during pregnancy is

conflicting. A study conducted in Europe, North and South America and Australia reported that almost 80% of pregnant women used at least one legal drug during their pregnancy, and around 60-70% have reported using at least one over-the-counter medication[152]. According to an epidemiological study conducted by Mitchell, Gilboa, and Werler (2011), by 2008, 50% of women reported using at least one medication. Some of the most common drugs used by pregnant women are antihistamines, decongestants, expectorants and antitussives, analgesics and antipyretics, topical creams and antacids and antidiarrheal agents[154].

Antihistamines and antiemetics are used during pregnancy for the treatment of nausea and vomiting principally and approximately 15% of pregnant women use these medications[155,156]. Etwel et al. (2017) undertook a systematic review and meta-analysis to study the effects of antihistamines. The authors concluded that the risk of stillbirth after 20 weeks gestation was not higher for women who had used antihistaminic agents during the first trimester (OR 1.23). Decongestants are used to treat conditions such as respiratory tract infections, allergic rhinitis, or pregnancy rhinitis[158]. Källén and Olausson (2006) conducted a study on the effects of the use of oral decongestants during pregnancy. The authors concluded that there was no statistical effect on the use of decongestants on any of the pregnancy outcomes they looked at, with an OR for perinatal death of 0.53[159].

Other commonly used drugs during pregnancy are analgesics such as acetaminophen (paracetamol), ibuprofen, naproxen, aspirin and others. More than 60% of women use acetaminophen during pregnancy, and around half of them do so in the first trimester[155]. A review published in 2010 aimed to study the pregnancy outcomes of women who had used acetaminophen during their pregnancy for analgesic or antipyretic purposes. As mentioned above, Pastore, Hertz-Picciotto, and Beaumont (1999), found an increase in the risk of stillbirth after using pain medication in the first two months of pregnancy[160]. However, there was no increment in that risk when looking specifically at acetaminophen (aOR 1.3)[160]. There are a number of studies that look at the benefits of aspirin in women with the previous history of pregnancy

loss with no reported issues. Aspirin has been used in women with a history of recurrent miscarriage showing benefits by improving vascularisation and altering the uterine blood flow[161]. The EAGeR Trial (Effects of aspirin on Gestation and Reproduction) concluded that, compared to placebo, women who adhere to low-dose aspirin use for at least 4 days per week over the entire follow-up period had fewer pregnancy losses (RR 0.69), and increase odds of livebirths (RR 1.33) in their subsequent pregnancies[162]. The authors also concluded that the effects of aspirin use were greater if the treatment was initiated before conception and continued throughout pregnancy in comparison with starting treatment at 6 weeks pregnancy[162] .

Brogly et al. (2021) conducted a cohort study of pregnancies that had been exposed to opioid analgesic prescriptions (butorphanol, buprenorphine for pain, codeine, fentanyl, hydrocodone, meperidine, methadone for pain, morphine, opium, oxycodone, pentazocine, tapentadol, and tramadol) in women with no history of opioid dependency or consumption. The authors concluded that exposure to prenatal opioid analgesics during the first and second trimesters increased the risk of stillbirth[163] (RR 1.2 and RR 1.3 respectively). Additionally, there is increased recognition internationally that prescribed opioid medication can cause addiction[164], which might become an additional challenge for pregnant women who may become dependent[165].

Antibiotics account for almost 80% of the medication that is usually prescribed during pregnancy, and about 20 to 25% of women will take antibiotics during pregnancy[166] because in some clinical scenarios antibiotics are used in second and third-trimester pregnancies to prevent infections. Ziv et al. (2018) conducted a systematic review and meta-analysis to assess the safety of Quinolones during pregnancy. The authors concluded that exposure to Quinolones during pregnancy does not increase the risk of birth defects or stillbirth (OR 1.32)[167].

According to a study conducted in Canada, around 18% of pregnant women had used a psychotropic drug (antidepressants, anxiolytic/sedative-hypnotics, antiepileptics,

antipsychotics, lithium, stimulants) before, during or after pregnancy, and this tendency has increased significantly over the years[168]. Management of depression with non-pharmaceutical interventions is generally recommended. However, major depression is frequently accompanied by antidepressant medication[169] which has led to a study of the safety of these medications during pregnancy. Stephansson et al. (2013) designed a population-based cohort study in Nordic countries to study the risk of stillbirth associated with exposure to selective serotonin reuptake inhibitors (SSRIs) during pregnancy. The results show that the risk of stillbirth for women who had been exposed to SSRIs was higher than those who had not (4.62 vs. 3.69 per 1000 births, OR 1.25)[169]. However, when the authors adjusted the data for maternal characteristics, country and year of birth, exposure to SSRIs was no longer significantly associated with a higher risk of stillbirth (aOR 1.17)[169]. Based on these data, the authors concluded that the higher rates of stillbirth among women exposed to SSRIs was likely to be related to the severity of the psychiatric diseases and other risk factors rather than to the exposure to the SSRIs themselves. It is important to consider the different influences that factors associated with mental health disorders might be having on the risk of stillbirth. As noted previously, mental health disorders are commonly associated with other socioeconomic challenges[170] and substance use[171], as well as the use of pharmaceutical medications. Hence, better quality studies are necessary to study the potential cofounders over the risk of stillbirth when studying mental health disorders.

Herbal remedies are also widely used during pregnancy for different purposes such as palliating morning sickness, urinary tract infections, nausea or vomiting. The number of pregnant women using herbal supplements ranges from 7% to 55% depending on different geographical, social and cultural settings and ethnic groups[172]. Holst et al. (2010) also undertook a review of the literature and surveyed women that were at least 20 weeks pregnant to assess the safety and efficacy of the most used herbs during pregnancy. The authors found from their surveys that 57.8% of the pregnant women included in their study had used at least one herbal remedy, the most widely used being herbal preparation of ginger (33.6%), cranberry (25.3%),

raspberry leaf (23.7%), chamomile (13.1%), peppermint (9.3%) and Echinacea (4.3%). However, there is little data about their safety in human pregnancy[158,172].

Smeriglio, Tomaino, and Trombetta (2014) conducted a review of the literature for data on the most frequently used herbal remedies during pregnancy and their potential side effects for the mother and the fetus. The authors concluded that few clinical trials have been performed to assess the safety and efficacy of herbal remedies[174]. Although some of these herbs have been classified as safe and effective, not enough data about their use in pregnancy and lactation is available (EMA 2018). Some of the studies included in this review criticise the use of herbal medicines to treat conditions such as urinary tract infection, as this infection, without the appropriate treatment, could lead to complications and adverse pregnancy outcomes such as preterm delivery and stillbirth. The authors concluded that none of the studies included in their review detected a great risk to the mother or fetus from the use of herbal substances as long as they are not used in absence of evidence-based treatments, however, the studies are very limited in size and normally do not include the effects pregnancy outcomes in their measurements[174].

Some authors have also studied the effect of the use of daily multiple micronutrients on the risk of stillbirth or as an intervention to prevent stillbirth. Ronsmans et al. (2009) undertook a meta-analysis including twelve randomised controlled trials comparing either the use of one recommended dietary allowance of multiple micronutrients or a dose of folic acid and iron in low-income countries. The authors defined stillbirth as the death of a fetus after 28 weeks' gestation but before delivery. Results of the meta-analysis showed that supplementation of multiple micronutrients during pregnancy did not result in a reduced risk of stillbirth (OR 1.01). The researchers found that the heterogeneity was low for stillbirth (I2=3%, p=0.42) so they excluded a large study conducted in Indonesia, where the OR for stillbirth was 1.12. Smith et al. (2017) published a meta-analysis including 17 randomised controlled trials on the effects of maternal multiple micronutrient supplementation on stillbirth (no specific definition) and other birth outcomes, in

low-income and middle-income countries. The authors compared the use of micronutrient supplements containing iron-folic acid against using iron-folic acid only. The results showed that there was no effect of the multiple micronutrient supplements on stillbirth (RR fixed effects 0.92; RR random effects 0.98), however, the authors found a reduction of 15% on the risk of neonatal mortality and other outcomes such as very low birthweight, low birthweight, early preterm birth, preterm birth and small for gestational age. The authors also concluded that there was an increased risk of being born large-for-gestational-age among women who had taken the supplementary micronutrients. These effects varied depending on the gestational week when women initiated using the supplements.

Maternal weight

Maternal weight, although not a behaviour itself, is an important lifestyle factor that has the potential to be modified through interventions addressing behavioural aspects including nutrition, diet and physical activity. According to the World Health Organisation, obesity has almost tripled since 1975[177]. In 2016, 39% of adults were overweight, and 13% had obesity[177]. The prevalence of overweight and obesity during pregnancy is hard to establish due to the lack of population-based surveillance data of BMIs during pregnancy[178]. However, surveillance data of women of reproductive age provides insights, and an increasing trend, similar to the one in the general population, has been observed[178,179]. According to Vahratian (2009), around 23% of women of reproductive age (20-44 years old) are considered to have obesity in the USA. The World Obesity Atlas 2022 predicts that 1 in 5 women and 1 in 7 men will be living with obesity by 2030[181]. The relationship between obesity and stillbirth is unclear, although the increased risk of fetal death caused by placental dysfunction is well supported, which can be a consequence of metabolic complications[182]. Some authors hypothesise that obesity may induce insulin overproduction though a biochemical pathway different to diabetes, which might be leading to uncontrolled fetal growth[183]. Another hypothesis is the association between sleep apnea and hypoxemia with obesity that can lead to fetal growth restriction and could potentially explain the high prevalence of babies with a birthweight under the 10th centile in

women that had obesity[183]. Further, some deaths in women who had obesity have also been associated with increased risk of infections, birth complications and congenital anomalies[182].

In the latest Irish perinatal mortality data, women who had obesity (BMI ≥30) were overrepresented (30.2%, n = 98 of 324) among those who experienced a perinatal death in 2020 (PM rate of 8.4 per 1000 births); women who had obesity had almost twice the risk of experiencing a perinatal death compared to women with a healthy BMI in 2020 (RR 1.80)[3]. Chu et al., (2007) conducted a systematic review and meta-analysis of the available epidemiologic evidence establishing the association between maternal overweight and obesity and the risk of stillbirth. The meta-analysis included 9 studies in total; three of which did not define stillbirth, one defined stillbirth as a fetal loss at 22 weeks or later, and five defined stillbirth as fetal death at 28 weeks or later[184]. Findings of this review indicated that the odds of stillbirth were 1.47 and 2.07 times higher among overweight (BMI 25 – 29.9) and obese (BMI ≥30) pregnant women respectively compared to women who were not overweight or did not have obesity[184]. According to their estimations, the risk of stillbirth was twice as high among pregnant women with obesity compared to pregnant women without obesity[184]. Yao, Goetzinger, & Caughey (2018) undertook a study to examine the risk of stillbirth at 24 weeks gestation or more with increasing severity of obesity and the impact at different gestational lengths. The results of this study show a dose-response effect[186]. The levels of HR increase significantly as overweight increases in severity but also as the pregnancy progresses, reaching a peak point of 5.84 (CI 95% 2.15 – 15.85) at 40-42 weeks gestation in cases of extreme obesity[186]. In all five categories of severity of overweight (overweight [25.0-29.9 kg/m^2], CLASS I [30.0-34.9 kg/m^2], CLASS II [35.0-39.9 kg/m^2], CLASS III [40.0-[429.9 kg/m^2], and Extreme Obesity [≥50 kg/m^2]), the highest level of HR is found at 40-42 weeks gestation[186]. At term (37 to 39 weeks), the risk of stillbirth was 1.3 times higher (HR 1.36) in the overweight group compared to the normal weight group, and 2.29 times higher (HR 2.29) in the extreme obesity group[186]. All HR at any pregnancy interval was significantly increased compared to normal weight pregnancies[186]. These results are

in concordance with results found previously by the same group[187]. The authors concluded that almost 20% of the stillbirths included in their study were associated with obesity. Furthermore, the results showed that almost 25% of the stillbirths that occurred at term (37 weeks gestation or more) were associated with obesity[187]. Similarly, Elberle et al. concluded in their retrospective population-based cohort study that women living with overweight or obesity had an increased risk of stillbirth, which increased with each BMI class and gestational age (obese class 1 at 39 weeks (OR 1.15), obese class II at 38 weeks (OR 1.21) and obese class III at 37 weeks (OR 1.30)[181].

Bodnar et al. (2015) conducted a study to identify possible pathophysiologic contributors or causes of stillbirth associated with pre-pregnancy obesity. The authors found that obese and overweight women were more likely to have a stillborn baby (defined as fetal death at 16 weeks gestation) due to placental disease (Adjusted HR 1.3; 2.0 respectively), maternal hypertension (Adjusted HR 1.6; 2.2 respectively), fetal genetic or structural abnormalities (Adjusted HR 1.3; 2.2 respectively), umbilical cord abnormalities (Adjusted HR 1.8; 3.3 respectively) and antepartum infection (Adjusted HR 1.3; 2.0 respectively)[188]. However, it was not associated with stillbirth caused by obstetric conditions (Adjusted HR 1.2; 1.3 respectively) or placental abruption (Adjusted HR 1.1; 1.4 respectively) [188]. Obesity during pregnancy can cause gestational diabetes and gestational hypertensive disorders, two conditions that have been associated with stillbirth and have been discussed in this review, and although obesity is also considered an independent risk factor for stillbirth[1,189–191], the joint effect of obesity and PGDM over the risk of stillbirth is stronger than either risk factor alone[183].

Other authors have studied the influence of gestational weight gain (GWG) on the risk of stillbirth. Nohr et al. (2021) conducted a cohort study in Denmark to examine how early gestational weight gain was associated with stillbirth[182]. The authors were able to identify a U-shaped pattern between early GWG and the crude risk of stillbirth, with the highest risk at lower gains (lost weight or gained <50 g per week)

and high gain (gained 300 g or more) in women with a BMI < 25. These patterns were not identified in women with BMI ≥ 25[182]. When adjusting for BMI, age, parity and social status, the risk of stillbirth was only higher for women with a BMI < 25 who gained less than 50gr/week (aHR 1.43)[182]. Yao et al. (2017) reported similar findings in their retrospective cohort study. The authors conclude that inadequate weight gain and weight loss were associated with an increased risk of stillbirth in all BMI categories except in the morbid obesity category, and especially in the weigh-loss group after 36 weeks gestation (HR 18.85)[192]. Amongst women with obesity (≥30.0 kg/m2), or morbid obesity (≥ 40.0 kg/m2), excessive weight gain demonstrated an increased risk of stillbirth from 29-33 weeks gestation (aHR 1.42 and HR 1.73 respectively). Weight loss was not associated with an increased risk of stillbirth in women with morbid obesity at any gestational age[192].

Inter-pregnancy weight changes also have relevance regarding stillbirth. Timmermanset al. (2020) conducted a systematic review to study the effects of inter-pregnancy changes in BMI on pregnancy outcomes. The authors conclude that, regardless of the BMI category, interpregnancy BMI gained resulted in a higher incidence of GDM, general hypertension, preeclampsia, caesarean section delivery, and large for gestational age infants[193], which are also all risk factors for stillbirth.

Sleeping habits

Sleeping in the supine position has been associated with an increased risk of stillbirth[194], but other sleeping related habits have also been explored in the literature. Stacey and Thompson (2011) conducted a prospective population-based cohort study in New Zealand assessing maternal snoring, daytime sleepiness, and sleep position at the time of going to sleep and on waking (left side, right side, back or other) and the risk of late stillbirth (at 28 weeks gestation or later)[196]. The authors concluded that snoring was not associated with a higher risk of stillbirth when comparing the results between women who snored and had a stillbirth, and women who snored and had a living baby (adjusted OR 1.12)[196]. There were no differences between levels of daytime sleepiness either (P=0.51)[196]. However, the researchers

found differences in the risk of late stillbirth depending on the maternal sleep position both at the time of going to sleep and at the time of waking up, especially during the last night of the pregnancy, but also during the last month[196]. The authors concluded that the risk was higher for those women who did not go to sleep or wake up on their left side (aOR left side 1.00; right side 2.54; and back 2.54), and they determined sleeping on the back to be the least safe position[196].

O'Brien et al conducted an internet-based survey case-control study of women who had a stillbirth at ≥28 weeks' gestation. The authors concluded that sleeping over nine hours in the previous month before the stillbirth and waking up on the right side were associated with an increased risk of stillbirth (aOR 1.75 and aOR 2.27 respectively). The authors also found that non-restless sleep in the last month and not waking up more than one time on the last night before the stillbirth was also associated with an increased risk (aOR 1.73 and aOR 2.03 respectively). However, these authors did not find a relationship between the risk of stillbirth and the going to sleep position, although only 2.4% of the women in their sample reported settling to sleep in the supine position[197].

Another study exploring the risks associated with sleeping position concluded that a supine sleeping position on the last night of pregnancy before the stillbirth was associated with a higher risk of stillbirth (adjusted OR 3.67). These authors also concluded that the risk was higher for term (aOR 10.26) compared with preterm stillbirths (aOR 3.12)[198], which is in concordance with the results found by the The Midland and North of England Stillbirth Study (MiNESS) study that reported that women who had a late stillbirth were 2.3 times more likely to have reported a supine sleeping position on the night before their stillbirth[194].

One hypothesis that explains these results associates the mother's sleeping position with cardiac outputs and fetal oxygenation. Some authors have suggested that the pressure exerted by the enlarged uterus over the vena cava and the aorta when lying on a supine or right lateral position could produce a decrease in the uterus blood flow and subsequently lead to fetal hypoxia[198–201]. This hypothesis has been

confirmed using functional magnetic resonance imaging which associated maternal supine position in late pregnancy with reduced uteroplacental blood flow and oxygen transfer across the placenta[202]. NICE has recently published guidance regarding maternal sleep position and pregnancy[203]. This guidance reiterates that there is evidence in the literature suggesting an increased risk of stillbirth after 28 weeks associated with going to sleep in the supine position compared to going to sleep on the left side. The evidence also suggested that there are no differences in the risk when going to sleep in any other position compared to the left side going to sleep position[203].

Antenatal care attendance

The World Health Organisation currently advises women to attend at least four antenatal visits during their pregnancy[204], when there are no complications. Antenatal care allows healthcare professionals to offer health information about the services available to improve women's pregnancy outcomes, while it is also an opportunity for them to monitor the development of the pregnancy and risks of adverse outcomes such as stillbirth[205].

Stacey et al. (2012) conducted a study to examine the role of antenatal care in the prevention of stillbirth in high-income countries. The authors compared cases of women who experienced a late stillbirth (≥28 weeks' gestation) between 2006 and 2009 in Auckland with ongoing random pregnancies at the same gestational week in which the stillbirth occurred[206]. The authors concluded that gestational age at booking with a prenatal care provider was not associated with a higher risk of late stillbirth; aOR 0.78 when booking between 10 and 20 weeks' gestation and aOR 0.63 when booking after 20 weeks gestation[206]. The number of recommended visits as per the NICE guidelines utilised in this study was 10 for uncomplicated pregnancies. Women who attended less than 50% of the recommended visits were 3 times more likely to suffer from late stillbirth (aOR 2.68); the authors also undertook a trend analysis and found a significant relationship between the decrease in the number of visits attended and the increase in the risk of stillbirth ($p = 0.0005$)[206]. The researchers

also concluded that fetuses who were small-for-gestational age were more likely to be stillborn if they were not identified before birth than those who were identified (aOR 9.46)[206]. This means that identifying small-for-gestational-age (SGA) fetuses could reduce the risk of stillbirth, reinforcing the importance of regular antenatal care attendance as this provides opportunities to detect SGA fetuses[206].

Such previously observed associations may have led to more detrimental outcomes during the COVID-19 pandemic. For instance, Chmielewska et al. (2021) concluded from their meta-analysis that global and fetal outcomes have worsened during the COVID-19 pandemic, with increases in maternal deaths, stillbirths, rupture ectopic pregnancies and maternal depression[207]. The authors suggest that these findings might be associated with the inefficacy of the health-care systems to cope with the pandemic[207]. Another recently published review exploring the influence of the changes imposed on maternity care provision during the COVID-19 pandemic concluded that these changes might have contributed to the general increased rates of adverse pregnancy outcomes, including stillbirth. The reduced antenatal care and reduced uptake of antenatal screening for infections and fetal anomalies consequence of these modifications in antenatal care provision might have contributed to this increase in risks[208].

1.8 Previous efforts to reduce the rates of stillbirth in high-income countries.

In this section, different initiatives taken in high-income countries to prevent stillbirth or reduce stillbirth rates are presented. All of these initiatives acknowledge that a large proportion of stillbirths in high-income countries are avoidable, and these initiatives are designed to reduce the rates of stillbirth by tackling different factors. The two main approaches adopted internationally to improve prevention efforts have two different foci: (1) designing and implementing perinatal mortality clinical audits and perinatal mortality reviews to identify causes and risk factors for stillbirth, and (2) raising awareness regarding risk factors for stillbirth and changing clinical practice to identify and intervene in risk pregnancies though public health initiatives. The approaches discussed in this section are based on two main focal points.

1.8.1 Perinatal mortality audits, reviews and confidential enquiries

Using epidemiological data to identify risk factors and sub-optimal care is essential to produce recommendations that might enhance care in the future, hence, perinatal mortality audits, reviews and confidential enquiries are necessary to help preventive efforts. These types of initiatives and their findings contribute to informing the focus and development of public health campaigns or interventions to tackle modifiable risk factors like the ones discussed below in section 1.8.2.

Definition and aims of perinatal mortality audits and reviews.

A clinical audit is a process that measures practice against standards of care[209]. This process should be described as a cycle, which illustrates the continuity of the process. Hence, a clinical audit involves a process of data collection, analysis, recommendations, implementation and re-evaluation[210]. Perinatal audit is defined as "the systematic, critical analysis of perinatal care, including procedures used for diagnosis and treatment, the use of resources and the resultant outcomes and quality of life for women and their babies"[211]. One of the aims of clinical perinatal mortality audits is to calculate perinatal mortality rates (PMR). The PMR is the number of stillbirths and neonatal deaths per 1000 total births and it is used often as an indicator of the quality of care provided to women and babies[8,212]. National Perinatal Mortality Audits can also serve to identify avoidable deaths and risk factors or factors contributing to suboptimal care as well as inform and enhance clinical practice by making recommendations[210,213].

Local perinatal mortality reviews collect data on the local deaths and associated factors and make recommendations accordingly[210]. Confidential enquiries are a type of perinatal mortality review that is conducted by an expert multidisciplinary team on cases with adverse outcomes that are anonymised and reviewed individually[210], these confidential enquiries also produce recommendations and influence clinical practice, health policy, and education[214]. Despite consensus on the importance of recording data that will allow for the identification of modifiable risk factors and suboptimal care associated with perinatal deaths, there is no standard method to

conduct perinatal mortality reviews, audits or in-depth confidential enquiries[215]. In 2021 the WHO published a guide entitled "Maternal and Perinatal Death Surveillance and Response: materials to support implementation". This guide proposes standard definitions and instructions on how to perform perinatal mortality reviews or audits, but it primarily focuses on setting them up at the facility level[216].

In addition, there are a number of factors that impact the reporting of perinatal mortality. For instance, Gutman et al. (2022), found in their systematic review issues related to completeness and standardisation of data such as lack of standardised reporting criteria, differencing in classification and differences in definitions. Other barriers identified regarding the completeness of data were poor staff training and capacity, lack of standardisation of audit methodology, lack of guidelines, poor guidance for data collectors, and poor data collection systems. Further, the lack of linkage between medical and civil records was also identified as an issue[217]. Despite such issues however, the data obtained through national clinical audits and reviews in order to identify risks and suboptimal care has shown results around different high-income countries in reducing the rates of stillbirth. These reductions are discussed in the following sections.

Perinatal national mortality audits and reviews conducted in high-income countries

Only four high-income countries are currently conducting national perinatal mortality audits: Ireland, the UK, New Zealand, and the Netherlands[218].

Ireland

The National Clinical Perinatal Mortality Audit in Ireland is conducted by the NPEC[213]. This audit was established in 2009, and produces perinatal mortality reports with the collaboration of a multidisciplinary perinatal mortality group annually, with the exception of 2018 and 2019 where the report presented biannual data[8]. The NPEC also generates hospital-specific reports. The nineteen maternity units across Ireland submit data on all cases of stillbirth, early neonatal death and late neonatal death through the NPEC's online data collection system. For this audit and in accordance with the Irish Stillbirth Registration Act[219], stillbirth is defined stillbirth as a child

weighing 500 grams or more or having a gestational age of 24 weeks or more who shows no sign of life.

United Kingdom

The national perinatal clinical audit is conducted in the UK by National Perinatal Epidemiology Unit (NPEU) and Mothers and Babies: Reducing Risk through Audits and Confidential Enquiries across the UK (MBRRACE-UK). This audit collects data on all perinatal deaths from 22 weeks across England, Scotland, Wales and Northern Ireland. MBRRACE-UK publishes reports on their findings every year including perinatal deaths from 22+0 weeks gestational age (including late fetal loses, stillbirths and neonatal deaths) of babies born between the 1st of January and the 31st of December of a given year[63]. The report defines late fetal losses as a baby born between 22+0 and 23+6 weeks gestational age showing no signs of life; stillbirths as a baby born at or after 24+0 weeks gestational age showing no signs of life, irrespective of when the death occurred; and neonatal deaths as a liveborn baby (born at 20+0 weeks gestational age or later) who died before 28 completed days after birth. Deaths of babies under 24 weeks of gestational age and terminations of pregnancy are excluded from the mortality rates reported. MBRRACE also produces reports for individual organisations / Trusts[220] and conducts confidential enquiries on selected topics[221]. Currently, MBRRACE is conducting a confidential enquiry on baby deaths to mothers of black, Asian and white ethnicity[222].

Also in the UK, the Royal College of Obstetricians and Gynaecologists (RCOG) has led the national quality programme called "Each Baby Counts"[223]. The aim of this programme was to reduce the number of babies who die during term labour (intrapartum stillbirths)[223] by conducting local investigations across the UK into stillbirths and neonatal deaths. The Each Baby Counts programme was launched in 2014 and has the ambition of reducing perinatal deaths and injuries by 50% by 2025[223]. In their 2020 final report, the RCOG stated that the programme has been useful to raise the profile of maternity safety and the importance of this cohort of babies, providing definitions that were used by multiple agencies, and generating

recommendations[223]. The latest EMBRRACE-UK report states that stillbirth rates have decreased in the UK by a 21% from 4.20 per 1,000 total births in 2013 to 3.33 per 1,000 total births in 2020[19].

New Zealand

New Zealand's national perinatal mortality audit is conducted by the Perinatal & Maternal Mortality Review Committee (PMMRC) and was established in 2005[218]. This audit has the aim of reviewing and reporting on perinatal deaths in order to decrease their perinatal mortality rates and improve care[224]. The audit collects data on any perinatal death defined as any infant death from 20 weeks gestation or at least 400 grammes up until 28 days of life. Data are provided by healthcare professionals in the different maternity units through a website platform. The PMMRC publishes publicly available reports with national findings and recommendations to improve care[224].

The Netherlands

The Netherlands has a system of national audits conducted by Perined, which is a merger organisation of PAN (Perinatal Audit Netherlands) and PRN (Perinatal Registry Netherlands), founded in 2015[225]. The Perinatal Audit Netherlands (PAN) was established in 2010 as a result of the comparison of the Netherlands' perinatal mortality rates and reduction progress with other countries. This audit includes babies born at least 22 weeks gestation[225]. As with the other national audits, the Perined Perinatal National Audit also has a standardised online data collection tool that healthcare professionals use to report perinatal deaths[225]. Perined publishes reports with national data and special articles focusing on specific topics which include recommendations[218].

Regional audit and/or data reporting

Some countries report data on their perinatal death figures, but do not have an audit or review process that generates recommendations to improve care. For instance, Canada publishes perinatal mortality rates composed of perinatal deaths defined as the death of a child under one week of age or a stillbirth of 28 or more weeks

gestation, late fetal deaths defined as a stillbirth of 28 or more weeks gestation, and early neonatal deaths which is the death of a child under one week of age (0 – 6 days)[226]. In the USA, the Centre for Disease Control and Prevention publishes a report on fetal mortality rates, including fetal deaths at 20 weeks gestation or more[227]. The descriptive report provides information about general fetal mortality rates, associations with specific risk factors and causes of death, but no recommendations for improvements[227]. Similarly, Australia also reports their perinatal mortality rates on the Australian Institute of Health and Welfare website. The definition of stillbirth used in Australia is a fetal death before the birth of a baby born at 20 weeks gestation or more, and/or weighing 400 grams or more[228].

There are other initiatives conducted internationally to reduce the rates of stillbirth through auditing and case reviews. The Euro-PERISTAT Network is a project comprising representation form 31 European countries to establish a high-quality European perinatal information system[229]. UNICEF, the WHO, the World Bank Group and the United nations also recently published a report with data on stillbirth globally, highlighting global inequalities and progress made in the last two decades, as well as projected trends for the future[230]. Finally, the "Making Every Baby Count" guidelines for audit and review of stillbirth and neonatal deaths, published by the WHO, outlines key steps to introducing a system for capturing the numbers of stillbirths and neonatal deaths and reviewing selected cases systematically[230].

1.8.2 Raising awareness: Public health approaches internationally

The Public Health initiatives discussed in this section include components related to awareness about behavioural risk factors or behavioural modification components to reduce stillbirth rates. Inclusion of these components is what makes them especially relevant to this Thesis given its focus on modifiable behavioural risk factors. To date, there has not been any initiative involving behavioural risk awareness and/or modification components carried out in Ireland. The following sub-sections are examples of initiatives conducted in different high-income countries that could serve as examples to inform the development of an initiative in Ireland.

Scottish Patient and Safety Program (SPSP) Maternity and Children Quality Improvement Collaboration (MCQIC)

The SPSP is a national initiative launched in 2008 with the objective of improving the whole of National Health service (NHS) Scotland. Although this programme focuses on different settings and aspects of the Scottish health care system, it also has a specific focus on maternity and children's care[231]. The SPSP Maternity and Children Quality Improvement Collaboration (MCQIC) was launched in 2013 and covers three areas: maternity, neonatal, and paediatrics. The SPSP MCQIC is currently focusing on reducing stillbirth rates, reducing postpartum haemorrhage rates, reducing neonatal mortality, and highlighting the importance of the essentials of safe care in maternity care[232].

The stillbirth prevention component of the SPSP MCQIC programme focuses on assessing fetal wellbeing, monitoring fetal movements and fetal monitoring. In terms of behavioural risk factors, HCP are encouraged to engage in discussion with women highlighting: the importance of being aware of baby's movements and their babies' uniqueness; ensuring women are making contact with maternity units if concerned; the importance of raising awareness about the link between reduced fetal movements and fetal wellbeing; and how this can be associated with the risk of stillbirth[233]. Furthermore, in terms of behavioural risk factors, the programme also aims to offer all women carbon monoxide monitoring when booking their antenatal care appointment, to refer women who are smokers to smoking cessation services and to provide tailored packages of care to all women who continue to smoke. From inception in 2013, the programme has supported a national aggregated reduction in stillbirth rates of 22.5%[234].

NHS Saving Babies' Lives Care Bundle (SBLCB), United Kingdom.

The NHS SBLCB is a care bundle designed and implemented in the NHS to reduce perinatal mortality. This initiative was created in response to the 2014 announcement of the Secretary of State for Health of a new ambition to reduce the rate of stillbirths by 50% in England by 2025, and by 20% by 2020[235]. The NHS SBLCB

comprises four key elements of care based on available evidence to reduce stillbirth rates. These elements are: reducing smoking in pregnancy; risk assessment and surveillance for fetal growth restriction; raising awareness of reduced fetal movements; and effective fetal monitoring during labour[236]. The second version of the care bundle includes a fifth element: reducing preterm birth[237]. This care bundle is a good example of addressing different behaviours at the same time to reduce the rates of stillbirth, since, as mentioned above, these behaviours rarely occur in isolation[130–133]. The NHS SBLCB initiative uses training for healthcare professionals, information leaflets and videos for patients, social media engagement (i.e., hashtags like #movementsmatter, #quit4baby, #sleeponside, #growingmatters, #letstalktiming) and informative websites as resources. The smoking elements include carbon monoxide testing of all pregnant women at antenatal booking appointments and referral to smoking cessation services by midwives[236]. The other behavioural element, monitoring fetal movements, includes providing information to women through a leaflet with information and guidance in case of concerns, and the use of a checklist to manage the care of women who report reduced fetal movements[236].

The evaluation of this care bundle across different participating NHS Trusts concluded that stillbirth rates have declined by 20% over the period in which the SBLCB was implemented[238], but the authors were not able to determine if the implementation of the SBLCB or any of its components per se was associated with the reduction in rates of stillbirth[238]. Regarding smoking, the authors found that the proportion of women recorded as smoking at delivery declined from 14.3% before SBLCB to 11.8% after SBLCB[238], however, there was no evidence of an increase in smoking cessation rates[238]. The reports states that this decline in the proportion of women recorded as smoking at delivery might be a reflection of societal changes as less women were recorded as smokers at booking[238]. Carbon monoxide testing was found to be acceptable, but referral and engagement with smoking cessation services were poor[238]. Regarding awareness of monitoring fetal movements, the authors

concluded that awareness was good and that most Trusts were using the information leaflet provided[238].

The latest MBRRACE-UK report with 2020 data states that rates of stillbirth have reduced by 21% from 4.20 per 1,000 total births in 2013 to 3.33 per 1,000 total births in 2020[239]. While not entirely attributable to the NHS SBLCB, reductions in stillbirths recently reported by MBRACE-UK could be related to the care bundle[238]. The wider uptake of the Care Bundle started in 2017 in England, which correlated with a fall in the stillbirth rates of 5.1% from the stillbirth rate in 2016[238].

National Stillbirth Action and Implementation plan and the Safer Baby Bundle, Australia

The National Stillbirth Action and Implementation plan recognises stillbirth as a public health issue in Australia. The focus of this plan is on stillbirth after 28 weeks, and its overarching goal is to support a sustainable reduction in rates of preventable stillbirths, with a goal of 20% or more reduction over five years from 2020[240]. The plan includes five priorities: 1) ensuring high-quality stillbirth prevention and care; 2) raising awareness and strengthening education; 3) improving holistic bereavement care and community support following a stillbirth; 4) improving stillbirth reporting and data collection; and 5) prioritising stillbirth research[240]. Each priority has several action areas associated.

Some of the main considerations that this National Stillbirth Action and Implementation plan highlights is making sure that stillbirth prevention is linguistically and culturally appropriate, which might be achieved by co-desigining and implementing strategies and tools to reduce stillbirth rates in collaboration with communities at increased risk[240]. The plan also proposes that information on how to discuss risk factors for stillbirth with women should be included in the national clinical practice guidelines[240]. Further, the plan proposes promoting community awareness by developing a community awareness package with consistent information about stillbirth, its prevalence and its associated risks and the creation of a national stillbirth education program for health professionals[240]. Other elements

of this National Plan are related to enhancing bereavement care, improving care in subsequent pregnancies for bereaved mothers, and providing national guidelines on bereavement care following a stillbirth[240] as well as improving stillbirth reporting and data collection through standardised investigations and reporting of stillbirth[240].

Action area 1 "Implementing best practice in stillbirth prevention" which is associated with priority one of this plan involves implementing the Safer Baby Bundle (SBB)[241]. Implementation started in the State of Victoria in 2019 and it has been implemented widely in the following years[241]. The SBB has five main objectives: supporting women to stop smoking during pregnancy, improving detection and management of impaired fetal growth, increasing awareness about fetal movements, providing advice on maternal sleep position, and supporting shared decision-making around the timing of birth[242]. This care bundle also highlights the importance of continuity of care, especially in women with high-risk of stillbirth, and the need to establish a perinatal mortality audit[240]. Results of the effectiveness of this approach in reducing stillbirth are currently available for the 15 Victorian health services that completed both phases of the Safer Baby collaborative. Results show a decrease in stillbirth rates by 21%, from an average rate of 0.24% to 0.19%. The days between stillbirths increased by 131%, from an average of 3.5 days to 8.1 days, including periods of 42 and 32 days without a stillbirth occurring at any of the reporting sites[243]. The rates of smoking cessation of women during pregnancy increased by 200%, from an average rate of 11% to 33%[243].

Still Six Lives, Australia

Still six lives is a public awareness campaign to raise awareness about stillbirth and reduce stillbirth rates in Australia[244]. The campaign is composed of a website that provides information about stillbirth prevention for women and partners, family stories, and news. The behavioural focus of this campaign is on smoking cessation, monitoring fetal movements, and sleeping on the side after 28 weeks gestation[244]. The website provides a rationale as to why these factors increase stillbirth risk, and provides contact and information support for parents. In addition to the website, the

campaign also includes with videos, podcast scripts and ads and social media posts[244]. The developers of this campaign reported a high number of engagements from media and website users, but potential reductions in the rates of stillbirth associated with this campaign have not been reported to date[244]. This campaign is part of the "Together we can stop stillbirth: Education campaign" around the Safer Baby Bundle discussed above.

Count the Kicks, USA

"Count the kicks" is a public health campaign that was developed in 2008 and designed by five bereaved mothers in Iowa, USA[245]. The campaign has a focus on raising awareness and training mothers to monitor their babies' movements. The campaign provides healthcare professionals with informative posters and brochures to provide to women, as well as videos and resources in different languages, and a social media presence. The campaign also provides a mobile app that is used to inform mothers about the importance of monitoring foetal movements during pregnancy and to facilitate the baby's movement tracking[245]. The authors claim that in the first 10 years of the implementation of the "Count the Kicks" campaign in Iowa, the stillbirth rate in the state of Iowa decreased by 32%[246]. No peer-reviewed publications on the efficacy of this public health campaign are available, however, according to information on their website, some preliminary data on the use of the app shows that users were more likely to contact their healthcare providers with concerns and that there was a reduced rate in stillbirth for the pregnancy in which women used the app compared to earlier pregnancies[247].

Sleep on Side; Stillbirth Prevention Campaign, New Zealand

The Sleep on Side stillbirth prevention campaign is a public health campaign designed and implemented by the University of Auckland, Cure Kids and the New Zealand Ministry of Health[248]. The campaign focuses on raising awareness about the importance of side sleeping during late pregnancy using informative leaflets and videos, as well as a specific website and media presence. However, there is as yet no

available evidence of the efficacy of this campaign over the rates of stillbirth in New Zealand.

Canadian Collaborative for stillbirth prevention (CC4SP)

The CC4SP is a petition developed by stakeholder individuals and organisations to the Canadian Government. It is proposed in this petition that the Australian stillbirth prevention plan (discussed in the section "National Stillbirth Action and Implementation plan and the Safer Baby Bundle, Australia" above) be adapted to the Canadian context[249]. The five key areas that the plan suggests to target are the same exposed in the Australian National Stillbirth prevention plan: ensuring high quality and stillbirth prevention care, raising awareness and strengthening education, improving holistic bereavement care and community support following a stillbirth, improving stillbirth through reporting and data collection, and prioritizing stillbirth research[249]. So far, the organisers are gathering expert letters to be sent to the Health Minister and collecting signatures to force action from the government on developing and implementing this plan[249].

International Stillbirth Alliance (ISA) prevention working group (PreWG)

The PreWG is a group of bereaved parents and researchers working towards enhancing stillbirth prevention as part of the ISA. The PreWG align their work with recommendations made in the Lancet Stillbirth series 2011 and 2016[250] and has four key areas of work: 1) development and implementation of the Improving Perinatal Mortality Review and Outcomes Via Education (IMPROVE) workshop which is an educational program for maternity and new-born health care professionals to train them on best practice care around the time of a perinatal death; 2) the classification system project, which aims to improve the quality of the data on causes of stillbirth and neonatal death though implementing a standardised classification system; 3) increasing parent engagement in perinatal mortality audits; 4) and updating and producing position statements such as the one produced for fetal movement monitoring[250,251].

1.8.3 Potential for improvement in Ireland

As discussed in the previous section, the establishment of perinatal mortality audits and reviews, and the development of public health initiatives with behaviour change components are the two main approaches that have been adopted internationally to reduce the rates of stillbirth.

In Ireland, some efforts have been conducted to date to reduce the risk of stillbirth, including the establishment of the Perinatal Mortality National Clinical Audit mentioned above. Ireland's Perinatal Mortality Clinical Audit was endorsed by the National Clinical Effectiveness Committee (NCEC) in 2022[252,] which highlighted the relevance of this audit and its methodological quality. In their reports, the NPEC produces recommendations and makes comments on the stage of implementation of recommendations made in earlier reports. The NPEC has repeatedly recommended in their reports that "A pubic health education programme on perinatal deaths and modifiable risk factors should be developed"[253,254]. Enhanced efforts should be made to ensure these recommendations are implemented by relevant organisations. Another strategy to reduce the risks of stillbirth is the improved efforts to identify risk factors and provide individualised care plans to all women in the Irish maternity unites, however, limited financial and time resources hinder the day-to-day implementation of this measure[253].

In 2016, the National Women and Infant Health Programme (NWIPH) launched the National Maternity Strategy Implementation Plan. One of the recommendations proposed in this report is that antenatal care should *"encompass a holistic approach to the woman's healthcare needs including her physical, social, lifestyle and mental health needs"*[255]. One of the action plans associated with this recommendation is to develop the "Make every contact count (MECC)" programme available for all maternity units[255]. This programme focuses on increasing awareness and detection of issues related to mental health, domestic violence, substance use and lifestyle issues[255]. The National Maternity Strategy Plan (NMSP) makes other recommendations relevant to the behavioural risk of stillbirth such as having dietetic

and smoking cessation services in each maternity unit, developing a consistent approach towards informing women about the risks of alcohol consumption, providing services with Drug Liaison Midwives, providing additional support to vulnerable women, and standardising access to perinatal psychiatry and psychology services[255]. However, these recommendations are not framed within the context of stillbirth prevention. In the NMSP Revised Implementation Plan for 2021-2026 published in 2021, most of these initiatives are in progress but some others, like the implementation of the MECC programme, have not been started yet[256].

Currently, a National Clinical Practice Guideline focusing on all aspects relating to stillbirth is being developed in Ireland. This guideline focuses on risk factors, diagnosis, investigations, management, classification, audit and review, follow-up care, and future research priorities[257]. The guideline includes a section on the identification of risk factors and proposes adequate clinical practices and recommendations. Hence, regarding modifiable risk factors, the guideline proposes the following recommendations:

- "Women should be screened for modifiable risk factors for stillbirth at their booking visit, as simple antenatal intervention may significantly impact on antenatal course and clinical outcomes."[257]
- "While some modifiable risk factors may be addressed in a normal-risk pregnancy, some may warrant specialist multidisciplinary input and management in the specialised care pathway."[257]
- "Women should be included in the decision-making process as maternal engagement is important for tackling modifiable risk factors."[257]

New approaches that can help address the guideline aims can be informed by examples of care bundles, public health approaches, and behaviour change interventions utilised in high-income countries. Evidence from such examples has shown that stillbirth rate reductions are possible and can serve learning points to develop new approaches in countries like Ireland[237,242]. Although Ireland's stillbirth

rates are comparable to other high-income countries, they are rising rather than falling and there is much scope for improvement[253]. To date, no public health campaigns in Ireland exist despite the availability of research showing the lack of awareness about stillbirth of the Irish population[258]. Hence, additional efforts need to be considered. In line with this, in the latest Perinatal Mortality National Clinical Audit Report (2022), NPEC advocates for the introduction of a "Care Bundle" approach in an attempt to reduce the perinatal mortality rates in Ireland the same way that they have been reduced in other high-income countries using similar approaches[3]. Amongst other elements, the care bundle proposed by the NPEC includes a public health awareness programme focusing on reducing smoking in pregnancy, weight management to achieve adequate BMI, raising awareness of stillbirth and reduced fetal movements, and increasing awareness of HCP on modifiable risk factors for PM[3]. The proposed bundle also includes elements of staff training and surveillance for fetal growth restriction using a standard national approach, as well as advocating for effective monitoring of the fetus during labour for a reduction in Neonatal Brain Injury and intrapartum deaths[3]. Such a care bundle, together with all of the other efforts made to date, could have the potential to reduce the rates of stillbirth in Ireland[3].

1.9 Behaviour change

As discussed previously, a number of existing approaches to reduce the risk of stillbirth include components designed to tackle modifiable risk factors for stillbirth. In many instances such factors are behavioural, for instance, substance use, attendance to antenatal care, sleep position or weight management behaviours to reduce weight-related risks[237,242]. Development and implementation of potentially effective and appropriate behaviour change interventions to prevent stillbirth must be based on a good understanding of the behaviours that need to be addressed, as well as behavioural theory[259]. The following sub-sections discuss literature regarding behaviour change during pregnancy, as well as the importance of the use of behaviour theory to inform behaviour change interventions and, will describe some of the most commonly used behaviour change theories and frameworks. Lastly, a methodology for behaviour change intervention design will be described.

1.9.1 Behaviour change during pregnancy

As described in Section 1.7.5, some risk factors for stillbirth are behaviours or lifestyle factors that women can adopt and/or maintain, and that have the potential to be modified. Pregnancy has been described in the literature as a teachable moment[260–262]., which is described as a naturally occurring life transition or health event that teaches individuals to adopt risk-reducing behaviours spontaneously[263]. It is argued that "teachable moments" might contribute to behaviour change by enhancing the person's perception of risk and outcome expectations, provoking strong emotional reactions, and/or forcing a redefinition of one's concept or social role[263]. However, Olander et al. (2015) argue that thinking of behaviour change during pregnancy as something that naturally occurs, as proposed by the Teachable Moments theory[263], is too simplistic and may hinder the discovery of potential fruitful behaviour change possibilities or conceal barriers altering capabilities or opportunities[264]. For instance, even though women might have increased levels of motivation to adopt changes in their behaviour, it is vital to recognise that other psychosocial or medical factors might be acting as barriers or facilitators.

Given that pregnancy represents a period during which women can/may change their behaviours and the previous associations of certain behaviours to undesired pregnancy outcomes such as stillbirth, there has been an effort to design behaviour change interventions targeting such behaviours. For instance, in a scoping review of existing literature related to behaviour change in pregnancy more broadly, rather than in the context of stillbirth[265], the authors identified 30 studies that demonstrated considerable variation in the interventions or programmes used. Their findings show that education has been the predominant approach towards behaviour change during pregnancy; however, as the authors acknowledge, information provision alone is not sufficient, and interventions should address other behaviour change components[265]. Other approaches identified by these authors were health-coaching interventions, motivational interviewing, cognitive behavioural theory (CBT) and national social and behavioural change champaigns[265]. Regarding effects of prenatal behaviour change interventions on behavioural outcomes, Heslehurt et al (2020) conducted an umbrella review examining the effectiveness of interventions delivered during pregnancy on changing women's smoking, alcohol consumption and diet and/or physical activity[266]. The authors concluded that the most consistent evidence was for interventions targeting dietary behaviours compared with the evidence supporting interventions targeting other behaviours[266]. The authors identified a gap in the evidence relating to reviews of interventions to reduce alcohol consumption, as the number of systematic reviews identified for this behaviour was much lower than for the other two (4 reviews for alcohol vs. 16 reviews for smoking and 16 reviews for diet and/or physical activity)[266].

A number of interventions have been developed and examined for smoking cessation. Chamberlain et al. (2017) conducted a systematic review of psychosocial interventions to support women to reduce smoking during pregnancy. Psychosocial interventions may include non-pharmacological strategies such as cognitive-behavioural therapy or motivational and supportive therapy (e.g., counselling, health education, feedback, financial incentives, social support or exercise)[136]. Compared to usual care, the authors found that counselling, feedback, and the use of incentives

had positive effects on smoking cessation rates; the effects of health education, social support from peers and exercise were less clear. The authors concluded that psychosocial interventions have the potential to support women to stop smoking during pregnancy and do not have any overall negative physical or psychological effect on women[136].

Gilinsky (2011) et al. conducted a systematic review of interventions delivered during antenatal care to reduce alcohol consumption in pregnancy. These interventions included motivational interviewing, self-help manuals, supportive counselling, high feedback ultrasound and basic education[267]. The authors concluded that there was some evidence that face-to-face brief interventions had positive effects on alcohol abstinence during pregnancy and that allowing women to choose their own goals and involving their partner also showed positive results[267]. Some of these brief face-to-face interventions incorporate educational aspects and motivational interviewing, while others involve verbal and financial reinforcement[267].

Previous research has also attempted to explore the evidence regarding interventions related to weight during pregnancy. Campbell et al (2011) conducted a meta-analysis of behavioural intervention for weight management in pregnancy. The meta-analysis included findings from 5 controlled trials describing "complex interventions"[268]. Among the interventions, four of them encompassed both dietary and physical activity components, while one of the interventions solely focused on providing dietary advice[268]. The study concluded that there were no significant differences in gestational weight gain amongst participants in the intervention group[268]. The authors also conducted a meta-synthesis of qualitative research to assess factors influencing intervention effectiveness, and concluded that some of the barriers that women described in the qualitative studies were not addressed by the interventions[268]. This findings highlights the importance of collecting enough evidence, including from intervention users, to inform the development of behaviour change interventions. On the other hand, Walker et al (2018) conducted a systematic review and meta-analysis to assess the efficacy of interventions to prevent excessive

gestational weight gain (GWG)[269]. The findings of this meta-analysis indicated that diet interventions including low Glycaemic diet, calorie restriction or healthy eating advice, were more effective than physical activity or lifestyle interventions to prevent excessive GWG[269]. Similarly, Du et al. (2018) explored the effects of physical activity interventions on maternal outcomes in their systematic review and found that prenatal physical exercise in overweight and pregnant women with obesity was effective to reduce gestational weight gain and the risk of diabetes[270]. Other authors have explored interventions to prevent the decline of physical activity during the progressions of the pregnancy. Currie et al (2013) conducted a systematic review of physical activity interventions in pregnant women with the purpose of reducing PA decline during pregnancy[268]. The review findings show that interventions included counselling, structured exercise, and education, and some of the techniques they used were goal setting, planning, feedback, repetition and substitution, shaping knowledge and comparison of behaviours. Eight out of the ten interventions analysed for effectiveness showed positive results whereas women in the intervention arm were more physically active than controls[268].

While there have been multiple interventions designed and implemented to tackle behaviours that have been associated with an increased risk of stillbirth outside the context of stillbirth prevention[271–273], the number of behaviour change interventions that have been explicitly designed to reduce stillbirth rates is limited (as will be discussed further in Chapter 8). In order to enhance the likelihood of effectively reducing stillbirth rates through behaviour change interventions, such interventions should be designed and implemented in the context of stillbirth prevention. Although some interventions tackling the behavioural risk factors for stillbirth already exist, it is important to develop a behaviour change intervention that address all the different behavioural risk factors taking into account the particularities regarding risk perception in the context of pregnancy and stillbirth, which can lead to different behavioural outcomes.

1.9.2 Theories of behaviour change

Behavioural theories are the accumulated knowledge of the assumptions of what behaviour is, how is it influenced, and what mechanisms of action will produce a change in behaviour[274]. Behavioural theories are important because they can help explain and facilitate understanding of the factors that influence behaviour, and through which mechanisms this behaviour is influenced[275]. Applying theory to intervention development is useful for many reasons. Firstly, it facilitates identifying triggers or antecedents of behaviour and its influences, which might facilitate selection and tailoring of the different intervention elements[274]. This means that the causal determinants of change associated with a given behaviour could be directly targeted by the intervention. Secondly, it also allows for further investigation of the mechanisms of action (e.g., attitudes, perceived self-efficacy, motivation) composing a theory which might enhance understanding of why or how an intervention works[274]. Hence, researchers would be able to determine if an unsuccessful intervention did not work because it did not have effects over the hypothesised mechanisms of action, or because the selected mechanisms of action (successfully influenced) had no effects over the behaviour[274]. Thirdly, it enhances the cumulative knowledge on behaviour change in different contexts, populations, or behaviours, and lastly, it enables researchers to test theory through applying theory-based interventions[274]. However, despite these advantages, most interventions are not designed with explicit reference to behaviour theory[276].

Despite the importance and usefulness of behavioural theory, many behaviour change interventions have been designed without evidence or with a poor application of theory[277]. For instance, Prestwich et al (2014) conducted a systematic investigation of the theory used in physical activity and dietary interventions. The results showed that 56% of the interventions reported a theory base, but 90% did not report links between the components of the interventions and specific theoretical constructs[278]. In a systematic review of theory use in the design of guideline dissemination and implementation strategies, only 22.5% of the included studies were found to be underpinned by theories[276]. Similarly, in a scoping review

to identify behavioural and behaviour change theories with potential relevance to public health interventions across psychology, sociology, anthropology and economics, 82 different theories were identified[274]. However, four theories were present in 63% of the interventions that used theories: (1) Transtheoretical Model of Health Behaviour Change, (2) Theory of Planned Behaviours, (3) Social Cognitive Theory and (4) the Information-Motivation-Behavioural-Skills Model[274]. The authors of this scoping review propose several potential reasons why only a few theories might account for more than half of the studies identified, indicating uneven distribution of frequency of use[274]. Firstly, how often a theory is used might be influenced by the year of introduction of that theory, and the level of how much that theory is discussed and hence, the research community has awareness of it[274]. Another hypothesis is that the most used theories might be of better quality because they have stronger evidence to support them. However, it has been shown in previous research that this is not always the case[274]. Lastly, the authors proposed that researchers and intervention developers are not aware of the full range of theories available, and so they choose the most commonly applied in the literature[274]. Irrespective of researcher rationale for choice of behavioural theories, what theory is used and how it is used in intervention development is of importance. The previously found weak associations between the behavioural theory used and the efficacy of an intervention can be a result of poorly applying such theory, or of utilising an inappropriate behaviour theory for a specific behaviour[276]. Some of the most commonly used theories identified by Davis et al (2015) have been applied in the context of behaviour change during pregnancy. In the following paragraph, we will discuss the Transtheoretical Model (TTM) of Health Behaviour Change, the Theory of Planned Behaviour (TPB) and the Social Cognitive Theory (SCT) and their use in the pregnancy context and applicability to stillbirth research.

According to the TTM of Health Behaviour Change, health behaviour change involves progressing through six stages of change which represent the temporal dimension[279]. These phases are defined based on the readiness/willingness of people to engage in behaviour change and range from “Precontemplation” to “Maintenance” and

"Termination" of the behaviour change process. The TTM model has also been used to explain or address some of the relevant behavioural risk factors for stillbirth such as smoking[280], weight-related risks through diet and physical activity[281], and alcohol consumption[282]. Application of the TTM model to behaviour change interventions during pregnancy has involved designing strategies to facilitate women to move from one stage of the model to the next. For example, in the intervention designed by Lowrey (2019) to tackle alcohol drinking behaviours, activities such as decisional balance exercises, readiness rulers, and goal setting[282], whereas Pinheiro de Freitas et al. (2020) used strategies such as reassessment of the environment, consciousness-raising, self-reevaluation, social liberation, contingency management and counterconditioning in their intervention to control weight gain during pregnancy[281]. However, there are some limitations to this theory that are especially relevant for the behaviours associated with stillbirth. The TTM model does not take into account the social context in which the behaviour occur and does not take into account other potential cofounding health behaviours, which is crucial in the case of the behavioural risk factors for stillbirth, where social support and environmental barriers have demonstrated important effects. Further, there is no clear idea on how much time is needed for change and how much time a person can spend in each stage, this is especially relevant to the behaviours associated with stillbirth, where the time window for behaviour change is so short[283,284].

The central factor in the TPB is the intention of the individual. This model assumes that the stronger the intention a person has to engage in a behaviour, the more likely it will be to happen. However, the model acknowledges that the performance of a behaviour might be influenced by non-motivational factors such as the availability of opportunities or resources to perform the behaviour. The TPB states that there are three conceptually independent determinants of intention: attitudes towards the behaviour[285], the subjective norms" [285], and the perceived behavioural control[285]. The TPB has also been used during pregnancy in association with the behavioural risk factors for stillbirth to predict behaviours and to inform intervention development[286,287]. For example, in a meta-analysis conducted by De Vivo et al

examining the application of the TPB in explaining exercise intentions during pregnancy, the authors concluded that women's attitudes towards exercise show the strongest association with performing the behaviour; however, perceived behavioural control and subjective norms were also very relevant[287]. The TPB also has some limitations relevant to stillbirth related behaviours. It does not take into consideration other factors that might be affecting intention such as emotional factors (fear, thread, mood changes)[288]. Further, environmental factors that are highly relevant during pregnancy are also not considered[288]. The TPB also assumes that individual decision-making is linear and does not consider changes over time. This is particularly problematic during pregnancy where women can be having very different experiences and perceptions of their pregnancy as it progresses[288]. Lastly, the timeframe between intention and behaviour change is not considered, which is also very relevant in pregnancy where changes need to be made in such a short period of time[288].

The Social Cognitive Theory (SCT) distinguishes between three constructs that interact with each other to influence behaviour: internal personal factors (cognitive, affective and biological events), behavioural patterns, and environmental events (which include imposed physical or socio-structural environment, selected environment and constructed environment)[289]. Self-efficacy and observational learning have also been identified in this theory as having a strong influence on behaviour change[288]. The uniqueness of this theory is that it emphasizes the importance of social influences and of internal and external reinforcements. The following examples illustrate the used of the SCT in the context of pregnancy to inform behaviour change interventions. Arefi et al (2022) recently examined an educational intervention based on the SCT that improved nutritional behaviour in a group of pregnant women[290]. The structures of the SCT were measured with questionnaires including questions related to "outcome value", "self-efficacy", "social support", "self-regulation", "mutual determinants", "knowledge" and "nutritional behaviour" administered pre and post intervention. The authors concluded from their Structural Equation Model that 39% of the variance in

nutritional behaviour during pregnancy could be explained using the SCT model[290], with "self-efficacy" having the highest predictor effect on nutritional behaviour[290]. The SCT also has some limitations relevant to behaviour change in stillbirth prevention, however. The theory assumes that changes in the environment automatically lead to changes in the person's behaviour[291], which might not be the case as other psychological factors related to the person's motivation such as unconscious motives, emotions or personality traits, and hormonal and biological processes, so relevant during pregnancy[292], will also have an effect on their behaviour change process[291]. The theory is based on the idea of interactions between the person, the behaviour and the environment, but there is not a clear idea as to how these factors affect behaviour or which factor is more influential[291].

All of the theories mentioned above have limitations that make them unsuitable to be used in the context of stillbirth prevention. These limitations are not specific to the context of stillbirth prevention but have been identified broadly in the context of behavioural science. Efforts have been made previously to develop a comprehensive robust theory of behaviour change that includes all the constructs relevant to behaviour change and that addresses some of the stated limitations. Mitchie et al. developed the Theoretical Domains Framework (TDF) for this purpose[293]. The TDF was the result of a project with the objective of identifying and agreeing on a set of key theoretical constructs for use in implementation science and intervention development[293]. Through a process of identifying theories and theoretical constructs, simplifying them, evaluating the constructs' importance and validating the construct list, the multidisciplinary study group identified 128 explanatory constructs from 33 psychological theories considered relevant for implementation purposes[293]. Hence, the refined TDF is composed of 14 domains covering 84 theoretical constructs: Knowledge, skills, social/professional role and identity, beliefs about capabilities, optimism, beliefs about consequences, reinforcement, intentions, goals, memory, attention and decision processes, environmental context and resources, social influence, emotions, behavioural regulation[293]. The fourteen domains of the Theoretical Domains Framework can be condensed into three main core components

to explain behaviour change: Capability, Opportunity and Motivation (COM-B model)[294].

The COM-B model is a framework for understanding behaviour and how to change behaviour[259]. This model was generated to develop a behaviour change design methodology informed by behaviour theory. The authors identified three main factors that are considered sufficient and necessary for behaviour change: the necessary skills, a strong intention to perform the behaviour, and no environmental constraints to make the behaviour possible[274]. These are operationalised as Capability, Motivation, and Opportunity respectively (See Table 1.3). In the COM-B model, the three components interact and influence each other, hence, altering one has the potential to alter the others. In the COM-B model, Capability is defined as "the individual's psychological and physical capacity to engage in the behaviour"[274] through, for example, knowledge or skills; and it is divided into Physical Capability and Psychological Capability. Opportunity is defined as "all the factors that lie outside the individual that make the behaviour possible or prompt it"[274]; and it is subdivided into social opportunity and physical opportunity (See table 1.3 for definitions and examples). Finally, Motivation is defined as "all those brain processes that energise and direct behaviour"[274] such as habitual processes, emotional responses, and analytical decision-making; and it is subdivided into reflexive motivation and automatic motivation.

Table 1. 3: COM-B elements and definitions obtained from www.behaviourchangewheel.com/[259]

COM-B Elements		Definitions	Examples
Capability	Physical	"Physical skills, strength"	Being able to walk 15 minutes despite feeling pregnancy related nausea
	Psychological	"Knowledge or psychological skills to engage in the necessary mental processes"	Being able to understand written antenatal education booklet
Opportunity	Physical	"Opportunity afforded by the environment involving time, resources, locations, cues, physical 'affordance'"	Free access to healthy eating classes for pregnant women
	Social	"Opportunity afforded by interpersonal influences, social cues and cultural norms that influence the way that we think about things, e.g. the words and concepts that make up our language"	Partner providing supporting pregnant woman's quitting behaviour by quitting as well
Motivation	Automatic	"Automatic processes involving emotional reactions, desires (wants and needs), impulses, inhibitions, drive states and reflex responses"	Anticipating an undesired pregnancy outcome
	Reflective	"Reflective processes involving plans (selfconscious intentions) and evaluations (beliefs about what is good and bad)"	Intention to attend all antenatal appointments

The TDF and the COM-B model represent potentially useful theories of behaviour that can be used in the context of stillbirth prevention because they incorporate internal factors, such as attitudes, co-occurring behaviours and co-founding factors as well as external factors[295]. The importance of the social context has been described in the literature in relation to behaviour change during pregnancy[262]. Moreover, it is also well established that the behaviours that have been associated with an increased risk of stillbirth often can co-occur (e.g., smoking and alcohol consumption[296], smoking and illicit drug use, illicit drug use and lack of attendance at antenatal care, substance use and physical inactivity)[296–298]. Therefore, utilising a theory that takes into account factors that influence behaviour such as the physical and social environment is important to understand behaviour change during pregnancy[299]. The COM-B model has not been widely tested in the context of stillbirth prevention. However, previous research mapping factors influencing dietary behaviour, physical activity, smoking, and alcohol use[262] during pregnancy to the elements of the COM-B has concluded that all its factors (capability, opportunity and motivation) have a role in directing behaviour[299]. This framework and its relevance for this work will be discussed in the following section.

1.9.3 The Behaviour Change Wheel (BCW)

The BCW is a systematic framework providing a methodology for designing behaviour change interventions. This methodology proposes the elements of the COM-B model as mechanisms of action (See Fig. 1.2). Despite having a vast amount of theory available and some existing frameworks for intervention design, interventions are usually designed without formal analysis of the behaviour they are trying to address, or the theoretically predicting mechanisms of actions associated with these behaviours[277]. Mitchie et al (2011) hypothesise that this is because the existing frameworks to date did not meet intervention designer's needs, as they either contained poorly defined constructs or did not produce enough level of detail to address what is effective in an intervention and what is not[277]. Hence, Michie and

colleagues developed a new framework for systematically developing behaviour change interventions, the BCW.

The BCW has at its core the elements of the COM-B Model (Green, see Figure 1.2). The red wheel illustrates the different Intervention Functions, and the grey wheel illustrates Policy Categories. These core elements will be discussed in detail in this section.

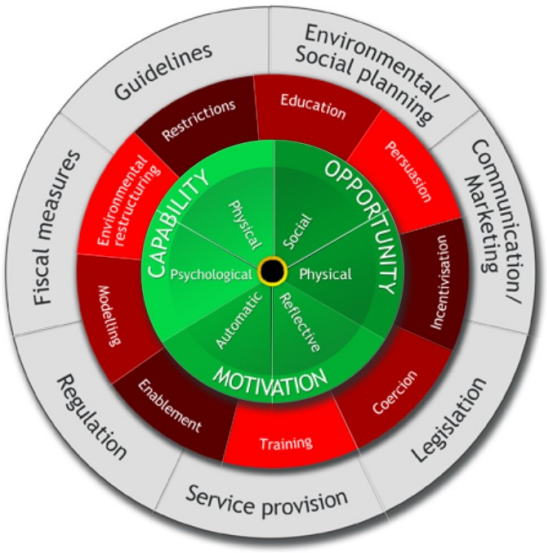


Figure 1. 2: The Behaviour Change Wheel obtained from Michie et al. (2014).[259]

The BCW provides intervention designers with a systematic process for intervention design composed of 3 main stages and 8 steps exposed in Table 1.4.

Table 1. 4: Stages and steps composing the Behaviour Change Wheel systematic process for intervention design.

Stage 1: understanding the target behaviour	Step 1: define the problem in behavioural terms.
	Step 2: Select the target behaviour.
	Step 3: Specify the target behaviour.
	Step 4: identify what needs to change.
Stage 2: Identify intervention options	Step 5: Identify intervention functions.
	Step 6: Identify policy categories.
Stage 3: Identify content and implementation options	Step 7: Identify behaviour change techniques (BCTs)
	Step 8: Identify the mode of delivery.

The objective of Stage 1 is to understand the problem in behavioural terms by being as specific as possible about what needs to change. This includes detailing what behaviours need to change to solve the problem, specifying the location of the behaviour, and the individual or group[259] ; it is recommended to gather information about the behaviour from as many sources as possible[259]. An important aspect of this stage is conducting a behavioural diagnosis, which can be defined as "the assessment of influences on the desired behaviour"[300] and includes taking into consideration individual, social, environmental, and medical factors that may facilitate or impede the behaviour[300]. Identifying barriers to behaviour change and mapping them to the components of the COM-B model is helpful to identify which elements of the model need to be addressed in intervention design. Though it has been noted that the broadness of the COM-B model could restrict the ability to test the model in a specific context, each domain of the TDF relates to a COM-B component. Thus, if a more detailed understanding of a behaviour is required, the TDF can be used to expand on the COM-B components identified as relevant in the analysis of the behaviour[259]. The

behavioural diagnosis performed using the COM-B components and/or the TDF is a key step to designing an intervention as it identifies potential levels of change[259]. In the case of stillbirth prevention, the selected behaviours relevant to an increased risk, and hence the focus of this Thesis were smoking, alcohol consumption, physical activity, healthy eating, regular attendance at antenatal care, and side sleeping; these behaviours were identified after a review of the literature as discussed in Chapter 2. These behaviours should be adopted throughout pregnancy by all pregnant women. However, the questions about what needs to change require more detailed behavioural analysis using the COM-B to be answered.

Once the relevant elements of the COM-B model are identified for the target behaviours, the next stage of the BCW involves selecting intervention functions and policy categories[259]. Intervention functions are defined as "broad intervention components which describe how an intervention aims to change behaviour"[259] and policy categories are defined as "broader population level strategies that could enable those interventions to occur"[259] (See Supplementary file 1.2). The components of the COM-B model are linked to the intervention functions, which are in turn linked to policy categories[277]. When mapping and identifying intervention functions and policy categories, multiple options may be identified. Thus it is recommended to utilise the APEASE criteria to select the most appropriate intervention functions and policy categories[259] based on the parameters of affordability, practicability, effectiveness and cost-effectiveness, acceptability, safety and equity[301].

The last stage of the BCW (Stage 3) involves identifying intervention content and implementation options. To do so, the first step is to select Behaviour Change Techniques (BCTs), which are defined as "active components of an intervention designed to change behaviour"; as such they represent the proposed mechanisms of change[259]. The BCT Taxonomy (BCTTv1) includes 93 hierarchically clustered and defined BCTs[302], which are linked to the different intervention functions in the BCW, facilitating identification of potentially useful BCTs from the preceding stages[259]. BCTs

can then be prioritised and selected using the APEASE criteria and translated into tangible intervention elements. For example, the BCT "Information about health consequences" could be translated into developing a leaflet with factual data about the increased risk of stillbirth when smoking during pregnancy[259].

The BCW framework has been used for behaviour change intervention design in the context of pregnancy. For example, Gould et al. (2017) successfully used the BCW to design an intervention to target smoking amongst Australian indigenous pregnant women[301]. Another example of an intervention designed using the BCT is the "stay-active" smartphone app developed by Smith et al (2022) to increase physical activity in pregnant women[303]. The authors of both interventions report that utilising the BCW provided them with a systematic approach that facilitated the process. The process described in the studies published by both authors outlining their use of the BCW to inform the development of their interventions led to the design of two different interventions that are now being tested through feasibility studies.[304,305]. Hence, there is evidence in the literature that the BCW is a methodology suitable for intervention design applicable to pregnancy.

1.10 Summary

Stillbirth is a devastating pregnancy outcome that has implications for women's physical and mental health, as well has to their partners, other children, and future pregnancies. Stillbirth also affects the healthcare staff involved in care of bereaved parents, and it also supposes an economic burden for families and for the health system. Some issues in relation to the different definitions and data capture that are interfering with prevention efforts are captured in this section, with a rationale to change the current definition in Ireland to benefit parents and babies. The challenges associated with preventing stillbirth are different for low-, middle-, and high-income countries. Whereas low-, and middle- income countries need to focus their efforts in improving intrapartum care and preventing infections, high-income countries should focus on risk identification and enhancing antenatal care. The risk factors for stillbirth

are quite diverse, including sociodemographic factors, medical factors, obstetric history related factors, placental factors, and behavioural and lifestyle factors. The focus of this Thesis is on the latter given their importance in high-income countries.

While the BCW provides a promising approach to informing design of behaviour change interventions that target behaviours associated with increased risk of stillbirth, to date, no intervention has been designed using the BCW and adopting a multi-target approach that might incorporate all of the relevant behaviours. Evidence from public health initiatives implemented in high-income countries provide evidence that further reduction in stillbirth rates is possible in Ireland. Hence, the main objective of this Thesis was to develop a comprehensive evidence base to inform the future development of a behaviour change intervention relevant to all behaviours associated with an increased risk of stillbirth. It is important to gather enough evidence using different sources that will inform all stages of intervention development. The studies included in this Thesis provide a comprehensive evidence base that informs the early stages of intervention design as guided by the BCW. The exploration and examination of women's experiences, perceptions and attitudes to behavioural stillbirth risk factors, as well as examination of existing interventions and information provision, provides an evidence base upon which future work can build to develop an intervention that addresses the behavioural risk factors for stillbirth. The evidence base built through the studies forming this Thesis enhances the understanding of such behavioural risk factors and discusses what has been conducted to date to address the behavioural risk factors for stillbirth.

1.11 Stakeholder engagement in this research

The studies within this Thesis have been conducted in the context of the Pregnancy Loss Research Group (PLRG), based at the Cork University Maternity Hospital and led by Prof. Keelin O'Donoghue. The PLRG is a multidisciplinary team of health and social care professionals and researchers that conducts research in all types of pregnancy and infant loss.

The PLRG is composed of professionals from different disciplines, including midwifery, medicine, social work, epidemiology and psychology, as well as parent advocates. The group meets once a month, in those meetings, the work of all of the members is discussed, and they are given the opportunity to request advice or support to other members of the group to support their own research projects.

Participating in these meetings provided this project with much needed informal stakeholder input to make sure the work was always informed by best research and clinical practices. Some examples of stakeholder input from the members of the PRLG can be found in Chapter 3, where members reviewed the list of included websites and suggested additional ones, and in Chapter 7, where members input was crucial in informing the best way to approach recruitment of postpartum women on the postnatal wards.

1.12 Thesis aims and objectives

As stated, the overall aim of this Thesis was to build the evidence base to inform the development of a behaviour change intervention for stillbirth. The specific objectives for each chapter are presented in Table 1.5.

Table 1. 5: Aims and objectives of each individual chapter.

Thesis Chapter	Objective(s)
Chapter 2 - Modifiable risk factors for stillbirth: a literature review	• To explore and examine the available evidence in relation to behavioural risk factors for stillbirth. • To define the problem and select target behaviours.
Chapter 3 - Stillbirth and risk factors: an evaluation of Irish and UK websites	• To assess whether the current online resources for pregnant women are useful to obtain information about stillbirth and behavioural risk factors.
Chapter 4 - Facilitators and barriers to seeking and engaging with antenatal care in high-income countries: a meta-synthesis of qualitative research.	• To assess the literature in order to identify barriers and facilitators to women's behaviour change regarding attendance and engaging with antenatal care from the pregnant women's perspective.
Chapter 5 - Facilitators and barriers to substance-free pregnancies in high-income countries: A meta-synthesis of qualitative research	• To assess the literature in order to identify barriers and facilitators to women's behaviour change regarding substance use from the pregnant women's perspective.

Chapter 6 - Facilitators and barriers influencing weight management behaviours during pregnancy: a meta-synthesis of qualitative research	• To assess the literature in order to identify barriers and facilitators to women's behaviour change regarding engaging with weight management behaviours from the pregnant women's perspective.
Chapter 7 - Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth	• To explore women's experiences of behaviour change during pregnancy & awareness regarding stillbirth and associated risk factors.
Chapter 8 - A systematic review of behaviour change techniques used in the context of stillbirth prevention	• To identify the behaviour change techniques used to date in stillbirth prevention interventions.

In addition to the studies presented in the chapters of this Thesis, another study was conducted in collaboration with a member of the Pregnancy Loss Research Group (PLRG), which was relevant to the general purpose of this Thesis. Even though I was not lead author on this study, I was heavily involved through the whole process. Hence, these findings are incorporated in the discussion section. This study is entitled "Exploring Healthcare professionals experiences when communicating with pregnant women about stillbirth" and was conducted with the objective of exploring how HCP communicate about stillbirth and behavioural risk factors for stillbirth with women, and to gain understanding regarding HCP's perceived barriers to addressing this issue. From now on, this study will be referred as "Survey study". More information about this study including the study protocol, the utilised survey and the manuscript in preparation can be found in Supplementary file 1.1.

This Thesis is hence composed of nine chapters, one introductory chapter, eight studies, and a discussion chapter that brings together the findings of all the research studies discussed in the different chapters.

Chapter 2 - Modifiable risk factors for stillbirth: a literature review

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Chapter 2 - Modifiable risk factors for stillbirth: a literature review

2.1 Abstract

A stillbirth is defined as an infant born weighing 500grammes and/or more or at a gestational age of 24 weeks who shows no signs of life. Having a stillborn baby has a wide range of consequences that can affect parents, family and the healthcare professionals involved. Several risk factors have been associated with an increased risk of stillbirth: including maternal medical factors, maternal characteristics, fetal factors, sociodemographic factors and behavioral factors. The aim of this work is to review the literature on risk factors that have a behavioral component. The main behaviors modulating the risk of stillbirth that have been more widely studied in the literature include use of substances (smoking, alcohol, illicit drugs and medical drugs), weight management, attendance at antenatal care and sleeping position. There is evidence in the literature that supports that all those behaviors have an impact on the risk of stillbirth, especially in the cases of smoking and drugs consumption during the pregnancy. Hence, more research is needed to establish interventions targeting these behaviors as preventive measures to reduce the risk of adverse obstetric outcomes.

Keywords: stillbirth; modifiable risk factors; pregnant women; health behavior

2.2 Introduction

The loss of an unborn child is one of the most devastating life events that parents can face during their lifetime. Having a stillborn baby in a family has a wide range of consequences that can affect the parents, the rest of the family, and even the healthcare professionals involved.

According to the Irish Stillbirth Registration Act (1994), a stillbirth is defined as an infant born weighing 500grammes or more or at a gestational age of at least 24 weeks who shows no signs of life. However, stillbirth is defined differently around the world depending on weeks of gestation and weight; there is no globally accepted definition of stillbirth versus second trimester miscarriage (see Supplementary File 2.1). Therefore, the challenge of comparing birth rates and associated risk factors is greater[10,306].

Stillbirths are associated with 2.6 million deaths each year all over the world[307]. Although rates of stillbirth for high-income countries are lower than in middle or low-income countries, 1 in 200 babies will be stillborn and this rate has remained steady since 2000[308]. According to the last report published by the National Perinatal Epidemiology Centre in 2016, the rate of stillbirth at >24 weeks gestation or a birthweight of >500grammes was 3.9 per 1,000 births in Ireland[254].

There is a wide range of risk factors that have been studied in relation to stillbirth, including maternal medical factors, maternal characteristics, fetal factors, sociodemographic factors and behavioral factors. The purpose of this review is to focus on behavioral factors as they consist of different habits, lifestyles or choices that women make; these are smoking, alcohol and drug consumption, weight management, attendance at antenatal care and sleeping positions. Establishing which risk factors are modifiable and understanding how they can be changed can help develop prevention strategies to empower women to reduce their own risks of experiencing a stillbirth. This review also gives an overview of some of the evidence available and highlights areas of controversy, serving as a tool for researchers to understand where to focus their efforts (see Table 2.1). Although this area has been

a matter of concern for some time among healthcare professionals, the well-informed groups are very specialized. This review can serve as an informative document for a wider audience, including practitioners in primary care and pregnant women themselves.

To conduct this review, a non-systematic search was performed using different search engines (Google Scholar) and a database (Pubmed) The following keywords *"stillbirth", "risk factors", "behavioural risk factors", "smoking", "alcohol", "illicit drugs", "medication", "over the counter drugs", "supplements", "high BMI", "overweight", "obesity", "sleep position", "sleep habits", "attendance antenatal care", "prenatal care", "pregnancy outcomes",* were used in isolation and in different combinations, focusing on one risk factor at a time.

Relevant articles pertaining to the different selected risk factors were identified and organized using Mendeley. However, the search continued as the work progressed. Hence, the identification and revision of papers facilitated the identification of other relevant studies. Although there were no publication date restrictions, most recent publications were prioritized to make sure we were including the most up-to-date research and findings, enhancing the overall quality of the review. The review is organized by sections, each one corresponding to one of the above-mentioned risk factors. Each section aims to give an overview of the evidence in relation to specific risk factors for stillbirth. Evidence regarding early miscarriage or other types of perinatal loss are not included, as it is not within the scope of this review.

Table 2. 1: Summary of the evidence exposed in this review

Associated with stillbirth/ Not recommended	**No association/ Recommended**	**Conflicting evidence**
Smoking	**Prescription drugs**	**Alcohol and illicit drug use**
Active smoking	Prescription nausea medication	Small amounts of alcohol
Passive smoking	Prescription pain medication	
Ever smoking		**Prescription drugs**
Highest influence in first trimester	**Herbal and dietary supplements**	Acetaminophen
Higher amounts increase the risk	Iron folic acid	Aspirin
		Antihistamines
Alcohol and illicit drug use	**Sleep position**	Selective Serotonin Reuptake Inhibitors
High amounts increase the risk	Sleeping on the left side	
Use of any illicit drugs	Maternal snoring	**Herbal and dietary supplements**
	Daytime sleepiness	Herbal remedies (ginger, cranberry, raspberry, chamomile, peppermint)
Prescription drugs		
Migraine medication on second trimester.	**Attendance to antenatal care**	Vitamin C
Ultrasound exposure on first and second trimester	At least four antenatal visits during pregnancy	Vitamin D
Naproxen		**Attendance to antenatal care**
		Gestational age at first booking
BMI		
Unbalanced energy and protein dietary supplementation		**Sleep position**
		Sleeping less than 6h per night

BMI over 25 Severity of overweight increases the risk	
Sleep position	
Sleeping on the back	
Sleeping on the right side	
Supine sleep position	
Not getting up to the toilet or just getting up once during the last night of pregnancy	
Regular daytime sleep	
Attendance to antenatal care	
Attending 50% less than the recommended visits. Decrease in the number of visits increases the risk	

2.3 Findings

2.3.1 Smoking

Smoking is one of the risk factors associated with stillbirth that has been widely studied and has been associated with an increased risk of stillbirth (see Table 2.2). Hyland et al. found suffering a stillbirth is 1.44 times more likely for women who were ever-smokers than for women who were never-smokers. Furthermore, these authors also concluded that never-smoking women exposed to passive smoking showed a 22% increase in the odds of experiencing a stillbirth compared to never-smoking women not exposed to passive smoking [141]. A meta-analysis conducted by Marufu et al., (2015) found that smoking during pregnancy was associated with a 47% increase in the odds of stillbirth. Accordingly, The Lancet published a different meta-analysis showing that smoking during pregnancy was associated with a 36% increase in the odds of stillbirth and this risk was bigger in smoking pregnant women of 40 years or older [87].

Varner et al. conducted a study to explore the effects of the dose used on the risk of stillbirth. They concluded after their case-control study that increasing amounts of self-reported smoking were associated with an increase in the incidence of stillbirth, being 1.77 times more likely for women who reported smoking 1 to 9 cigarettes per day and 2.17 times more likely for women who reported smoking more than 10 cigarettes per day[139].

Some researchers have tried to study the effect of time of smoking exposure on stillbirth. Dodds and colleages concluded in their study that the effect of smoking on stillbirth was similar in women who quit smoking before 16 weeks of pregnancy and women who continued to smoke after 16 weeks[140]. The authors reported that these findings give support to the hypothesis that the first trimester of a pregnancy might be the more susceptible to the influence of smoking in regard to stillbirth, and that cessation pre-pregnancy would be ideal.

Table 2. 2: Characteristics and results from the studies related to smoking

Author	Type of study and population	Objectives	Main outcomes	Results
Hyland et al. (2014)	Cross sectional study 80762 women	Study association between tobacco exposure and pregnancy outcomes	Ever active smokers	OR 1.44 (95% CI 1.20 to 1.73)
			Never active smoker exposed to passive smoking	OR 1.22 (95% CI 1.02 to 1.47)
Marufu et al. (2015)	Meta-analysis including 34 studies	Study the association between maternal smoking and risk of stillbirth	Smoking during pregnancy	OR 1.47, (95% CI 1.37 – 1.57, p<0.0001)
Flenady et al. (2011)	Meta-analysis including 96 studies	Identify priority areas of stillbirth prevention	Smoking during pregnancy	aOR 1.36 (95%CI 1.27–1.46)
Varner et al. (2014)	Case control study 663 stillbirths 1932 live births	Compare illicit drug use and smoking in pregnancies with and without stillbirth	Smoking 1-9 cigarettes a day	OR1.77 (95% CI 1.13 – 2.80)
			Smoking ≥10 cigarettes a day	OR 2.17 (95%CI 1.25 – 3.78)
Dodds et al., (2006)	Population based case control study 105 stillbirth cases and 389 live-births	Identify risk factors for stillbirth and explore possible causes based on time of exposure	Quitting before 16 weeks of pregnancy	HR 2.6, 95%CI 1.5 – 5.8
			Quitting after 16 weeks of pregnancy	HR 2.2, 95%CI 1.2 – 4.1

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2.3.2 Alcohol and illicit drug use

Alcohol and illicit drug abuse have also been studied in association with the risk of stillbirth (See Table 2.3). Ethanol is the most commonly abused substance during pregnancy and it is a recognized public health problem[309]. Globally, about 10% of women consume alcohol during their pregnancy[143]. Aliyu et al. (2008) concluded in their study that more than 3% (N = 120) of cases of stillbirth were associated with mothers that consumed alcohol during pregnancy, with a stillbirth rate of 8.3 per 1000 births versus a rate of 5.3 per 1000 births in the abstinent reference group.

Some researchers have tried to establish a correlation between the amount of drinks consumed and the risk of stillbirth by studying the maximum amount considered safe. Henderson et al. (2007) found in their systematic review and meta-analysis that most of the studies relating alcohol consumption with stillbirth were subject to recall bias; only one article was except from bias as it measured alcohol consumption with a validated questionnaire. These authors concluded that low-moderate levels of consumption were associated with slightly higher rate of stillbirth, but the association was not statistically significant. Similarly, Andersen et al. (2012) found that the adjusted risk of stillbirth was higher as the amount of drinks consumed per week increased, however, their results were not statistically significant.

Illicit drug consumption also has an impact on pregnancy outcomes (See Table 2.3). The drugs that have been more commonly found to be used during pregnancy besides tobacco and ethanol are cocaine, amphetamines, opioids, marijuana, hallucinogens and toluene-based solvents [143]. Varner and colleagues concluded in their study that a positive toxicology test for any drug increased the risk of stillbirth almost a 94%, and this risk was more than 3 times higher in women who also reported having used illicit drugs. The most common drug that these authors found was Tetrahydrocannabinolic Acid (THCA), which is the active component of cannabis, and was significantly associated with a 2.34 times higher risk of stillbirth [139].

Table 2. 3: Characteristics and results from the studies about alcohol consumption.

Author	Type of study and population	Objectives	Main outcomes	Results
Aliyu et al. (2008)	Retrospective study 65000 pregnancies	Study the association between maternal alcohol consumption and early stillbirth	Alcohol consumption during pregnancy	aHR 1.4 (95%CI 1.2-1.7)
Andersen et al. (2012)	Cohort study 92719 participants	Assess the risk of fetal death according to maternal alcohol consumption	½ to 1 drink per week	HR 0.90 (95%CI 0.73-1.12)
			2 to 3½ drinks per week	HR 0.88 (95%CI 0.63-1.24)
			4 or more drinks	HR 1.20(95%CI 0.76-2.18)
Varner et al. (2014)	Case control study 663 stillbirths 1932 live births	Compare illicit drug use and smoking in pregnancies with and without stillbirth	Positive toxicology test	OR 1.94 (95%CI 1.16-3.27)
			Women who reported using illicit drugs	OR 3.30 (95% CI 1.54 - 7.03)
			Consuming cannabis	OR 2.34 (95% CI 1.13 - 4.81)

2.3.3 Use of medical drugs and supplements

Prescription and over-the-counter drugs

Some authors have also studied the effects of using medically prescribed drugs, over-the-counter drugs and food or herbal supplements on the outcomes of pregnancy (See Table 2.4).

A study conducted in Europe, North and South America and Australia reported that almost 80% of pregnant women used at least one drug during their pregnancy, and around 60-70% have reported using at least one over-the-counter medication [152]. Some of the most commonly drugs used by pregnant women are antihistamines, decongestants, expectorants and antitussives, analgesics and antipyretics, topical creams, antacids and antidiarrheal agents [154].

Antihistamines and antiemetics are principally used during pregnancy for the treatment of nausea and vomiting and approximately 15% of pregnant women use them[156]. Following a meta-analysis, Etwel et al. concluded that the risk of stillbirth was not higher for women who had used HI antihistaminic agents during the first trimester. However, the authors did not rule out a possible association between both factors due to a lack of information on the cause of death of the stillborn infants[157].

Other common drugs used during pregnancy are analgesics such as acetaminophen (paracetamol), ibuprofen, naproxen and others. More than 60% of women use acetaminophen during pregnancy, and around half of them do so in the first trimester[311]. However, some studies have associated the use of ibuprofen, aspirin, and naproxen with a higher risk of spontaneous miscarriage and other fetal anomalies, especially when used around the time of conception[312] (Hernandez et al., 2012).

According to a study conducted in Canada, around 18% of pregnant women had used psychotropic drugs (antidepressants, anxiolytic/sedative-hypnotics, antiepileptics, antipsychotics, lithium) before, during or after pregnancy, and this tendency has increased significantly over the years. Depression is one of the most common

conditions with a prevalence among 7% to 19% in developed countries (Leong et al., 2017).

A population-based cohort study in all Nordic countries first showed that the likelihood of experiencing a stillbirth for women who had been exposed to Selective Serotonin Reuptake Inhibitors (SSRIs) was 25% higher than those who had not. However, when adjusting the data for maternal characteristics, country and year of birth, exposure to SSRIs was no longer significantly associated with higher risk of stillbirth. The authors concluded that the higher rates of stillbirth were likely to be related to the severity of the psychiatric diseases and other risk factors rather than to the exposure to the SSRIs themselves [169].

Table 2. 4: Characteristics and results from the studies about prescription drugs and over-the-counter drugs.

Author	Type of study and population	Objectives	Main outcomes	Results
Pastore et al., (1999)	Case control study 332 stillbirths and infants deaths within 24h after birth 357 control livebirths	Study the association between stillbirth and 14 medical exposures	Migraine medication 2nd trimester	aRR 1.6 (95% CI 1.1-2.3)
Etwel et al. (2017)	Systematic review and meta-analysis 37 studies included	Determine whether exposure to antihistaminic during the first trimester is associated with negative obstetrics outcomes	Stillbirth	OR 1.23 (95%CI 0.48-3.18)
Stephansson et al., (2013)	Population based cohort study 1633877 singleton births, 6054 stillbirths; 3609, neonatal deaths; and 1578 post-neonatal deaths	Study the influence of exposure to SSRIs on the risk of stillbirth	Exposure to SSRI	aOR 1.17 (95%CI 0.96-1.41, P=0.12)

Herbal and Dietary Supplements

Herbal remedies are also widely used during pregnancy for different purposes such as palliating morning sickness, urinary tract infections, nausea or vomiting and hence some authors have tried to establish their safety during pregnancy (See Table 2.5). The amount of pregnant women using herbal supplements ranges from 7% to 55% depending on different geographical, social and cultural settings and ethnic groups[172].

Smeriglio and colleagues wrote a review to establish the most used herbal remedies during pregnancy and their potential side effects. The authors concluded that few clinical trials have been performed to assess the safety and efficacy of these remedies. According to the findings of these authors, the most common herbs used during pregnancy are ginger, cranberry, raspberry, chamomile, peppermint and Echinacea [174]. Although some of these herbs have been classified as safe by the European Medicines Agency, not enough data about their use in pregnancy and lactation is available and they cannot replace medical treatments[173]. Some of the studies included in their review criticise the use of herbal medicines to treat conditions such as urinary tract infection, as this infection, without the appropriate treatment, could lead to bigger complications and adverse pregnancy outcomes such as stillbirth.

The use of daily multiple micronutrients on the risk of stillbirth has also been studied. Smith et al. (2017) conducted a meta-analysis including 17 randomized controlled trials on the effects of maternal multiple micronutrients supplementations on stillbirth (and other birth outcomes, in low-income and middle-income countries. The authors compared the use of micronutrient supplements containing iron-folic acid against using iron-folic acid only. The results show that the use of micronutrient supplements did not increase the risk of stillbirth (See Table 2.5).

Table 2. 5: Characteristics and results from the studies about herbal and alimentary supplements.

Author	Type of study and population	Objectives	Main outcomes	Results
Holst et al. (2010)	Literature review	Review the literature on safety and efficacy of the most common used herbal remedies	Safety of different herbal remedies	Not enough data on efficacy of herbal remedies during pregnancy and lactation
Smith et al. (2017)	Meta-analysis 31 Randomised control trials	Identify modifiers on the effect of micronutrient supplements on stillbirth and other birth outcomes	Odds for stillbirth	RR 0.92 (95%CI 0.86-0.99)

The World Health Organization recommends balanced energy and protein dietary supplementation for pregnant women in undernourished populations to reduce the risk of stillbirth in their "WHO recommendations on antenatal care for positive pregnancy experience" report. Use of daily oral iron and folic acid with 30mg to 60mg of elemental iron[b] and 400 g (0.4 mg) of folic acid[c] to prevent maternal anemia, puerperal sepsis and low birthweight is also recommended. On the other hand, the WHO recommends against using zinc supplements, multiple micronutrient supplements, vitamin B6 (pyridoxine) supplements, Vitamin E and C supplements and Vitamin D supplements [204]. However, there are discrepancies among the recommendations made by the different health authorities regarding this issue (See Table 2.6).

Table 2. 6: Comparison of the recommendations of four different health authorities.

	Recommended	Not recommended
World Health Organization	▪ Folic acid supplements	▪ Zinc supplements ▪ Micronutrients supplements ▪ Vitamin B6 ▪ Vitamin E ▪ Vitamin C ▪ Vitamin D
National Institute of Health and Care Excellence (NICE)	▪ Folic acid ▪ Vitamin C ▪ Vitamin D	▪ Vitamin A ▪ Iron Supplements
Royal College of Obstetricians and Gynaecologists, London, UK	▪ Folic acid ▪ Vitamin C ▪ Vitamin D	▪ Vitamin A ▪ Vitamin E ▪ Vitamin B (other than folic acid)
The American College of Obstetricians and Gynecologists	▪ Folic acid ▪ Vitamin A ▪ Vitamin C ▪ Vitamin D ▪ Iron	▪ N/A

2.3.4 Maternal weight

According to the World Health Organization, obesity has almost tripled since 1975. In 2016, 39% of adults were overweight, and 13% were obese[177]. Although, the prevalence of overweight and obesity during pregnancy is very hard to establish, an increasing trend similar to the one in the general population has been observed[179].

Obesity during pregnancy can cause gestational diabetes and gestational hypertensive disorders, two conditions that have been associated with stillbirth, but obesity is also considered an independent risk factor for stillbirth (See Table 2.7)[1]. However, most women who try to conceive are not aware of these risks[178].

Chu et al. (2007) found in their systematic review and meta-analysis that overweight women were 47% more likely to experience a stillbirth than normal weight women, whereas in the case of obese women, the risk of stillbirth was 2.07 times higher compared with the normal weight group.

When examining the risk of stillbirth Yao et al. (2018) found that the hazard ratio (HR) increased significantly as overweight increased in severity but also as the pregnancy progressed (See Table 2.7). The same authors concluded in previous work that almost 20% of the stillbirths included in their study were associated with obesity[187].

Table 2. 7: Characteristics and results from the studies about overweight and obesity.

Author	Type of study and population	Objectives	Main outcome	Results	
Chu et al. (2007)	Meta-analysis 6 studies	Summarize the available epidemiological evidence on the relationship between weight and stillbirth	Overweight group	OR 1.47 (95%CI 1.08-1.94)	
			Obese group	OR 2.07 (95%CI 1.59-2.74)	
Yao et al. (2018)	Retrospective cohort study 3279846 births included	Examine the risk of stillbirth with increasing obesity severity and progression of the pregnancy	Overweight group	33-36 weeks	HR 1.41 (95%CI 1.24-1.59)
				37-39 weeks	HR 1.36 (95%CI 1.19 – 1.54)
				40-42 weeks	HR 1.42 (95%CI 1.08-1.86)
			Class I Obesity	33-36 weeks	HR 1.52 (95%CI 1.31-1.75)
				37-39 weeks	HR 1.76 (95%CI 1.52-2.04)
				40-42 weeks	HR 1.95 (95%CI 1.43-2.66)
			Class II Obesity	33-36 weeks	HR 1.62 (95%CI 1.35-1.96)
				37-39 weeks	HR 2.36 (95%CI 1.98-2.82)
				40-42 weeks	HR 2.15 (95%CI 1.41-3.29)
			Class III Obesity	33-36 weeks	HR 1.61 (95%CI1.28-2.03)
				37-39 weeks	HR 2.26 (95%CI 1.80-2.83)
				40-42 weeks	HR 2.53 (95%CI 1.50-4.27)

2.3.5 Sleeping habits

One of the maternal everyday habits that have been investigated for having an effect on the risk of stillbirth are the sleeping practices of pregnant women (See Table 2.8). Stacey and Thompson (2011) conducted a prospective population-based control-study in New Zealand assessing maternal snoring, daytime sleepiness, and sleep position at the time of going to sleep and on waking (left side, right side, back or other) and the risk of late stillbirth. Snoring, daytime sleepiness and hours of sleep were not associated with higher risk of stillbirth when adjusting for co-founders. However, women who slept during the day regularly had 2.04 times higher risk of having a stillborn baby. Further, not getting up or getting up just once to the toilet during the last night of pregnancy increased the risk of stillbirth 2.42 times

The researchers also found differences in the risk of late stillbirth depending on the maternal sleep position both at time of going to sleep and at time of waking up, especially during the last night of the pregnancy, but also during the preceding month. The risk was higher for those women who did not go to sleep nor woke up on their left side and the authors determined sleeping on the back the least safe position [195]. Similar results were found in a different study where the authors also concluded that the risk was higher for term compared to preterm stillbirths[198]. This is in concordance with the results from other studies that concluded that women who had a late stillbirth were 2.3 times more likely to have reported a supine sleeping position on the night before their stillbirth[194,315].

Warland et al. (2018) undertook a systematic review and meta-analysis exploring the association between maternal sleep during pregnancy and poor fetal outcomes. The authors looked at four different areas of maternal sleeping: disordered breathing, sleep duration, sleep quality and sleep position. According to the authors, all these aspects of maternal sleeping can be associated with different fetal outcomes such as fetal growth and preterm birth. However, with regard to stillbirth, studies showed that women who sleep less than 6 hours or more than 8 hours per night in late pregnancy have more than an 80% increase in the odds of experiencing a stillbirth

(See Table 2.8). Furthermore, in accordance with Stacey and Thompson, the attributable risk of supine going-to-sleep position in the studies they included in their meta-analysis ranged between 4% and 37%.

One hypothesis that explains these results associates the mother's sleeping position with cardiac outputs and fetal oxygenation. The pressured exerted by the enlarged uterus over the vena cava and the aorta when lying in a supine or right lateral position could produce a decrease in uterine blood flow and subsequently lead to fetal hypoxia[198,199].

Table 2. 8: Characteristics and results from the studies about sleep habits.

Author	Type of study and population	Objective	Main outcomes	Results
Stacey et al. (2011)	Population based control study 155 late stillbirths 310 controls	Determine whether certain sleeping habits are associated with stillbirth	Snoring	aOR 1.12 (95%CI 0.75-1.67)
			Daytime sleepiness	Epworth sleepiness score 5.9(4.1) for cases and 5.6(3.8) for controls (P=0.51)
			Hours of sleep	Less than 6h aOR 1.81, (95% CI 1.14 to 2.88)
				More than 8h (aOR 1.83 95%CI 1.14-2.94)
			Regular daytime sleep	aOR2.04 (95%CI 1.26 – 3.30)
			Getting up one or zero times last night of pregnancy	aOR 2.42(95%CI 1.46 – 4.00)
			Right side sleeping position	aOR 1.74(95%CI 0.98-3.01)
			Back side sleeping position	aOR 2.54 (1.04 – 6.18)
McCowan et al. (2017)	Multicentre case-control study 164 late stillbirths 569 controls	Test the hypothesis that maternal non-left and supine going-to-sleep position is a risk factors for stillbirth	Stillbirth at term	aOR 10.26 (3.00 to 35.04
			Stillbirths pre-term	aOR 3.12 (0.97 to 10.05)

Warland et al., (2018)	Systematic review and meta-analysis	Summarize current literature on maternal sleep including sleep disordered breathing, sleep quality, sleep duration and supine sleep position	Less than 6h sleep per night	aOR 1.81 (95%CI 1.14 – 2.88)	
			More than 8h sleep per night	aOR 1.83 (95%CI 1.14 – 2.94)	
			Sleep position	Heazell et al. (2018)	aOR 2.31 (95%CI 1.04-5.11)
				McCowan et al. (2017)	aOR 3.67 (95%CI 1.74 – 7.78)
				Owusu et al. (2013)	aOR 8.0 (95%CI 1.5-43.2)
				Gordon et al. (2015)	aOR 6.26 (1.2 – 34.0)
				Lakshmi et al., (2017)	aOR 2.95 (95%CI 1.5-5.8)

2.3.6 Attendance at antenatal care

The World Health Organization currently advises women to attend at least four antenatal visits during their pregnancy[204], when there are no complications. Antenatal care gives healthcare professionals an opportunity to educate women and offer health information, and it is also an opportunity for them to monitor the development of the pregnancy[319].

Stacey et al. (2012) conducted a study to examine the role of antenatal care in the prevention of stillbirth in high-income countries. The authors compared cases of women who experienced a late stillbirth with ongoing pregnancies at the same gestational week in which the stillbirth occurred. The authors concluded that gestational age at booking with a prenatal care provider was not associated with higher risk of late stillbirth (See Table 2.9). However, women who attended less than 50% of the recommended visits, were almost 3 times more likely to suffer a late stillbirth; the authors also found a significant relationship between the decrease in the number of visits attended and the increase in the risk of stillbirth. The researchers also concluded that small-for-gestational-age fetuses were almost 10 times more likely to be stillborn if they were not identified prior to birth than those who had been identified. This means that identifying small-for-gestational-age fetuses could reduce the risk of stillbirth, reinforcing the importance of regular antenatal care attendance[206].

Table 2. 9: Characteristics and results from the studies about antenatal care.

Author	Type of study and population	Objective	Main outcomes	Results	
Stacey et al. (2012)	Cohort study 215 stillbirth cases 310 control	Asses the relationship between antenatal care and risk of stillbirth	Gestation age at booking	10-20 weeks gestation	aOR 0.78 (95%CI 0.46-1.30)
				More than 20 weeks gestation	aOR 0.63 (95%CI 0.28-1.39)
			Attending less than 50% of recommended visits	aOR 2.68 (95%CI 1.04 – 6.90)	

2.4 Conclusion

Risk factors are crucial in prediction and prevention of stillbirth and raising awareness about them is essential to ensure women are making informed decisions. Smoking, substance abuse, non-attendance to antenatal care, and sleeping positions have been established as behavioral risk factors for stillbirth and should be taken into consideration in every pregnancy. Behavioral risk factors can be reduced or eliminated through antenatal interventions as a preventive measure before pregnancy, or if necessary, can also be addressed in ongoing pregnancies [306]. A study published in the Lancet showed that some behavioral risks associated with stillbirth such as smoking, substance abuse and maternal obesity, were underestimated by healthcare professionals, and also the authors identified a lack of awareness among the wider community [307]. This work summarizes the information regarding the risk factors that can be modified by the mothers, giving healthcare practitioners an opportunity to empower women.

Some of the results included in this review are contradictory, especially the older studies, and should be considered with caution. Some of the limitations found when revising the literature had to do with the difficulties in establishing proper comparisons due to differences in the definitions of stillbirth used. Further, different studies use various methodologies and measure different outcomes.

Another limitation of this review is related to several ethical issues to consider when conducting clinical research with pregnant women. Traditionally pregnant women have been excluded from research as this population are considered vulnerable that need to be protected from the potential risks of participating in research [320]. Hence, given pregnant women's exclusion from research trials, there is a paucity of studies which are eligible for inclusion in a systematic review on behavior change interventions targeting these risk factors,

Despite the difficulties mentioned above, it seems clear that there is evidence in the literature supporting the fact that certain maternal behaviors have an impact on the risk of stillbirth.

Smoking and abuse of illicit drugs seem to have the highest impact. In the case of medication and herbal or food supplements, the evidence is not clear, but there is a wide consensus in the literature that more clinical trials are needed to establish the safety and efficiency of their use during pregnancy. Maternal weight is another factor that needs to be taken into consideration when a woman is planning a pregnancy. As seen in the literature, it is not only an independent risk factor, but it can also lead to other risks such as gestational hypertension or diabetes [321]. With regard to maternal sleeping habits, it seems that avoiding sleeping on the right side might be a simple measure to reduce the risk of stillbirth, being possibly the most easily modifiable behavior among the different ones included in this review. On the other hand, it seems clear that attendance to antenatal care visit is very relevant in the prevention of stillbirth, as it is the only way healthcare practitioners can monitor pregnancies to detect other risk factors and diagnose pregnancy complications.

Despite some reviews and trials being currently conducted, not enough data is available on the efficacy of the preventive interventions established in regards with stillbirth specifically, with this research focusing mostly on sleep position. More research is needed in this area to establish the efficiency of interventions targeting these behaviors among pregnant women as preventive measures with the aim of reducing the risk of stillbirth and other adverse obstetric outcomes.2.5 Inter-Chapter Synopses

Chapter 2 aimed to identify the modifiable/behavioural risk factors for stillbirth with the strongest evidence in the literature. The findings of Chapter 2 suggest that modifiable risk factors such as smoking, alcohol consumption, maternal age, maternal obesity, and inadequate prenatal care were associated with an increased risk of stillbirth. Chapter 2 emphasizes the need for targeted interventions to address these modifiable risk factors to reduce the incidence of stillbirths. The findings obtain from this study regarding the selected behaviours informed the rest of the studies in this Thesis. Chapter 3 aimed to evaluate the information available on Irish and UK websites regarding stillbirth and the risk factors associated to stillbirth identified in Chapter 2. Before confucting the study in Chapter 3, it was well known that the public awareness around stillbirth in Ireland was limited[258], and that the internet is an acceptable resource for people to access health information[322,323].

Chapter 3 - Stillbirth and risk factors: an evaluation of Irish and UK websites

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Chapter 3 - Stillbirth and risk factors: an evaluation of Irish and UK websites

3.1 Abstract

Background

Research indicates that the internet is used to source health-related information, including pregnancy-related information. The aim of this study was to examine websites targeted at the pregnant population and to assess the content therein relating to stillbirth risk factors and maternal behaviours.

Methods

The study was limited to websites hosted in Ireland and the UK, and results organised by provider and topic. A codebook was designed to record the information found on the websites. Data were collected on different website characteristics, and a search was undertaken for basic information related to stillbirth (prevalence, causes, procedures, consequences, etc.), and information relating to modifiable risk factors (smoking, alcohol/drug use, medicines use, sleep position, attendance at antenatal care, and weight management).

Results

92 websites were included in the study, of which 39.1% (n=36) contained basic information related to stillbirth and 29.3% (n=27) contained information related to modifiable risk factors. In total, only one website (1.1%) contained all the information that was sought. Websites hosted by charities (n=28) were more likely to contain basic information related to stillbirth, (39.3%, n=11 of the 28 websites hosted by charities) whereas the commercial sites (n = 15) were more likely to contain information about behavioural risk factors (53.3%, n = 8 of the 15 commercial sites).

Conclusion

The results of this study illustrate that websites directed at the pregnant population are a poor source for information related to stillbirth. Some stillbirth risk factors are modifiable; therefore, it is crucial that women and stakeholders can avail of reliable sources of information to make informed decisions.

3.2 Introduction

Pregnancy represents a transition period for women into motherhood; for many, this can be a transition to the unknown[324]. Due to having to cope with the uncertainty of being pregnant for the first time, many women and their partners draw upon the internet for information[325]. This information can influence their health-care related decisions[326].

Previous studies have concluded that around 80% of internet users use the Internet to find health-related information[322,323]. Andreassen et al. (2007) conducted a questionnaire-based study to explore the internet information seeking behaviours of the general population[327]. From a sample of 7934 people from seven different European countries, 44% reported that they had used the internet for health purposes, with respondents from northern Europe (German, Denmark and Norway) more likely to use the internet to seek health information compared to southern countries (Portugal and Greece)[327].

Many women also use the internet as a source of information during their pregnancy[326,328–330]. Sayakhot & Carolan-Olah (2016) conducted a systematic review to describe use of the Internet as source of information for pregnant women. They concluded that around 90% of the women who participated in the studies had accessed the internet for information, while factors such as having higher education levels, being married, being employed or being a first-time pregnant woman were associated with increased use[331]. Similarly, a recent study conducted by Jacobs et al. (2019) found that 96% of women in their Dutch sample used the Internet as an information source before or during their pregnancy[333].

Lagan et al. (2010) conducted a questionnaire-based study that confirmed that all women had used the internet to search for health information during their pregnancy. In their sample, 13% of women stated that the internet was the only source of information they used[326]. Although more than 95% used general search engines to find the information, less than half reported trusting that information. Government sites were cited as the least used, yet rated as the most trusted[326].

The internet has the potential to serve as a tool to empower and inform users in healthcare-related issues and in supporting patient choices[334]. There is evidence that interventions aimed at primary prevention have been delivered successfully onlin [335–337]. Previous research on internet use by people with different health conditions has shown that finding health-related information on the internet clarified many of their concerns and broke the feelings of isolation[338,339]. The same authors also concluded that finding this information online motivated discussions between these patients and their healthcare professionals[338,339]. In the aforementioned study by Lagan et al. (2010), around 50% of pregnant women reported that the Internet helped them be better prepared for their next antenatal appointment and feel more involved in the decision-making process[326].

However, not all types of pregnancy-related topics are uniformly represented online. Globally, there are 2.6 million cases of stillbirth every year[307]. Most of these stillbirths occur in low and middle income countries, where preventive strategies focus on improving care during labour and birth[1]. In high-income countries, the rate of stillbirth (after 28 weeks of gestation) varies from 1.3 to 8.8 per 1,000 births depending on the country. In Ireland, a stillbirth is defined as an infant born weighing 500 grams or more or at a gestational age of at least 24 weeks who shows no signs of life ("Stillbirth Registration Act, 1994", 1994), with a rate of 3.8 per 1,000 births in 2017[340].

Although not all stillbirths can be prevented, a systematic review published in the Lancet Stillbirth series found that a large proportion of stillbirths in high-income countries are associated with factors that are avoidable[87]. The authors of this systematic review also argue that increased clinical and community awareness of the risk factors associated with stillbirth might improve management and lower stillbirth rates. Despite some existing controversies regarding the modifiable risk factors associated with stillbirth, there is strong evidence suggesting that smoking, high amounts of alcohol drinking, illicit drug use, high BMI, certain sleeping positions, and poor attendance at antenatal care, are associated with a higher risk of stillbirth[341].

Pregnancy could be a teachable moment to motivate women to adopt healthy behaviours and reduce risk-taking[342]. Teachable moments give healthcare professionals an opportunity to inform their patients and utilise the intrinsic motivation associated with them to promote health and wellness[343]. However, information relating to stillbirth is not widely available in general, and healthcare professionals do not tend to discuss stillbirth as a possible outcome of pregnancy with their patients[27], letting this teachable moment pass.

In a national cross-sectional study conducted by Nuzum et al. (2018), 56% of participants within the Irish population were not able to identify a single risk factors for stillbirth, demonstrating a lack of public awareness of the prevalence of stillbirth, or potential risk factors associated with it[258]. Nevertheless, 88% of the respondents felt that the possibility of stillbirth should be included in antenatal education programmes[258]. This issue is in keeping with a qualitative study conducted by Kelley and Trinidad (2012) which concluded that parents who had experienced stillbirth had no prior knowledge or expectations of stillbirth as a potential occurrence[345]. The women in this study with high-risk pregnancies were concerned about prematurity, but not about stillbirth; they reported that they had never been informed about stillbirth[345]. The authors suggest that the lack of information about stillbirth online could be explained by the taboo and stigmatization surrounding stillbirth with the awkwardness and discomfort that this topic produces in others[345].

This fact not only hinders possible prevention strategies for stillbirth, but also leaves parents bereaved by stillbirth feeling very isolated. In a qualitative study conducted by Pollock et al. (2019), bereaved parents felt that they would have preferred to live with some fear, to be prepared and aware of the risks, rather than have to face the shock and isolation surrounding their loss[27].

To be able to make informed decisions, women who are planning a pregnancy or are already pregnant need to have access to updated high quality information[326], and, as shown in previous research, the Internet is a source of information commonly chosen by pregnant women. Expectant parents should be aware that stillbirth is a possibility

in order to be able to recognise the warning signs, and seek medical attention, before it is too late[47].

Geller, Psaros, & Kerns (2006) conducted a study where they systematically assessed several websites targeted at people who had experienced a pregnancy loss. They selected eight high quality websites that were acceptable for women and professionals to use[346]. However, this previous work was neither focused on stillbirth, nor on risk factors for it[346]. In our study, we do not only focus on websites addressed to people who have experienced a pregnancy loss, but also on websites that target the pregnant population in general, and hence can have some influence on the prevention of stillbirth[346].

The aim of this research was to examine websites targeted at the pregnant population to assess whether or not they had content in relation to stillbirth, and maternal behaviours in pregnancy that are associated with an increased risk of stillbirth. Our research questions were as follows:

a) How many websites directed at pregnant women, or women who have had a pregnancy loss, contain information about stillbirth?
b) How many websites directed at pregnant women, or women who have had a pregnancy loss, contain information about modifiable risk factors for stillbirth?
c) What are the differences between the different types of websites (e.g., UK vs. Ireland, General vs. Specialised, type of provider, etc.) in terms of the information provided?

3.3 Methods

3.3.1 Context of the study

This study was focused on websites targeted at pregnant women in the Republic of Ireland and the UK, based on the similarities among the antenatal care systems of both countries. In Ireland, all pregnant women and their babies are entitled to access

a free program of care with their General Practitioners (GP) and antenatal hospital services under the Maternity and Infant Care Scheme[347]. This program includes eleven visits during a woman's pregnancy, which are alternated between visits to the maternity unit/hospital and GP. The care of the women is midwifery-led in cases of normal risk, and a combination of midwifery and obstetrician care is provided in higher risk cases[347]. Similarly, the National Health Service in the UK offers 10 pregnancy appointments to all pregnant women. The care is also midwifery-led combined with GP appointments in normal risk cases, and obstetrician-led care in higher risk cases.

Both antenatal care systems have a similar scheduling for the antenatal visits: first booking visit is recommended between 8-12 weeks of gestation; the dating scan is performed from weeks 8 to 14 in the UK, and 10 to 14 in Ireland; a fetal anomaly ultrasound scan is offered between weeks 18-22, and monitoring gets more intensive after week 28.

Furthermore, both health services use several modes to communicate their findings; through the publication of reports and reviews (e.g., the HSE National Reports on Women's Health or the NHS National Maternity Review) and through public health campaigns that can include posters, leaflets and social media messages. (e.g., the HSE campaign on Fetal Alcohol Spectrum Disorders or the NHS "Health b4 pregnancy" campaign). Finally, both services direct the pregnant population to their institutional websites which they use to disseminate pregnancy-related information.

3.3.2 Research design

We conducted a quantitative content analysis to answer our research question. Quantitative content analysis provides the researcher with a systematic way of gathering and analysing text, which can be anything written, visual or spoken, and serves as a means of communication[348]. For the purposes of this study, quantitative content analysis has been used to descriptively examine the content of pregnancy-related websites. The key steps in quantitative content analysis include: developing the research questions and conceptualisation of hypothesis, sampling, developing a

coding scheme, collecting and coding the data, testing for reliability, and exposing findings and conclusions[349].

3.3.3 Selection of websites and data collection method

One researcher compiled a list of websites from different sources: (1) internet searches; (2) discussions with stakeholders, such as obstetricians, bereavement specialist midwives, allied healthcare professionals, and researchers; (3) links provided in National Bereavement Standards[350]; (4) hand searches of included websites. All authors also included suggestions after examining the compiled list of websites. Websites were only included if they were in the English language and hosted in the UK or Ireland. The following keywords, used in different combinations, were used to search for relevant websites: “pregnancy”, “stillbirth”, “stillbirth support”, “stillbirth Ireland”, “stillbirth UK” and “pregnancy loss”.

Websites were classified into different categories according to: provider, depending on what type of group or organization created the website; hosting country, depending on where the website was created; topic/target audience, depending on the main focus of the content of the website (Table 3.1). For more information on what websites were included in each category, please refer to Supplementary File 3.1.

Table 3. 1: Website classification categories with definitions.

Category	Value	Definition
Provider	Professional bodies	Websites from official professional organisations
	Charities and voluntary organisations	Websites from public organisations that are registered as charities
	Support groups	Websites created by groups of parents or affected people not registered as charities
	Commercial websites	Websites provided by private companies not including private health providers/insurers.
	Health service websites	Websites from national health providers
	Private health websites	Private websites with the purpose to offer a service or provide information
Country	Republic of Ireland	
	United Kingdom	
Topic/Target audience	General	Websites where the main topic is pregnancy/maternity and which target all pregnant women
	Specialised	Websites where the main topic is pregnancy loss and which target those who have experienced a pregnancy loss
	Other	Websites that do not fit in any of the previous categories, for example, general bereavement support services

A codebook was developed to assign coding values to the different categories of interest (see Supplementary File 3.2). Availing of a codebook allows the process of the data coding to be systematic and replicable[349]. The categories included in the codebook were name and link of website, date websites were accessed, main topic of website (General vs. Specialised), website provider, target audience, hosting country, presence of advertisement, and accreditation from official organisations. In addition, the website content pertaining to basic information about stillbirth was searched for and recorded, including prevalence rates, procedures (e.g. legal procedures after stillbirth, post-mortem examination, etc.), medical or psychological consequences (including; bereavement process, physical changes, health-related associated issues, etc.), and support for parents. Information relating to modifiable risk factors for stillbirth was also included and recorded, specifically that relating to risk factors with a behavioural component (smoking, alcohol and drug use, medicine intake, sleep position, attendance to antenatal care, and weight management). Information about other types of risk factors for stillbirth (e.g: fetal growth restriction, high blood pressure, placental insufficiency, etc.) was recorded in a general variable, since the focus of this study were modifiable risk factors. Data were considered as absent when no information explicitly linked to stillbirth was found. Information about other types of pregnancy loss, or when risk factors were mentioned without reference to stillbirth, was not included.

3.4 Data extraction and analysis

An excel sheet was used to record the information obtained through the data collection process. Required information was searched for in each of the different websites, and recorded as per the codebook in the excel sheet. If the information was explicitly expressed in the body of the website, it was coded as present. However, if the data were not found after utilising the website's own navigation panel and the website's search box (when available) with the terms stillbirth, stillborn, pregnancy loss, and loss, it was then marked as absent. The data were recorded by the main author between May 2019 and July 2019. Given the nature of

websites, content can be updated regularly. Therefore, screenshots of the information on the date websites were accessed were taken, and stored in NVivo V12. The second author reviewed the Nvivo V12 file, including the screenshots, for consistency, and any discrepancies were discussed. Data were analysed using descriptive statistics and the analysis was conducted using SPSS statistical software v.25.

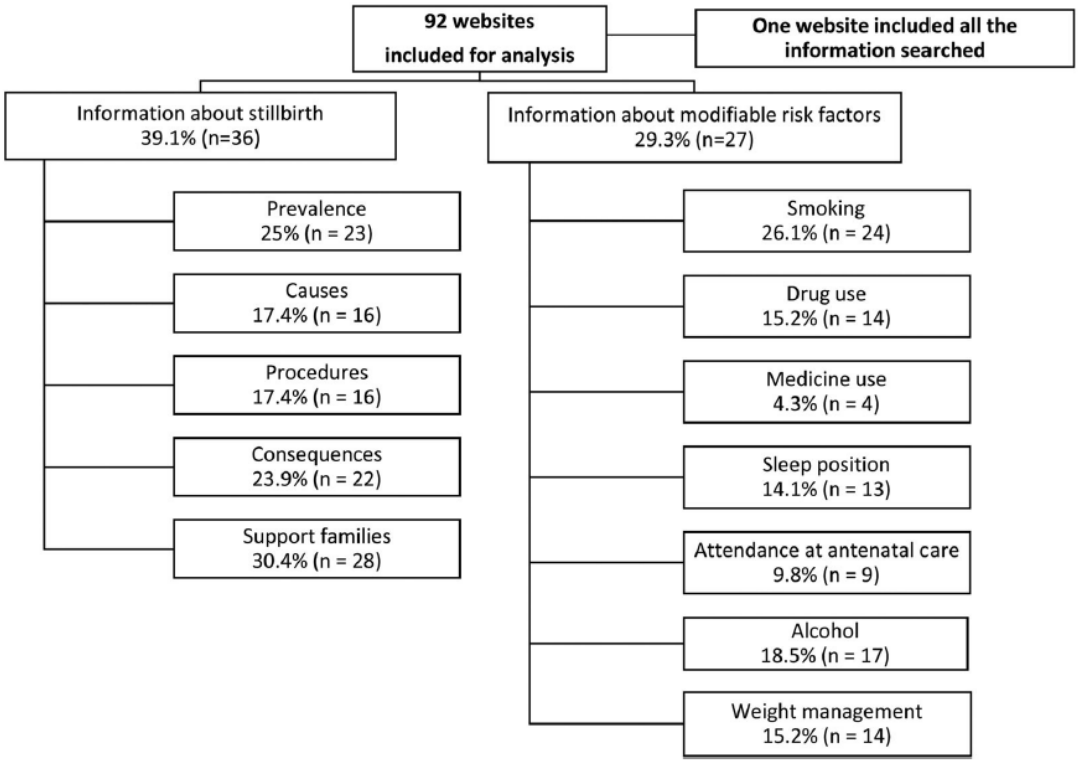
3.5 Results

A total of 92 websites were included in this study, of which 39.1% (n=36) websites were hosted in the UK and 60.9% (n=56) in the Republic of Ireland. The websites were divided into the following categories of websites, depending on the provider: professional bodies' sites (8.7%, n=8), charities and voluntary organizations sites (30.4%, n=28), support groups sites (13%, n=12), and commercial sites (16.3%, n=15), health services sites (14.1%, n=13) and private health sites (17.4%, n=16) (See Table 3.2). Regarding the classification per topic or targeted audience, the websites were divided as follows: specialised websites (32.6%, n=30), general websites (63.0%, n=58), and other (4.3%, n=4) (See Supplementary File 3.1).

At least one positive result relating to basic information about stillbirth was present in 39.1% (n=36) of the included websites, with information about support for families (30.4%, n=28) and prevalence (25.0%, n=23) being the most common topics. Almost 12% (n=11) of the 92 websites contained all the basic information about stillbirth that was sought. Information regarding modifiable risk factors was found in 29.3% (n=27) of the websites, with smoking being the most common risk factor mentioned (26.1%, n=24), followed by illicit drug intake (15.2%, n=14) and sleep position (14.1%, n=13).

Only one of the 92 websites contained all the information that was sought pertaining to basic information about stillbirth and associated modifiable risk factors (see Figure 3.1).

Figure 3. 1: Information searched on the websites and prevalence of appearance.
**Categories are not mutually exclusive.*



When examining the websites by provider, as outlined in Table 3.2, websites classified as charities were more likely to contain basic information about stillbirth (39.3%, n=11) than any other provider, followed by commercial sites (53.3%, n=8) and support groups sites (58.3%, n=7). Support for families was the most common topic, regardless of the provider. Websites hosted by support groups and commercial sites were the most likely to report on emotional or physical consequences after a stillbirth (41.7%, n=5 and 46.7%, n=7 respectively). On the other hand, charities and support groups focused on reporting stillbirth prevalence (32.1%, n=9 and 41.7%, n=5 respectively). Only two (25%) of the eight professional bodies' websites contained some basic information about stillbirth, focusing equally on prevalence, causes, consequences and support for families (12.5%, n=1). Seven (53.8%) of the health services websites contained some basic information about stillbirth focussing mostly on offering support for families (46.2%, n=6); and only one (6.3%) of the private

health sites contained some basic information about stillbirth, also giving information on support for families.

When looking specifically at information about modifiable risk factors for stillbirth by provider, websites hosted by commercial sites were the most likely to contain information about at least one risk factor (53.3%, n = 8), followed by the sites hosted by charities (28.6%, n = 8). Only two (25%) websites hosted by professional bodies and one (8.3%) website classified as a support group contained information regarding at least one modifiable risk factor for stillbirth. Most websites focused their content on smoking (25%, n = 7 for charities; 46.7%, n = 7 for commercial sites; 38.5%, n = 5 for health services sites and 18.8%, n = 3 for private health sites). Medicines intake was the least represented risk factor.

Table 3. 2: Information depending on provider

		Websites	**Professional bodies**	**Charities**	**Support groups**	**Commercial websites**	**Health services**	**Private health websites**
		n=92(%)	n=8(8.7)	n=28(30.4)	n=12(13.0)	n=15(16.3)	n=13(14.1)	n=16(17.4)
Basic information about stillbirth	Prevalence	23(25.0)	1(12.5)	9(32.1)	5(41.7)	5(33.3)	3(23.1)	0(0.0)
	Causes	16(17.4)	1(12.5)	8(28.6)	2(16.7)	3(20.0)	2(15.4)	0(0.0)
	Procedures	16(17.4)	0(0.0)	7(25.0)	3(25.0)	3(20.0)	3(23.1)	0(0.0)
	Consequences	22(23.9)	1(12.5)	7(25.0)	5(41.7)	7(46.7)	2(15.4)	0(0.0)
	Support families	28(30.4)	1(12.5)	10(35.7)	4(33.3)	6(40.0)	6(46.2)	1(6.3)
Information about behavioural risk factors	Smoking	24(26.1)	1(12.5)	7(25.0)	1(8.3)	7(46.7)	5(38.5)	3(18.8)
	Drug use	14(15.2)	1(12.5)	6(21.4)	0(0.0)	3(20.0)	3(23.1)	1(6.3)
	Medicine intake	4(4.3)	0(0.0)	1(3.6)	0(0.0)	2(13.3)	0(0.0)	1(6.3)
	Sleep position	13(14.1)	1(12.5)	6(21.4)	0(0.0)	5(33.3)	1(7.7)	0(0.0)
	Antenatal attendance	9(9.8)	0(0.0)	5(17.9)	1(8.3)	1(6.7)	2(15.4)	0(0.0)
	Alcohol	17(18.5)	1(12.5)	6(21.4)	0(0.0)	6(40.0)	3(23.1)	1(6.3)
	Weight	14(15.2)	1(12.5)	4(14.3)	1(8.3)	5(33.3)	3(23.1)	0(0.0)

When comparing the websites by country, websites hosted in the UK were more likely to report basic information about stillbirth than Irish websites (55.6%, n=20 vs. 28.6%, n=16, p=0.010). Websites hosted in the UK were also more likely to contain information about modifiable risk factors (41.7%, n=15 vs. 21.4%, n=12, p=0.037). Regarding other types of risk factors, there were no differences between the UK websites and the Irish websites (36.1%, n = 13 vs. 23.2%, n = 13, p=.180). (see Figure 3.2). As mentioned previously, the websites were also categorised by topic. Specialised websites (websites targeting people who have experienced a pregnancy loss) were more likely to contain basic information about stillbirth compared with general websites (46.7%, n=14 of 30 vs. 36.2%, n=21 of 58), however, this difference was not statistically significant (p=0.533). On the other hand, there were differences observed between the groups of websites when examining information regarding modifiable risk factors (13.3%, n=4 of 30 vs. 39.7%, n=23 of 58, p=0.015) (See Figure 3.3).

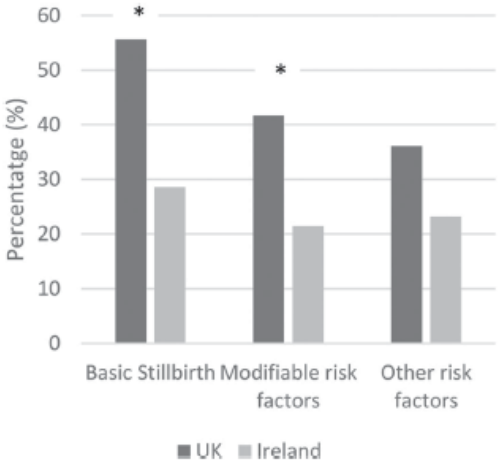


Figure 3. 3: Information by country

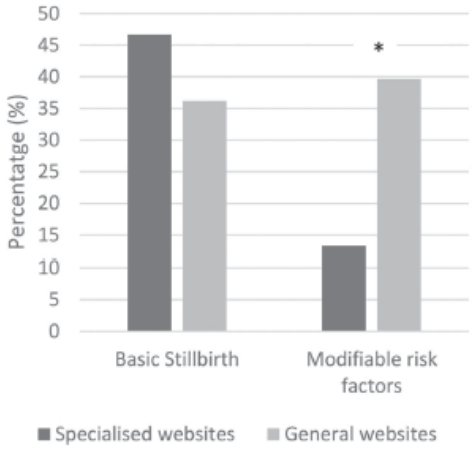


Figure 3. 2: Information by topic

A large proportion of the websites contained external links to either professional guidelines, patient information leaflets, or other websites. External links providing basic information about stillbirth were present in 43.5% (n=40) of the websites, whereas external links containing information about modifiable risk factors were present in 21.7% (n=20). Only 13 (23.2%) out of 56 websites not containing any basic information about stillbirth provided external links with this information. Seven (10.8%) out of 62 websites without information about any modifiable risk factor provided external links with this information.

Most of the external links provided redirected the user to other websites for both types of information (30.4%, n=28 in the case of basic information about stillbirth, and 13%, n=12 in the case of information about modifiable risk factors (see Graph 2)). However, clinical guidelines were frequently linked when providing information about risk factors (6.5%, n=6), whereas patient information leaflets were linked when providing basic information about stillbirth (8.7%, n=8) (see Figure 3.4).

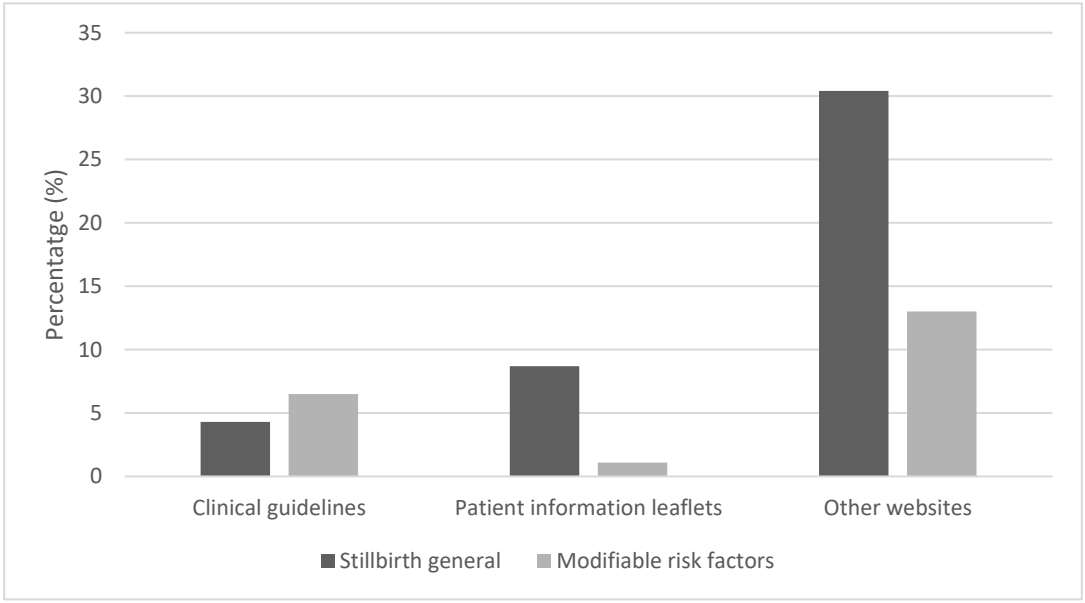


Figure 3. 4: Types of external links used

3.6 Discussion

The purpose of the current work was to examine a list of websites targeted at the pregnant population and hosted in the Republic of Ireland and in the UK in order to assess their content in relation to stillbirth, and maternal modifiable risk factors in pregnancy that have been previously associated with a higher risk of stillbirth. The results of the study illustrate that less than half of websites contained basic information about stillbirth, and approximately 30% contained information on at least one of the modifiable risk factors examined. However, only one website contained all the information that was specifically searched for.

The websites that contained information about stillbirth predominantly provided information on the prevalence of stillbirth and supports for families who are bereaved. Providing data about the prevalence of stillbirth is very valuable as demonstrated in a previous qualitative study conducted by Kelly & Trinidad (2012); which shows that parents who had experienced a stillbirth were surprised to learn both the rates of stillbirth worldwide and also the rate in their high income country[351].

The fact that websites include data about the prevalence of stillbirth in high income countries can have a positive impact on pregnant women's behaviours, since it could prevent biased beliefs such as thinking that stillbirth only happens in developing countries[344]. However, providing women with this information alone is not enough, given that the most successful communication behaviour change interventions use three or four categories of communication techniques engaging participants at different levels[352]. Hence, more comprehensive communication strategies are necessary.

Research shows that there are several maternal modifiable risk factors associated with stillbirth and other adverse perinatal outcomes[341]. Our study found that these websites focus their efforts on informing the public about the risks of smoking and illicit drug use during pregnancy. Providing this information to women is very positive since there is evidence associating these behaviours with a higher risk of stillbirth and other adverse pregnancy outcomes, and considering that many women use the internet to access pregnancy-related information in the antenatal period[332].

However, as this study illustrates, there are other maternal modifiable risk factors that have also been associated with a higher risk of stillbirth which are not included in website content. For example, the evidence related to the risk of stillbirth and the intake of over-the-counter medicines, herbal supplements and/or micronutrient supplements is very limited and conflicting[341], yet this information is not present in most of the websites, leaving women potentially uniformed.

Another issue of concern is that only two out of the eight of the professional bodies' websites contained some basic information about stillbirth, and just two contained information about risk factors. These websites tend to use links to clinical guidelines instead of summarizing the information, and/or adapting it for the general public, within the body of their websites. While these guidelines may contain rich evidence-based information, they need to be up-to-date, as well as user-friendly and adapted to the general population, to be able to serve as an informative tool [353]. For other types of website sources, using links to clinical guidelines might be due to a lack of expertise in the area, resulting in them referring to appropriately endorsed guidelines and recommendations.

This issue raises further questions relating to the reliability of information obtained online[354–357]. It is the official organisations' responsibility to disseminate their research efforts and provide evidence-based information. Dobbins et al. (2018) conducted a review of the literature to identify any available tools that assessed reliability of websites in a quick and easy-to-use manner. The authors concluded that the tools available had poor reliability, or had not been assessed for reliability. As a result, women have few resources available to assess the reliability of the information they are obtaining, besides the reassurance of using websites provided by official institutions that follow national guidelines when providing advice. However, as per our own experience, navigating these websites and their clinical guidelines is not easy, which potentially leads users to other potentially more attractive websites in terms of layout, ease of navigation, level of interaction, etc., but that might be providing unreliable information.

When comparing the specialised websites (those focused on pregnancy loss) to general pregnancy-related websites, there were no differences regarding the basic

information provided about stillbirth. However, general websites targeting all pregnant women tended to give information about modifiable risk factors more often than specialised websites. Having specialised websites giving information about stillbirth and bereavement support is very positive. Nevertheless, by not providing information about modifiable risk factors it seems that there is a lack of recognition from these specialised websites that women who have already experienced a stillbirth have a higher risk of stillbirth in their subsequent pregnancies[92], and hence, the need to inform these women about modifiable risk factors is even more relevant.

With the peak in popularity of social media platforms in the last few years, many content creators and social media users have used this resources to share and obtain health information[359]. Moorhead et al. identified several benefits of social media use to obtain health information, these including increased interaction with other people with the same health concerns, higher availability of shared and tailored information, peer/social/emotional support, real time low-cost communication for public health surveillance and monitoring of public response to health issues. For healthcare professionals, they provide a source of data about patient experiences and provide a platform for risk communication[360]. However, to our knowledge, no research has been conducted to explore the use of social media to obtain information about stillbirth.

The increasing availability of health information on social media platforms has raised concerns about the quality and reliability of such information[360]. Some of those concerns relate to the lack of regulation of social media platforms, which means that most users can post health information regardless of its scientific validity, which is also link to the spread of misinformation or "fake news"; as well as biased and conflicting information, as some content creators might have commercial interests when it comes to promoting certain products or therapies[360]. Other concerns identified by Moorhead et al. in their systematic review are lack of confidientiality and privacy, risk associated with communicating harmful information, information overload, difficulty to apply information found to individual's situation, adverse health consequences, negative health behaviours, or acting as a deterrent from

seaking professional health advice[360]. Hence, even though social media might be a useful resource to provide health information, more research is necessary to address what are the bests ways to utilise this resource and ensure the reliability of the information provided.

There are some limitations to this study. The website search was done manually and limited to the United Kingdom and the Republic of Ireland. However, we consider that this kind of search simulates what an internet user would do. The search for the information in the websites was done systematically page by page. As mentioned previously, there is not a wide recognition of stillbirth, and people tend to avoid hearing or speaking about it[27]. Further, previous research indicates that people seeking health information on the internet do not have a systematic plan[361]. Hence, as per our own experience navigating the websites, and previous studies conducted in the area of online health information seeking behaviour[333,362], we can assume that it is even less likely that pregnant women accessing these websites find the information relating to stillbirth or risk factors, unless they are specifically looking for it.

The findings of our study have several implications. Firstly, maintaining the silence around stillbirth hinders potential preventive strategies. Not giving attention to the fact that stillbirth can happen, and that there are measures that may reduce the risk, not only feeds into the stigma of stillbirth[27], but also promotes an altered perception of risk or a biased belief that "this only happens to other people"[33,344]. Knowledge and education are not enough to change behaviour; however, this is the first step in most instances. It is essential that people are educated, not only to be aware of risk factors associated with stillbirth, but also to understand why and how behaviour change is important[363]. Hence, by not using these websites to clearly inform people about stillbirth and the relevant modifiable risk factors, the opportunity to inform and empower them is lost.

Clear and effective communication in healthcare settings and in public health is essential and can have an effect on health outcomes[364,365]. To avoid misinforming the public, it is important to use the right terms and avoid euphemisms, for example using the word "stillbirth" instead of "loss". Euphemisms are often used as a way to

avoid talking about a topic that the speaker would rather avoid, normally in response to taboos[366]. Previous research in the field of palliative care shows that using misleading language to avoid causing pain denies that person the opportunity to prepare for the next steps[367]. Using clear language would avoid confusion and help give visibility to the fact that stillbirth can happen and have very serious consequences. On the other hand, one of the most important things to consider is to make sure the information is not only accurate but also user-friendly. Women need to be able to find and understand relevant information easily. Use of a strategic communication framework to ensure effective communication, such as the one proposed by the WHO, could be of use[368]. This framework can be used as a resource for the development of communication strategies, based on the principles of effective communication: accessibility, actionability, credibility and trustworthiness, relevance, timeliness and understanding.

There is evidence that people use the internet to find health-related information[325]. This information can subsequently influence their decision-making[326] and can have preventive effects for certain issues[335–337]. Hence, it is important to break the silence on stillbirth and make sure women can avail of reliable sources of information to make informed decisions, not only during their pregnancies, but also when they are planning their first or subsequent pregnancy. Currently, we are not aware of what sources of information people use to educate themselves about stillbirth, so further qualitative and quantitative research is necessary to identify these sources of information and improve information provision to parents.

3.7 Inter-chapter synopses

Chapter 3 aimed to evaluate the information available in Irish and UK websites regarding stillbirth and its associated modifiable/behavioural risk factors. Findings in Chapter 3 highlight that there was a lack of consistency in the information available in such websites, and that there were gaps in the information provided. Chapter 3 emphasised the need for accurate, complete, and readable information on stillbirth and modifiable risk factors online to be made available for parents, healthcare professionals and the general public. This lack of information online highlights one of

the potential barriers to behaviour change during pregnancy, as it is a resource that could be utilised to increase people's awareness. The following chapters (Chapter 4-6) provide a deeper exploration of other potential barriers and facilitators to behaviour change. Concretely, Chapter 4 imed to identify the facilitators and barriers to seeking and engaging with antenatal care in high-income countries. Before conducting the study exposed in Chapter 4, it was well known that lack of attendance at antenatal care is a modifiable risk factor for stillbirth[206], and hence, there was a need to further understand this behaviour in order to identify which influences are affecting it.

Chapter 4 - Facilitators and barriers to seeking and engaging with antenatal care in high-income countries: a meta-synthesis of qualitative research.

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Chapter 4 - Facilitators and barriers to seeking and engaging with antenatal care in high-income countries: a meta-synthesis of qualitative research.

4.1 Abstract

Inadequate attendance to antenatal care has been associated with negative maternal and fetal outcomes, including stillbirth. This study aimed to identify facilitators and barriers to antenatal care attendance. A systematic search was conducted in March 2019 and updated in January 2021. Qualitative studies involving pregnant or post-partum women up to 12 months from high-income countries that provided data about facilitators and barriers to antenatal care attendance were sought. Meta-ethnography was used to inform this meta-synthesis. Fifteen studies were included in the analysis. Findings indicate that inadequate antenatal care attendance is influenced at different levels. Aspects like sociodemographic factors, difficulties navigating the health system, administrative delays, lack of flexibility and tailored care, constant change of carer and communication issues also act as barriers. These issues affect women's access to knowledge and the formation of women's' beliefs and feelings towards seeking care. On the other hand, having a positive attitude towards the pregnancy, encountering empathetic healthcare professionals and availing of social support acted as facilitators. The reasons why women seek or delay attending antenatal care are multifactorial and can be explained using the Social Determinants of Health Framework. Any response needs to be taken across all levels of influence and not just focused on the individual. A better understanding of the barriers and facilitators to antenatal care might contribute to informing intervention or policy development addressing this issue.

Keywords: Facilitators; Barriers; Antenatal Care; Stillbirth; Meta-synthesis; Meta-ethnography; Qualitative research.

What is known:

- Delaying or avoiding care during pregnancy can result in adverse pregnancy outcomes, such as stillbirth.
- Although there is evidence of the benefits of attending antenatal care, there are some women who continue to delay or avoid seeking care due to different reasons.

What this paper adds

- The reasons why women might not attend or engage with antenatal care are multifactorial and the personal barriers identified are only one such aspect.
- Factors associated with the women, the individual healthcare professionals, the health-care system and the social environment have an influence on women capability and willingness to attend antenatal care in high-income countries.

4.2 Introduction

Within its continuum, antenatal care provides an opportunity for communication, health promotion, prevention, screening and diagnosis of diseases[319]. The WHO recommendations on antenatal care include a minimum of eight contacts: five in the third trimester, one contact on the first trimester and two contacts in the second trimester[204].

A previous study comparing the levels of utilization of antenatal care in different countries concluded that the proportion of late attenders was low in Finland (4%), France (4%), and Italy (4%), moderate in Belgium (7%), Germany (8%), Norway (7%), and Sweden (9%), and high in Greece (18%), Ireland (29%) and Portugal (18%)[369]. These reported differences are linked with women from lower socioeconomic groups[370,371], specific ethnic groups[370,372,373], and are associated with women living in deprived areas [374].

Previous studies have associated inadequate attendance at antenatal care with negative maternal and fetal outcomes, including stillbirth[206,375–377]. Stacey et al. (2012) reported that women who attended less than 50% of the recommended antenatal visits, were 3 times more likely to suffer from late stillbirth and a trend analysis found a significant relationship between decreasing visits and an increase in the risk of stillbirth[206]. Further, the researchers also concluded that fetuses which were small-for-gestational age were more likely to be stillborn if they were not identified prior to birth reinforcing the importance of regular antenatal care attendance[206]. Lack of attendance at antenatal care could prevent healthcare professionals from identifying difficulties during pregnancy. Moreover, antenatal care is also an opportunity for healthcare professionals to provide women with vital information about their health[319], which could lead to healthier lifestyles during the pregnancy decreasing the risk of adverse outcomes.

Previous research has demonstrated that women do not always use antenatal care services even when they are accessible, especially if they belong to ethnic minorities or marginalised groups[378]. Additionally, other evidence suggests that women might not find the medicalised antenatal care model suitable for their needs, or else find themselves in unpleasant situations while seeking care and cease to attend the

services[379]. This evidence suggests that there are different reasons or barriers that women might be encountering when trying to access antenatal care, other than accessibility and affordability, and that these need to be explored further.

Qualitative research allows the researcher to understand the nature, strength and interaction of the variables studied with a holistic perspective that preserves the complexities of human behaviour[380]. In our study, qualitative research helped us explore the values and beliefs that might be associated with the acceptability of antenatal care, in order to identify outcomes relevant to women. A meta-synthesis differs from a narrative review and a systematic literature review in that it involves a secondary analysis and re-interpretation, but based on previous findings instead of primary data[381].

The aim of this meta-synthesis is to analyse and synthesise all the evidence drawn from qualitative research in order to identify facilitators and barriers to antenatal care in high-income countries. This might potentially inform the development of a behaviour change intervention to minimise barriers and facilitate access to antenatal care.

4.3 Methods

4.3.1 Search strategy

We performed a comprehensive systematic search of the literature for all qualitative research that explored women's facilitators and barriers to seeking, attending, and complying with antenatal care during pregnancy. The databases searched were CINHAL, PsychINFO, Pubmed, SOCindex and Web of Science and the searches were conducted in March 2019 and then updated in January 2021.

Search terms were selected based on a preliminary scan of the relevant literature and the research aims. The search terms used were *facilitators, barriers, promoter, benefit, attitude, opportunity, determinant, promotion, intention, education, initiative, prevention, pregnancy, attendance, antenatal care, prenatal care, antenatal booking* (See supplementary Table 4.1).

4.3.2 Study selection

Three members of the research team (TES, LL, SM) independently reviewed the titles and abstracts of the studies resulting from the database search.

Studies were included for further review if (1) they used a qualitative or mixed methods design, (2) they were written in English, (3) the participants were pregnant women or up to 12 months post-partum, (4) they were conducted in high income countries; high-income countries were defined based on the World Bank Country Classification (Gross National Income per capita of $12,696 or more in 2020)[382] and (5) included extractable data about facilitators and barriers to seeking, attending or compliance with antenatal care.

Studies were excluded if they did not include qualitative data or if they were not original research. The decision to exclude previous reviews and meta-synthesis was based on concerns about duplication of data. Studies that included different types of participants (e.g., healthcare professionals and pregnant women, partners, and pregnant women) were only included if the data obtained from the pregnant women was differentiated from the rest. No year restrictions were applied (See supplementary table 4.1).

4.3.3 Data extraction of study characteristics

The characteristics of the studies were extracted and tabulated into a data extraction sheet by one author (TES). The following data was extracted from each study: country of publication, year of publication, aims, design, data collection method, sampling or recruitment strategy, consent process, number of participants, age of participants, pregnancy status, timing of data collection and method of data analysis.

4.3.4 Quality Assessment

Two authors (TES, SM) independently used the Critical Appraisal Skills Program (CASP) for qualitative studies to assess the quality of the studies. Since the CASP tool does not explicitly include scoring, we decided to score as following: 0 points for "No", 1 point for "Can't tell", and 2 points for "Yes". The CASP tool is composed of 10 items assessing validity of results, appropriateness of research design, aims and data collection methods, reflexivity and ethical issues, and value of the analysis and

findings of a study. Hence, the maximum score related to reporting quality that can be obtained with the CASP is 20 points.

Previous research in the area of quality appraisals has demonstrated a correlation between the value of a study as a source for a synthesis and the quality of its reporting [383], it was decided then that only high-quality studies would be kept for synthesis and those with a score under 10 were excluded (See supplementary file 4.2 for results of CASPS). Ethical approval for the study was not sought as it did not include human subjects.

To assess our individual review findings, we used the GRADE-CERQual approach. This approach facilitates assessment of how much confidence can be places on individual review findings from a synthesis of qualitative research [384] (See supplementary Table 4.3).

4.3.5 Process of synthesis

The synthesis of qualitative research was informed by meta-ethnography and we used the eMERGe reporting guidelines for meta-ethnography to write up this study [385]. Meta-ethnography is an interpretative approach originally developed by Noblit and Hare that facilitates "putting together" all of the research available by translating qualitative studies into one another [386]. By doing so, the analyst (TES) translated the studies into one another's concepts, and into their own world view, meaning that the synthesis will inevitably be partly a product of the author [387].

Noblit and Hare (1988) proposed a series of phases that overlap and repeat along with the conduction of the synthesis (see Supplementary table 4.4).

Phase 1 and 2 were completed through a systematic search of the literature detailed above (see Figure 1). Phase 3 involved reading the studies in depth in order to become familiar with them, during this phase data from the studies was being extracted. To do so, each paper was read carefully, and notes were taken identifying the different data that needed to be extracted by one author (TES). Once all the relevant study characteristics were identified, these were translated into our data collection sheet.

During Phase 4, the studies were imported into an Nvivo12 database to facilitate the identification of concepts and themes. One author (TES) conducted the line-by-line coding of the results and discussion section of each study, consulting with the rest of the team when in doubt. A second author (SM) coded a sample of the studies to ensure consistency in the coding process. As the coding progressed in each new study, we added to our list of codes or created new ones when necessary. Every sentence of the studies had at least one code applied.

The studies were comparable in that they were all contextualised in high-income countries which means that their antenatal healthcare systems could be comparable in terms of standards of care and resources; they share similar aims, exploring women's experiences during antenatal care and their samples are similar - pregnant or post-partum women, hence we considered they relate reciprocally.

Both first and second order constructs were extracted for analysis [388]. We understand that our access to first order constructs was limited by the fact that the data was already selected by the original authors. Hence, we utilised author statements and interpretations to obtain additional insights, context and explanations. In meta-ethnography, second order constructs complemented by participants narratives (first order constructs) are the usable data. Very few studies included reflective statements that could have helped us to assess the influence of the author's background over their own interpretations. The key concepts explored in the analysis were related to factors that either facilitated or hindered access to antenatal care for pregnant women in high-income countries.

Once the initial coding was completed, the text added to each code was examined to check consistency of interpretation and additional coding was performed when necessary. We obtained a list of concepts that were grouped into themes and categories using thematic analysis. These categories included "Women's individual factors", "Individual healthcare professionals", "Healthcare-system", and "Social environment". These categories were created inductively based on the data obtained from the studies analysed, and not by previous expectations or knowledge. The themes and categories were refined through discussions with all authors.

Phases 5 and 6 involved translating the studies into one another and synthesizing those translations. The themes were further refined through the coding process to identify if the themes reflected the meaning of each individual studies. For example, the code “Worries about being pressured into termination by social services” developed into “Worries about being pressured into termination” when, by adding more data to the code, it was clear that this phenomenon was influenced by many other sources besides the social services. The initial codes and themes were examined and combined when they described similar findings. To establish the relationships between the different concepts we utilised concept maps stating the influence of each concept over the other, and these maps were discussed with the research team.

As the context of the studies were similar, we did not deem it necessary to create sub-groups of studies to preserve the context of the studies in the process of translation. Similar contexts are beneficial for our study since the aim was to identify facilitators and barriers to antenatal care in high-income countries. We considered that with similar contexts, the more specific the identified facilitators and barriers would be. This may better inform the potential development of a context-specific intervention.

The influence of each study over each concept obtained after analysis is documented using references and quotes. The quotes are obtained from the primary study participants and by the primary author’s explanations and interpretations, and those which more clearly exemplified the interpretations made were selected.

As the analysis progressed, through the interpretative integration of findings, it became evident that the results were explainable by the Social Determinants of Health Model[389], which is discussed further in the discussion section.

The result of the translated concepts, their relationships and the primary data were used to create a line of argument, which is presented here (Phase 7).

4.4 Results

4.4.1 Search outcome

The PRISMA Flow Diagram in Figure 1 shows the process of inclusion of studies. The electronic databases searched identified 4517 studies. After screening titles and abstracts, a total of 4487 records were excluded and 33 studies remained eligible for full text review.

Detailed review of the remaining 33 full texts was conducted with 13 studies meeting the inclusion criteria. Seven additional studies were reviewed for potential inclusion following hand searching the reference list of the 13 included studies, and two additional studies were included. The final number of studies included for synthesis was 15 (Figure 4.1).

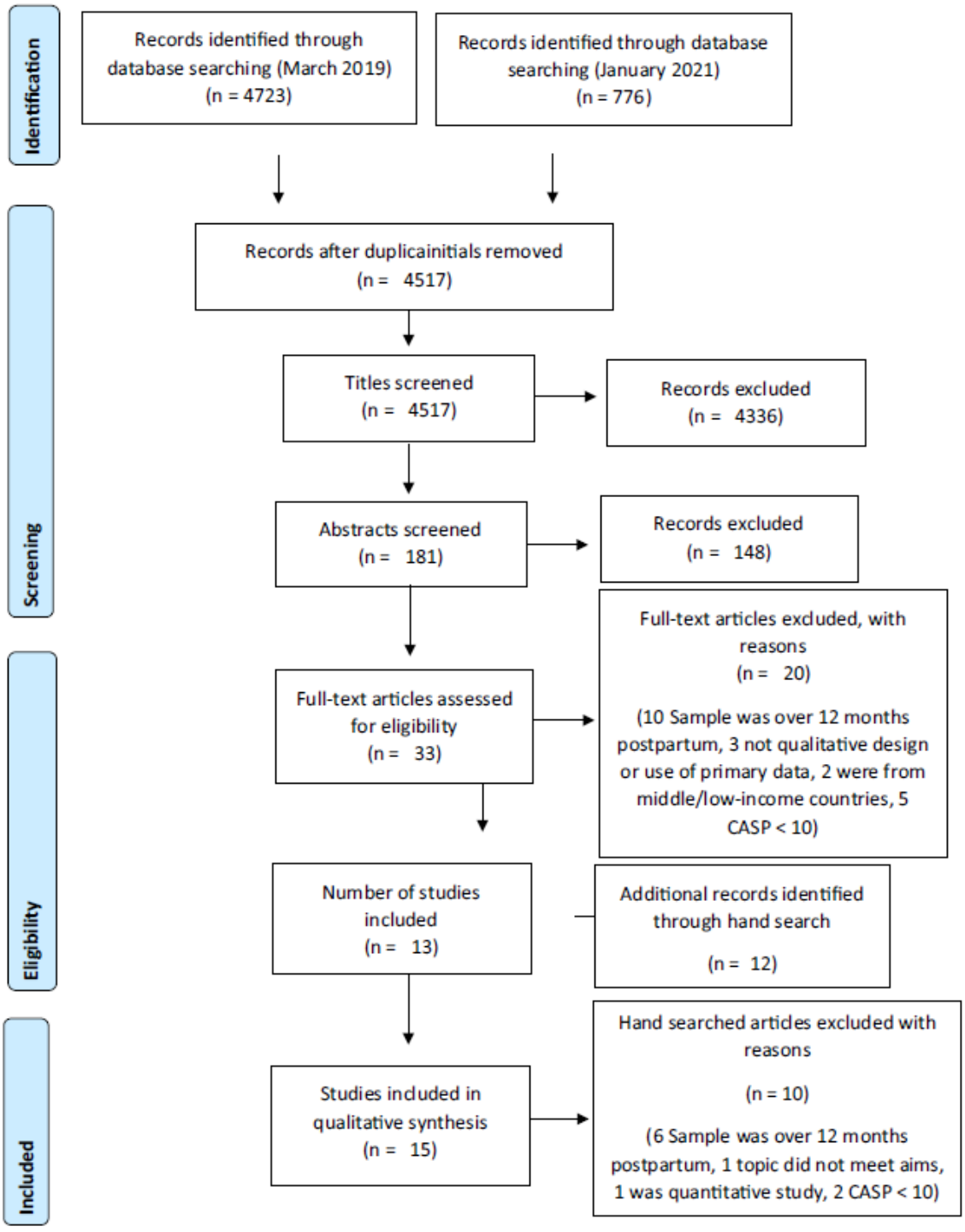


Figure 4. 1: PRISMA Flow Diagram showing the process of inclusion of studies. From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed100009

4.4.2 Study characteristics

The characteristics of the studies included are shown in Table 4.1. Of the 15 studies included for analysis, five were conducted in the UK, six in the USA, one in Sweden, one in Denmark, one in Switzerland and one in Ireland. The years of publication ranged from 1994 to 2020. Fourteen of the studies were qualitative and one study used mixed methods.

The studies used different data collection methods; ten used semi-structured interviews, five used focus groups.

The number of participants in the studies ranged from 9 to 48, with ages ranging from 16 to 43 years. Four of the studies included pregnant women only, five included postpartum women up to 12 months after birth only and six included both pregnant and postpartum women.

Table 4. 1: Characteristics of the included studies

Reference	**Callaghan et al. (2011)**	**Daniels et al. (2006)**	**Docherty et al. (2012)**	**Funge, J. K., et al. (2020).**	**Haddrill et al. (2014)**	**Hatherall et al. (2016)**	**Johnson et al. (1994)**
Country	UK	USA	UK	Denmark	UK	UK	USA
Year of publication	2011	2006	2012	2020	2014	2016	1994
Aims	Exploring late booker women's own accounts and perspectives on their relationship with NHS pregnancy care	Identify unique facilitators and barriers to low early prenatal care initiation among low socioeconomic Black women in Atlanta	Determine whether pregnant women's perceptions of current antenatal provision differed according to their socioeconomic deprivation background	Exploring undocumented immigrant women's experiences and their access to both formal and informal maternity care services in Denmark	Carry out a qualitative study with a broad range of late booking women to gain deeper understanding of the reasons why some women present late for antenatal care	Understanding what prevents or enables women living in an urban setting with high levels of diversity and social deprivation to begin a package of publicly-funded antenatal care early in pregnancy.	Explore the reasons given for not seeking prenatal care by women delivering at this same hospital
Design	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study.

Data collection method	Semi-structured interviews.	Focus groups.	Semi-structured interviews.	Semi-structured interterviews	Semi-structured interviews.	Semi-structured interviews and focus groups.	Semi-structured interviews.
Recruitment	Purposive-sampling from one maternity unit.	Purposive-sampling from a community health centre in Atlanta.	Purposive-sampling from one maternity unit.	Women were recruited through the Health Clinic for Undocumented Immigrants	Purposive-sampling from one maternity unit.	Purposive-sampling from one maternity unit, a health advocacy and a community based organization.	Women were selected from a group of women who reported delivering without prenatal care
Consent process	Written informed consent	Written informed consent	Consent obtained. Form not specified.	Written informed consent	Written informed consent	Written informed consent	Written informed consent
Number of participants	20	32	21	21	2	48	15
Age	16-43	16-36	17-40	20-42	15-37	20-34	20-32
Pregnancy status	Pregnant or within 6 months postpartum	Pregnant	Pregnant	Pregnant or up to 8 months postpartum	Pregnant	Pregnant or having given birth recently	Postpartum

Timing of data collection	Late 2006 and early 2008	December 2002 - March 2003	January 2007 - December 2008	January 2018 - January 2019	September 2006 – July 2008	March 2011	January 1989 - April 1989
CASP score	17	14	16	12	16	14	10

Table 4.1: Characteristics of the included studies (continuation)

Reference	**Larsson et al. (2017).**	**Mehta et al., (2017).**	**Meyer et al., (2016).**	**Roman et al., (2017).**	**Ross, E. (2012).** (Ross, 2012)	**Sami, J., et al. (2019).**	**Ward et al., (2013).**	**Tighe, S. M. (2010).**
Country	Sweden	US	USA	US	UK	Switzerland	USA	Ireland
Year of publication	2017	2017	2016	2017	2012	2019	2012	2010
Aims	Deepen the understanding of the meaning of caring in prenatal care from Swedish women's perspectives	Identify differences in barriers to and promoters of perinatal health in high versus low utilizing pregnant women of low socioeconomic background.	Gain a detailed and distinctive understanding of the barriers to perinatal care in rural and peri-urban areas of Georgia	Explore the perceptions of Medicaid-insured women about the process of care during the perinatal period	Obtain women's explanations for health-related behavioural changes during pregnancy, and to explore if the influence of maternal-fetal attachment on these behavioural changes	Describe de experiences of migrant women with pregnancy and maternity services at two main hospital	Examine the experiences of racial discrimination during prenatal care from the perspectives of African-American women in a low-income neighbourhood.	To explore the attitudes of first-time mothers towards antenatal education from the perspective of attenders and non-attenders
Design	Qualitative study	Mixed-methods study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative	Qualitative study

Data collection method	Semi-structured interviews.	Semi-structured interviews.	Semi-structured interviews.	Focus groups.	Semi-structured interviews and focus groups.	Focus groups	Focus groups	Focus groups.
Recruitment	Purposive-sampling from one maternity unit.	Purposive-sampling from one maternity unit.	The researchers posted flyers about the study in a variety of locations with distinctly different socio-demographics.	Community health workers recruited women from a program for African American women.	Purposive-sampling from a parenting charity.	Multiple recruitment strategies were used, contact at antenatal care, immigrant centres and swnowballing	Participants were recruited through a community-based organization.	Participants were recruited via their area public health nurse
Consent process	Written informed content	Written informed consent	Verbal informed consent	Informed consent	Informed content	Written informed consent	Verbal informed consent	No info
Number of participants	10 women	40	24	21	9	33	31	16
Age	25-37	Mean age 25 y.o.	Not reported	20-34	26-35	21-40	Not reported	No info
Pregnancy status	1-2 months postpartum	Pregnant	Women had given birth	Pregnant or up to 12	Pregnant or within a mean	Pregnant or up to 12	Women with a child less than 1 year old	Women had given birth within

			within last year	months postpartum	of 8 months postpartum	months postpartum		previous 6 months
Timing of data collection	Spring 2015	March - August 2014	Not reported	Not reported	May 2011	September 2015 - February 2016	Not reported	Not reported
CASP score	16	16	13	15	15	11	18	14

4.4.3 Quality of studies and findings

The utilisation of the CASP tool revealed that most of the included studies scored high when reporting aims, justifying the use of qualitative methodology, and reporting their findings and contribution to the field. However, most of the studies scored poorly regarding ethical issues and reflexivity.

Regarding the assessment of confidence in our review findings using the CERQual assessment tool, we can conclude that most of our findings scored high in level of confidence, and some of them obtained a moderate score. Most findings had minor or moderate concerns regarding methodological limitations. However, the studies contributing to these findings were assessed with the CASP tool and only high-quality studies were included in this synthesis. Hence, as per our judgement, these specific limitations do not have a lot of weight in the overall assessment of the finding using the CERQual tool. Additionally, some of the findings scored moderately regarding relevance since they only applied to a certain part of the population (e.g., specific socioeconomic background).

The limitations applicable to each finding, when relevant, have been exposed in the description of the theme, and can also be found in the CERQual assessment Analysis table (Supplementary file 4.3).

4.5 Synthesis

We were able to identify facilitators and barriers at four different levels within society. In each level, we can find both facilitators and barriers. Figure 2 shows a graphic representation of the themes identified in this synthesis embedded into the Social Determinants of Health Model.

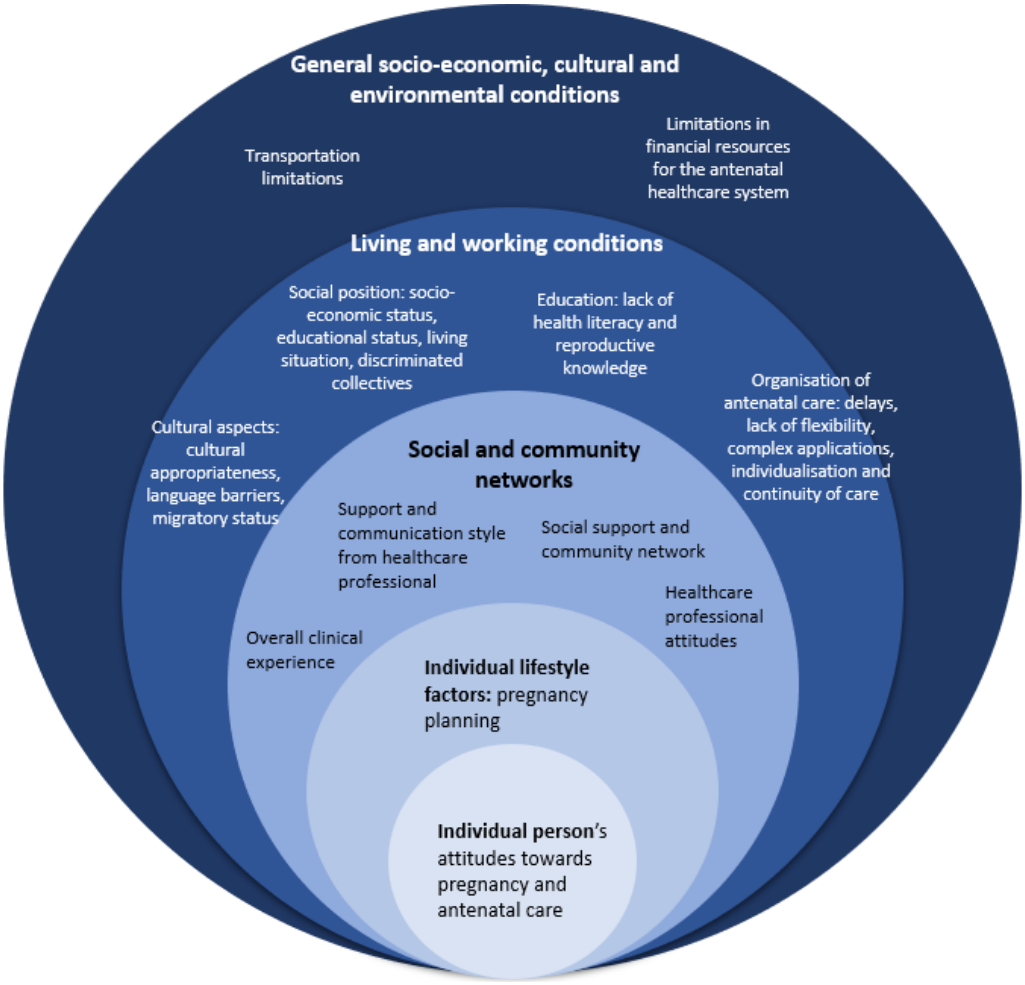


Figure 4. 2: Synthesis findings embedded in the Social Determinants of Health Model[389]

4.5.1 Women's individual factors

As barriers or facilitators associated with individual factors, we understand all those factors that have to do with the individual pregnant woman's cognitive, emotional and behavioural processes as well as her previous life experiences.

Theme 1: Attitudes towards pregnancy and antenatal care

Emotional processes play a role in the decision making around seeking antenatal care. Having feelings of ambivalence towards the pregnancy[390–397], denying the pregnancy[390,392,393,398], feeling overwhelmed or unprepared to be a parent[390,391,396], worrying about social services involvement[390,393,395,396,399,400] and fear[390,401] play a very important role can act as a barrier for women to seek antenatal care. Our analysis identified that not having a planned pregnancy intensified these feelings and acted as an additional barrier[390,391,394,395,398,402].

Nonetheless, there are other types of feelings that can act as facilitators to antenatal care. These feelings are associated with the desirability of the pregnancy. Having positive feelings towards the pregnancy[390,392,394–396], actively planning the pregnancy[392] and conducting actions that promote bonding with the baby[397] were mentioned as facilitators.

Analysis identified there are also certain beliefs and attitudes can act as a barrier for women to access antenatal care as some women underestimate the value of antenatal care[390–396,398,401,402] and hence, reduce their motivation to start a process which can be quite challenging for some. An example of these beliefs and attitudes are related to the understanding that pregnancy is a normal life event[390,393,396,402] and so medical attention is only necessary when feeling unwell[390,401,402] and that women can take care of themselves[390–392,402,403]. Further, certain collectives have a history of mistrust in the healthcare system, with midwives and doctors, and these beliefs also affect antenatal care attendance.

However, certain attitudes and beliefs act as facilitators to access antenatal care. Adopting an active role in their care[390,395,403], viewing antenatal care as an opportunity for behaviour change[394], and understanding the benefits of antenatal care[390–392,394–398,402,403] are attitudes and beliefs commonly reported in early

attenders. Women perceived antenatal care as acting as a preventive measure[395], being a source of knowledge, support and reassurance[391,392,394–396,398,400,402,403], and acting as a distraction and means of socialisation and peer mentoring[391,396]. Additionally, these women also reported high levels of trust in the system and their healthcare professionals[393,395,396] (Please see table 4.2).

Table 4. 2: Theme 1 Attitudes towards pregnancy and antenatal care

Barriers	Facilitators
"Well, I didn't start mine right away 'cause I went straight to the abortion clinic like I said at the beginning I was unhappy, but I didn't kill the first ones so I can't do nothing but stick with it."[392]	Many also created a bond by using more personal methods of feeling connected to the baby, such as naming or talking to the fetus.[397]
"…it actually took me a few weeks to feel anything. I didn't feel anything, because it wasn't planned"[395]	"I go to the fridge…if I saw some cheese in there and I thought I'd quite like it but I can't have it, I'll…say to myself 'well I'm not going to have it because it's bad for the baby', and then in your head you'll be like 'can you hear me baby? I'm not having this and it's because of you!'" [397]
The lack of preparation for pregnancy found in the study not only affected women's 'mindset' and delayed confirmation of the pregnancy, but also led to feelings of fear, depression and ambivalence amongst some women, particularly related to the consequences of the pregnancy,[390]	'You can always feel this presence, and you can almost, it sounds stupid, but bond with the baby, because you feel like you're together.'[397]
A small number of participants specifically noted that their pregnancy was unplanned; these women described they were ''in denial'' about their pregnancy, and therefore did not seek PNC [prenatal care] during	Access to antenatal care is heavily influenced by a woman's willingness to embrace her pregnancy and particularly the social aspects of the pregnancy [390]

their first trimester.[398] An intuitive process of 'do it yourself antenatal care' was reported by some women which included self-checks and active self-care, in order to promote and monitor the healthy progress of their pregnancy, until they felt able to access care[390] "… you are already experienced in giving birth. [ ] you'll know what to do and how to look after yourself"[402] "I had no reason to go. I had no need for it. I just didn't want it.[401] '... booking late is not a big deal to me because, number one [it] is not, is not a disease. I'm not sick or anything so I knew what's wrong with me …'[393] The women's responses suggested rather a differing set of priorities, a consideration of convenience and an assessment of the value of early care, linked to location, health and past experience.[390]	"It would frighten me not to have it [antenatal care]. To go through a whole pregnancy without it. It would frighten me when I'm in the labor room. It's very important."[392] "So your baby will be healthy and normal and all this stuff. Even though something could be wrong you're going through the proper channels to insure you have a happy child. 'Cause I hear stories now from some girls and the things that were wrong with their babies when they were born it's like it scares me.... But prenatal care is very essential to the baby."[392] "Prenatal care, it helps you feel a little bit safer, comfortable about what you're going through. Sometimes we get nervous. You don't know"[395]

4.5.2 Healthcare professionals and health-care system

The attitudes or communication style that one party adopts might have an influence over the other. This theme outlines the specific behaviours or attitudes adopted by healthcare professionals and that might have influenced the women's perception of care and thus, their willingness to engage with the antenatal services. Additionally, within this section we examine barriers and facilitators that are associated with the organisation, functioning or management of the antenatal services or health services providers at an institutional level. All these aspects may also have an influence on the women's perception of their antenatal care process and can contradict or reinforce possible beliefs that she might have had before attending, and therefore, may influence her behaviour.

Theme 2: Healthcare professionals 'attitudes and behaviours

Many women in the studies, especially those from ethnic minorities, reported feeling judged[390,392,393,396] and stigmatised[390,393–395,398] by healthcare professionals attitudes. Further, some racist behaviours[392,394,395,404] and a feeling of inequality between patient and professional[393,403] was also reported in several instances.

Lack of empathy or insensitivity from the healthcare professionals were also reported by several women in the different studies[392,393,395,402]. In some instances, women felt like they were not treated with respect, and that healthcare professionals were very task-oriented[393,395]. When conducting our CERQual analysis, we identified relevance concerns relating to this finding as most of the contributing studies explore women of low socioeconomic backgrounds (See supplementary file 4.3).

However, in other cases, women also reported different attitudes that healthcare professionals adopted to facilitate the antenatal care process for them, these attitudes influenced the women's perception of their carers in a positive way and made them feel cared for and valued[395]. Non-judgemental, honest, supportive and empathetic healthcare professionals were highly valued (Please see Table 4.3).

Table 4. 3: Theme 2 Healthcare professionals 'attitudes and behaviours.

Barriers	Facilitators
"Providers act like I don't know anything just because I am poor. I want to learn: didn't get nothing out of it; keep repeating; feel like I'm not smart enough to ask questions"[395]	"I mean, I just feel that they relate to a lot of things. You know say you going through things that you're dealing with, and when you find yourself in, you know, some type of crossroad, they help you, you know, try to navigate your situation. I mean, I just think they go above and beyond their duties"[395]
While examples of obvious behaviours (such as use of racial slurs) were described in some groups, other groups described more subtle forms of racism (such as providers' negative assumptions about them).[404]	
"She just came in and it's like she is so used to it and you are just another pregnancy, so she doesn't see you for an individual. She just does her thing and leaves"[395]	

Theme 3: Communication with healthcare professionals

Communication issues in different forms were reported through all the studies. Women reported receiving contradictive messages[395,396,402], having concerns dismissed[395,403], feeling uninformed[395,398,401,403], being confused due to the use of jargon[395,403] and feeling frustrated since healthcare professionals would not do any prior health knowledge assessment[403]. This poor communication not only might have altered the women's subjective perception of their carer but also of the whole antenatal process and in some instances, it might have led women to disengage and lose interest in their antenatal care. Regarding the CERQual assessment of this findings, we encountered concerns regarding adequacy as only three studies strongly support this finding, however, there are others that provide limited data to support it (See supplementary file 4.3).

However, according to the women, other practices that healthcare professionals adopted promoted positive communication and also helped women through their antenatal care process. Healthcare professionals who engage in active listening and took time to address women's concerns[391,398] and empower them to participate in the decision-making process[391,403] were seen as very encouraging by women and facilitated trust (Please see Table 4.4).

Table 4. 4: Theme 3 Communication with healthcare professionals

Barriers	Facilitators
"She did not spend a lot of time in the room. It did make me feel like you couldn't get your questions in. They immediately answered and then shoot you down. You know, when you are a pregnant woman, you are sincerely concerned about this health problem, because you are experiencing something that you feel is not normal and for it to just be immediately dismissed, it makes you feel like well, why did I even say anything"[395] ''They didn't really go over what to expect or what to do when this happens. They didn't really go over how the day would be, I guess.''[398] "The woman that we spoke to, she was going on about you know about protein in your urine or whatever and all this stuff and I just didn't have a clue what you're talking about. It is all very, I know they must do it all the time.[403]"	'The way she talked, the way she attend to you, you understand. That make you feel ... really feel good, you know.'[393] "She told me just about basically the different options of like how to have your baby about whether you wanted consultant led or midwife led and explained the difference between the two… so I kind of made a choice that I was just going to go with midwife led[403]" For engagement to be present we sought evidence of a case perceiving good communication with health professionals, perceiving a value in the quality and relevance of educational information and evidence of shared decision making[403]

Theme 4: Unsatisfactory clinical experiences

Some women included in these studies changed their behaviour after being exposed to antenatal care [392–394,396] (Please see Table 4.5).. Having an unsatisfactory clinical experience was reported by several women as a barrier to engage with the antenatal services [392,394,395,403]. In this instance, women had the motivation to access antenatal care but the subjective perceptions that followed their negative experiences reduced their levels of motivation leading to disengagement. Some of these negative perceptions were formed after experiencing rushed appointments[395,403], feeling that positive care was conditional on insurance status or sociodemographic factors[394,396], feeling excluded from the decision making[398,403] or feeling that the whole process was a waste of time[395]. Regarding the CERQual assessment, we attributed minor concerns in terms of relevance to this finding as most of studies contributing to it focus on women from low socioeconomic backgrounds (See supplementary file 4.3).

Table 4. 5: Theme 4 Unsatisfactory clinical experiences

Barriers
"It was kind of a waste of time, to sit there all that time, and then, you know, be rushed out; pretty much I didn't get anything accomplished with that. You know, I guess it's okay to know that your baby is doing fine or whatever"[395]
Most indicated the provider was not in the room long enough, they didn't get enough information, sometimes did not get questions answered, and often ended up feeling like they were leaving with nothing.[395]
Another mother regretted not discussing her desire for a vaginal delivery prior to labor, and said, ''It's something I would have appreciated more discussion about prior to the moment when that needed to happen. […That] decision was made really kind of around me, as opposed to with me, which was frustrating.''[398]

Theme 5: Organisation of antenatal care

The analysis shows that in some cases, women had the motivation to seek antenatal care and understood its value, however, they found several institutional barriers in their way that delayed their booking date. Examples of this are administrative delays[390,393,395,398,401] or problems with the referral process[393,395,402]. Other women reported that the lack of flexibility of the appointments[391,394,395,399] meant antenatal appointments clashed with their other commitments, which impeded their access. Women reported that they would have preferred a system that allowed for more flexibility or walk-in services[394], and in cases where this was available, it was reported as a facilitator for antenatal care engagement.

Some women found that the application to access antenatal care was too complex and required a lot of effort[395,398,400–402,404]; in this case women were willing to access antenatal care early but the application process itself was delaying them. This issue was especially relevant in the studies conducted in the US, where women needed pregnancy confirmation from a provider to be able to access the government insurance program Medicaid[395]. Other women reported not having enough information to access the different services available[399,401–403]. On the other hand, availing of extra professional support for help though this process was highly valued by some women[395].

Some women also reported that there was a lack of individualised care within the antenatal system. Women felt part of a production line, *"just another pregnancy"*, and in some instances, felt that their individual needs were not met[390,393,403].

Lack of continuity of carer[393–395,398,400] was another issue frequently reported by the women participating in the different studies. This lack of continuity in some instances related to an interruption in insurance coverage[394,395,398], resulting in frustration for the women affected. For some women, not seeing the same healthcare worker in each visit hindered their ability to build rapport and open up to them. This drove some women to avoid antenatal care services and attend their general practitioners only for their care. The studies showed that providers who facilitated continuity of carer [393,396,398] prompted better trusting relationships and promoted engagement with antenatal care [393,398] (Please see Table 4.6).

Table 4. 6: Theme 5 Organisation of antenatal care

Barriers	Facilitators
"It does take a long time because they don't take women without a GP at the hospital, so to get to the hospital she needed to get a GP, register with a GP, get an appointment and for her to be referred to the hospital it takes time. GP requires proof of address, some bill or something. And she did not have that address because they moved just recently so she was waiting for proof to come". [402]. "I think that with the Welfare, you have to have predetermination, and I didn't do something right. I went to the doctor's office, and they couldn't see me because I didn't know how to get on Medicaid until I got there. Then I had to reschedule and wait for them to get like the verification that I had it"[395] Women felt powerless to get early PNC, even when some reported that they had a prior high-risk pregnancy to the person scheduling appointments. [395] '... the second time I went it was a different doctor that was probably a locum or something and then the third time I went it was another doctor … and that's three times.' [393]	"I didn't call. I just figured since I'm around this area and I'm feeling that pain for 2 days now, why not just come over."[394] "My SB CHW connected me to everything else that I needed and she would just call me, like sign her up (daughter) and everything I needed. When we were into the appointment, she would like call the person and do this step, doing the work, everything"[395] "The only reason I did—I got in [accepted for Medicaid] is because my CHW was on my case. She was just e-mailing like, you know, hey, what's the hold up"[395] All of the mothers and key informants described the importance of seeing few providers consistently throughout their pregnancy to provide continuity of care.[398]

4.5.3 Social environment

As previously mentioned, the barriers identified after analysing the included studies affect the process of antenatal care from different levels in society. In this category, the barriers identified are related to the general socio-economic, cultural and environmental context where a woman lives.

Theme 6: Cultural aspects

Women from different cultural backgrounds reported issues related to cultural inappropriateness[401,403] or language barriers[390,399,400,402]. Once again, these issues hindered the possibility of forming a trusting relationship with their carer, which in many instances led women to seek care in their own communities or to travel home for their care [390,402].

Some of the studies included in the analysis looked at experiences of migrant women in high income countries. Women felt discriminated against due to their status as an immigrant [399,400], and in some cases, felt the need to justify their right to care[399].

Women with an illegal status were afraid of being deported if they attended antenatal care, and they were also afraid of being separated from their babies[399].

Due to their illegal immigrant status, some women had uncertainties about their ability to afford care, which led them to believe that they only should access care in emergencies or in labour[399]. Additionally, the lack of familiarity with the health system within the country increased their difficulties in accessing care[399,400].

Women found that availing of health clinics held specifically for immigrant women facilitated their access to care[399,400] as they felt safe there. Many women, were entirely dependent on these clinics as their only resource for their care, which was a source of concern [399].

In many instances however, these health service-related barriers were possibly consequences of reduced funding or resource limitations. In several of the studies, women reported interruptions in their care due to closure of their clinics or a delay in their booking due to a lack of providers in their area. These issues correlate with

the social environment in which those services are being provided (Please see Table 4.7).

Table 4. 7: Theme 6 Cultural aspects

Barriers	**Facilitators**
". . . but if the midwife in at the hospital says, "You are not Danish. you can go home or pay" I will think: I am not Danish, but I need help with my baby. I am a human being"[399]	"Honestly, they are so friendly here, and it's so nice to see that people really want to help you, even though you are foreign . . . and even though you don't have residency. You don't have nothing, but you have the clinic. And know that they won't report you to the police. It feels very safe"[399]
"I thought I was going to die. It didn't come abruptly. I felt more and more pain . . . But I knew we could not go to the hospital. But when I reached the point of thinking it's hospital or death, we went. I couldn't be deported if I was dead, you know. Dead people don't have passports"[399]	"...I solely depend on this place. If I could not go here, where would I go?"[399]
The women feared that the debt could increase their risk of deportation or affect their chances of attaining residence permits in the future. It also caused worries of whether they would be refused care during birth or be given inadequate care. Furthermore, they worried whether they would end up with unmanageable bills afterwards	

Theme 7: Social position and lifestyle factors

The social position and lifestyle factors are defined here as all those aspects associated with a specific way of living or socio-economic status. By social position or economic status we consider factors such as income, access to education, occupation, living conditions, access to health services and access to housing that will have an impact on a person's lifestyle[405].

Aspects like homelessness[392], intimate partner violence[391,394], living in deprived areas[403] or having low economic resources[390,392–395,401] have been identified as barriers for antenatal care attendance. Most of the time, women living in these conditions do not have their basic needs of survival covered and need to focus their efforts in their more immediate concerns[391,394,402,406]. Therefore, seeking antenatal care is not a priority for them[391,394,395,401,402].

Additionally, the analysis showed that the social discrimination of certain collectives within the society also acted as barrier for many women. It was reported in different studies that teenage women[390,398], substance users[390], women from ethnic minorities[392,394,395,404] or women from lower socioeconomic status[404] faced additional challenges when trying to access antenatal care. In some instances, women felt their care was of lower quality based on their insurance status[395,398,404].

Conversely, there are some lifestyle factors or social position related factors that act as facilitators to access antenatal care. Having a higher educational level[392,403], a higher economic status[392,403], a steady living situation[392], being older[392,403] and being a first-time mother[390] are factors associated with a better attendance and compliance with antenatal care. Women in more favourable socioeconomic positions are more likely have less immediate concerns and so they can focus on seeking care during their pregnancy. Furthermore, women with greater access to education and information might be more aware of the different risks during pregnancy and the importance of antenatal care (Please see Table 4.8).

Table 4. 8: Theme 7 Social position and lifestyle factors

Barriers	Facilitators
"I was mainly worried about my children eating, I didn't worry about myself."[394]	The early initiators were older, with an average age of 24 (ranging from 21-36), had higher household income [392]
"It hinders work, to miss work could be a problem when there's not enough money if you miss work."[401]	Women who reside within areas of low socioeconomic deprivation may be more likely to engage with the antenatal service.[403]
"It's difficult because I don't have a job. I get money from Welfare. I get food stamps. I get WIC. I've had plenty of times where it's been times where I've had no electricity. Right now, I need a hot water heater. I still need to figure out how to make arrangements with the water company and the gas company."[394]	
For example, one woman began to describe the better clinic that her friend (who had private insurance) was able to attend, but that, because they were the only Black girls in the waiting room, ''we waited longer than anybody else.''[404]	
"It's not gonna change. It's not, cause it's been like this forever. You learn to go last. No matter how much people talk about it, it is the same…. yes it is, it's been like that forever…That's how it is.[404]"	

Several mothers felt they had access to fewer providers and medical technologies than women with private insurance, which decreased their overall satisfaction with their PNC experiences.[398]

Theme 8: Social support and community network

In this theme, social support is understood as social external influences that might have an influence on a woman's ability and willingness to change her behaviour, and that can also accentuate or alleviate the influence of other types of barriers. These people could be either family, partner, members of the community or professional aids assigned to them.

An absence of social support heavily influenced women's decisions to attend antenatal care[390,392,394,399–401]. In some cases, this lack of support was due to fear of disapproval within the woman's community[392] or family[390,392]. According to Haddrill (2014), it is necessary that women obtain a certain level of approval within their social circle before they can accept the pregnancy and hence, seek antenatal care[390].

On the other hand, availing of social support from the baby's father, the community and the family[390,392,394] combined with a positive reaction facilitates the process of acceptance for the pregnant woman and promotes engagement (Please see Table 4.9).

Table 4. 9: Theme 8 Social support and community network

Barriers	Facilitators
"Like I said, I just moved to Phoenix, they (family) didn't really give a shit. It's just me and my problem for me to deal with, nobody else." [401] "I'm the only one of my friends that got kids. They mad at me right now. They do not want me to have no more kids. They're like 'Give it up.'" [392]	"The girls at my other job they are so supportive about the situation 'cause they know I really didn't want to have any more kids, but they are very supportive, and my mom and stepmom very supportive and my stepmom had her first child at 13. My stepmom, she's a very good listener. Very good for advice.[392]"

Theme 9: Transportation issues

Availability and cost of transportation[391,395,398,399] to attend antenatal care was also reported as a barrier. Some women reported not having any means to access their providers, and even those who were provided with private transportation like taxis, reported issues with the drivers and delays that made them miss their appointments[395] (Please see Table 4.10).

Table 4. 10: Theme 9 Transportation issues

Barriers
"It's just a process that goes along with getting there. Either driving, getting on the bus, something. A lot of the times I can't drive myself and I so then I have to park so far away because every spot around here is like you have the pay the meter or if you park in the hospital parking lot you have to pay them and that's an issue for me, having to pay for everything."[394]
"Didn't have someone to take me."[401]
This was particularly frustrating for women without the transportation, time, and money to travel to their provider's new location[398]

Theme 10: Lack of health literacy and reproductive knowledge

A lack of health literacy or lack of reproductive health knowledge and lack of access to knowledge was widely reported. Many women in the studies reported a delay in the recognition of their pregnancy[390,392,393,395,398] due to different reasons such as mistiming of the pregnancy[390], lack of knowledge of pregnancy signs and symptoms[390,392,393], attributing symptoms to other issues[390,395], having a history of irregular periods[390,395], not expecting the pregnancy[390] or not noticing the symptoms[390]. Contrarily, some women believed it was not possible for them to be pregnant due to their age [390] or due to using contraception[390,395] (Please see Table 4.11).

Table 4. 11: Theme 10 Lack of health literacy and reproductive knowledge

Barriers
"I just started throwing up. I just thought I had an upset stomach, so I waited for about 2-3 months and then I finally went to the hospital.[392]"
"Last time, it was just weird because I went for the Depo and they told me I was 25 weeks pregnant. I got caught on the Depo and I didn't know that, and I took the pills and I got caught on the contraceptive pill this time… and I didn't know with this one". [390]
"When they (the GPs) said 'I was going through the change' I thought 'well could I be' because at 37 I thought 'well I might be' because you hear women go through it earlier than I did and I think I got that into my head more than anything and I never contemplated that I was pregnant".[390]

Theme 11: Government level issues

By government level issues we understand barriers that might be derived from decisions made by governing institutions, policy makers or funding agencies. In this theme, the studies analysed showed that the biggest barriers were the limitations of financial resources for the antenatal healthcare system that explains a lot of the

healthcare system related barriers (e.g., small clinic capacity[394], shortage or closure of clinics[398] or overload of clinics resulting in long waiting times[392,394,395] and the limitations associated with the government health programs and costs of care[395,398,399,401,404].

On the contrary, it was also drawn from the studies that supporting women with the cost of antenatal care through government aids or by availing of free care for everyone was a facilitator to engage in antenatal care.

4.6 Discussion

The results of the synthesis identified facilitators and barriers associated within four different levels which we considered relevant to whether women seek and engage in the process of antenatal care. Firstly, we identified facilitators and barriers associated with the women's individual factors. The women's perceived value of antenatal care and their perceived clinical experiences during their current or previous antenatal care have an influence on their willingness to attend. The women's feelings towards their pregnancy and the idea of parenthood also played a role in their attendance behaviour. Secondly, we identified facilitators and barriers associated with individual healthcare professionals. According to the women's accounts, certain attitudes, and communication styles that healthcare professionals adopted had an impact on their antenatal care engagement. Thirdly, facilitators and barriers associated with the health care system were discussed. These barriers and facilitators relate to the complexities of the system itself, with women finding barriers associated with the organisation of the system and their own difficulties to navigate through it. Finally, we identified factors associated with the social environment, such as socioeconomic position, availability of social support and transportation, and lack of health literacy, that have an effect on women's attendance at antenatal care.

Although the facilitators and barriers identified in this study have been divided into different themes, they should not be considered as separate systems. On the contrary, the influences they have over each other are crucial to understanding the true dimensions of this issue. The reasons why women might not attend or engage

with antenatal care are multifactorial and the personal barriers identified are only one such aspect. Sociodemographic factors, geographical differences and limitations within the healthcare system can determine women's access to knowledge and ability to make informed decisions. These factors also influence beliefs and perceptions about the antenatal care process, ultimately affecting their behaviour.

In many instances, these attitudes, beliefs or perceptions are the result of a lack of information, limited health literacy or the influence of other people's experiences. Health literacy is associated with poorer health outcomes and poorer use of health care services in general [407,408]. This is also true for pregnant women; a study conducted by Kohan concluded that women with higher levels of health literacy were more likely to attend antenatal care early and frequently, and also had healthier outcomes for themselves and for their babies [409]. Similarly, previous research from our group has demonstrated that the level of health literacy and awareness that women have about a certain issue can have an impact on their engagement in behaviour change [410]. Previous studies have demonstrated that peer-led interactive interventions are more effective than lecture-style information provision in terms of increasing health literacy [411,412]. These elements should be considered when designing interventions targeting all women in a community. Further, involving women's voices in the development of interventions is also crucial to ensure that their needs are met. For example, Satwell et al. have designed an intervention to overcome some of the barriers identified in this study, including lack of health literacy, by co-producing an intervention with the involvement of their community to reduce the high rates of late initiation of antenatal care in the UK. The intervention is co-produced and locally tailored to each participating site[413].

The coronavirus disease (COVID-19) pandemic has forced the world to adapt in many ways, including provision of antenatal care. Some of the necessary antenatal care was delivered via online care programs. Online antenatal care could prove beneficial for those women who encounter economic, logistic or other types of external barriers to attend antenatal care at a healthcare facility, as it may be more convenient and cost-effective[414]. One way of tackling some of the barriers exposed in this article would be by providing high-quality antenatal care programmes, but to

do so, health services need to have skilled professionals and technology available, and governments should ensure access to electronic devices and relevant resources to their population. However, further work is necessary to optimise systems, as recent research has shown that women might find telehealth problematic and was less favoured than in-person care[415,416].

Our findings show that healthcare professionals should receive adequate education through their training programmes, and National Antenatal Education Standards published by the different Health Services should include an element of the importance of discussing health habits and risk factors for stillbirth or other potential adverse outcomes. Further training and guidance might encourage healthcare professionals to engage in these discussions with their patients with a higher level of confidence.

These results can be explained using the Social Determinants of Health model[389]. This model takes into account the impact of social and political conditions over health[417]. Aspects like the conditions in which a person is born and raised, availability of social support and community networks, access to education, job opportunities, housing, water and food supplies, access to health services, etc. have an impact on health outcomes[405,417,418]. Dahlgren and Whitehead developed a conceptual framework of the Social Determinants of Health mapping out the different categories that relate to public health[405]. This framework suggests different levels of intervention, ensuring that all relevant determinants of health are targeted when addressing any specific health issue. Hence, in order to intervene effectively, all levels need to be considered consistently while avoiding contradictive policies at different levels[405,419].

In the Social Determinants of Health model, Dahlgren and Whitehead positioned the constitutional/individual and lifestyle factors as the smallest arch of their rainbow[405]. These would correspond with facilitators and barriers related to the women's individual factors identified in this study. Although these personal factors, such as beliefs, capabilities and experiences, need to be set in their wider social context to be fully understood, they also play a role in health behaviours and subsequent health outcomes[420]. Therefore, the individual also needs to take responsibility and policy should be a tool to support the individual's personal autonomy when engaging with

well-informed decision making about their own health, without reaching a victim-blaming extreme[421]. The last three levels represented in the Social Determinants of Health model relate to social and community network, living conditions and general socio-economic, cultural and environmental conditions. We were able to identify factors at each level (See figure 2). Issues related to social position, cultural aspects, education, transportation limitations and lack of resources are responsibilities of local and national governments, and further work is necessary to address inequalities impacting women's access to antenatal care across all levels.

Better health outcomes may be achieved by a combined effort between society and the individual[421,422]. To support this purpose, co-production is an approach that has been applied to provision of public health services. By co-production we understand the direct user involvement in the production of research giving pleople with lived experiences an active role in all aspects of the research process and sharing decision-making power equally between researchers and stakeholders[423,424].

Another relevant approach is including Patient and Public Inovement (PPI) in research design. *This approach allows the incorporation of the perspectives, views and experiences of patients and the public into the different aspects of the research. However, within this approach the PPI representatives participate in specific aspects of the research and have different levels of involvement, and generally, the decision power resides with the researchers*[425].

Co-productive and public and patient involvement approaches can be used to obtain insights into the quality of care perceived by the users of a health service and improve that quality based on the insights obtained[426]. Some of the techniques that can be used feedback meetings, shadowing, using patients as educators, and experience based co-design, where patients and healthcare professionals work together for a period of time to co-design a service[426]. Haddrill et al. conducted studies to ask their participants about ways to facilitate attendance or engagement with antenatal care. The responses given by women included providing a non-judgemental contextually tailored service, better promotion and education about the value of early antenatal care[390], better pre-conceptual reproductive education, improved communication between services and simplifying the referral process[390,402]. This information

obtained by the women involved in antenatal care, in addition to insights obtained by healthcare professionals, could be part of a co-productive process to target those barriers that prevent women from attending antenatal care.

Our study has several limitations. First, our systematic review of the literature relied on a limited number of databases. These databases were chosen based on similar studies in the area, previous experience, and knowledge about the databases.

Further, we excluded studies based on quality assessment. Although this has enabled the findings from our study to be of a higher quality, excluding studies due to quality criteria can result in a loss of relevant input. However, previous research in the area of quality appraisal has shown that the quality of reporting of a study correlates with its value as a source for the final synthesis, and therefore it is appropriate to exclude inadequately reported studies[383].

The findings obtained in this study are based on interpretations of women's perceptions and beliefs, and therefore, they cannot be considered as facts. Additionally, by choosing not to include the experiences of other agents involved in the antenatal care process, we are not able to fully explore the issue from different perspectives. We considered that hearing the women´s voices is crucial in order to develop appropriate interventions. Nonetheless, further research should be undertaken to explore other stakeholders' experiences and opinions.

Finally, our findings are not based on primary data but on a secondary analysis of these women's views, opinions and experiences. This means that the original author's own views and interpretations should be acknowledged as potentially having some influence over our findings. However, the findings across the studies were consistent despite being conducted by different authors in different parts of the world, which strengthens our own findings.

The results of this study show that to tackle the issue of non-attendance at, or delaying access to, antenatal care, interventions need to be designed at different levels. Trying to increase the intrinsic motivation of women through tailored interventions that tackle the different personal barriers to antenatal care and working with healthcare professionals towards using better communication

strategies could result in a reduction of the barriers identified in this study. This issue needs to be considered by policy makers not only in terms of health, but also in terms of education, employment, housing and social equality. Moreover, the role of the individual cannot be forgotten. A co-production approach should be used where the social determinants of health are addressed to enable the individual to enhance facilitators and overcome barriers to securing better antenatal health[421].

4.7 Inter-chaper synopses

Chapter 4 aimed to identify the facilitatos and barriers influencing women's behaviour when deciding to seek and engage with antenatal care during pregnancy. The study identified several facilitators such as having a positive attitude towards antneatla care, social support, and having access to reliable information. However, multiple barriers were also identified including having negative attitudes towards the pregnancy, encountering communication issues or having unsatisfactory clinical experiences, as well as having to surpass organisational, institutional or social barriers to access care. The study emphasizes the need to address these barriers to improve access to and engagement with antenatal care services, which can lead to improved maternal and fetal outcomes. Chapter 5 had a similar aim to Chapter 4, but in this case, the behaviour of focus was substance use. Before conducting the study in Chapter 5, it was well described in the literature that smoking is one of the risk factors with the strongest evidence associated with the risk stillbirth[134], and, even though the evidence regarding alcohol and illicit drug consumption is a bit conflicting, the recommendations are also to avoid such consumptions during pregnancy in order to reduce the risk of stillbirth. Hence, further understanding of the influences affecting these behaviours were necessary to inform the development of future behaviour change interventions.

Chapter 5 - Facilitators and barriers to substance-free pregnancies in high-income countries: a meta-synthesis of qualitative research

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Chapter 5 - Facilitators and barriers to substance-free pregnancies in high-income countries: A meta-synthesis of qualitative research

5.1 Abstract

Background

Previous studies have associated substance use (alcohol, illicit drugs and smoking) to negative pregnancy outcomes, including higher risk of stillbirth.

Aim

This study aims to identify facilitators and barriers reported by women to remain substance free during pregnancy.

Methods

A systematic search was conducted in six databases from inception to March 2019 and updated in November 2020. Qualitative studies involving pregnant or post-partum women, from high-income countries, examining women's experiences of substance use during pregnancy were eligible. Meta-ethnography was used to facilitate this meta-synthesis.

Findings

Twenty-two studies were included for analysis. Internal barriers included the perceived emotional and social benefits of using substances such as stress coping, and the associated feelings of shame and guilt. Finding insensitive professionals, the lack of information and discussion about risks, and lack of social support were identified as external barriers. Furthermore, the social stigma and fear of prosecution associated with substance use led some women to conceal their use. Facilitators included awareness of the health risks of substance use, having intrinsic incentives and finding support in family, friends and professionals.

Discussion

Perceived benefits, knowledge, experiences in health care settings, and social factors all play important roles in women's behaviours. These factors can co-occur and must be considered together to be able to understand the complexity of prenatal substance use.

Conclusion

Increased clinical and community awareness of the modifiable risk factors associated with substance use during pregnancy presented in this study, is necessary to inform future prevention efforts.

5.2 Introduction

Prenatal smoking, consumption of alcohol and illicit drug use is a significant public health concern with important implications for women and infants[427]. Smoking, consuming alcohol and using illicit drugs during pregnancy are well established risk factors for a wide range of adverse pregnancy outcomes[341]. These include an increased risk of miscarriage[428], ectopic pregnancy[429], placental pathologies[430], low birth weight[431], small for gestational age[432], intrauterine growth restriction[433], preterm delivery[432], fetal alcohol syndrome[434], and neonatal death[428]. Prenatal smoking, alcohol consumption and illicit drug use are also associated with stillbirth[134,428,435].

The occurrence of stillbirth differs across countries. In 2008, 2.65 million stillbirths were estimated worldwide, with 98% of them occurring in low-income and middle-income countries[2]. In 2015, the rates of stillbirth per 1000 births at 28 weeks gestation or more in high-income countries varied from 1.3 in Iceland to 8.8 in Ukraine[6]. However, it is difficult to compare rates of stillbirth amongst countries due to the variability in the definitions used and the under-reporting of stillbirths under 28 weeks gestation.

In a systematic review and meta-analysis of 142 studies published in 2015, any active smoking and second-hand smoke exposure during pregnancy were associated with a higher risk of stillbirth (Standardised Rate Ratio (sRR) 1.46 and sRR 1.40 respectively)[428]. Previous studies have also associated heavy consumption of alcohol with an increased risk of stillbirth[310,436]. Aliyu et al. concluded in their retrospective cohort study with more than 650,000 pregnancies, that the likelihood of stillbirth between 20 and 28 weeks gestations was higher in women who drank alcohol gestation[310]. According to this study, women consuming five or more drinks per week during pregnancy experienced a 70% increase in the risk of stillbirth. Women consuming 1-2 drinks per week had a hazard ratio of 1.5 (95% confidence interval 1.0-2.1), no increase in risk amongst women drinking 3-4 drinks per week, and a peak increase in women drinking 5 or more drinks per week. In terms of illicit drug consumption, Varner et al. concluded that a positive test for any illicit drug in the umbilical cord homogenate was associated with an increase in the risk of stillbirth at

20 weeks gestation (OR 1.94)[437]. A recent meta-analysis reported that the global prevalence of smoking during pregnancy was estimated to be 1.7% [137]. This study concluded that the highest prevalence of smoking during pregnancy was in the European Region (8.1%) and the lowest in the African Region (0.8%)[137]. The five countries with the highest estimated prevalence of smoking during pregnancy, regardless of frequency or quantity, were Ireland (38.4%), Uruguay (29.7%), Bulgaria (29.4%), Spain (26.0%) and Denmark (25.2%)[137]. However, previous studies have reported variations in prevalence, such as a prevalence of 11 to 17% of smoking among pregnant women in Ireland[438–440]. Prevalence of prenatal alcohol consumption has been found to be higher than that of smoking. A systematic review and meta-analysis published by Popova and colleagues concluded that, globally, 9.8% of women consumed alcohol during their pregnancy[143]. The highest prevalence was observed in the European World Health Organization Region (25.2%), whereas the lowest was in the Eastern Mediterranean WHO Region (0.2%). The five countries with the highest estimated prevalence of alcohol use during pregnancy were Ireland (60.4%), Belarus (46.6%), Denmark (45.8%), United Kingdom (41.3%) and Russia (36.5%).

Cocaine, amphetamines, opioids, marijuana, hallucinogens and toluene-based solvents are the illicit drugs that have been found to be most commonly used by pregnant women[149,150], however, establishing the prevalence of illicit drug use in pregnant women may be more difficult than for alcohol and smoking. For example although social stigma can exist for all of these behaviours during pregnancy, legal repercussions associated with substance use might drive some women to avoid disclosure[150,441]. However, in the 2010 National Survey on Drugs and Health conducted among pregnant women aged 15 and 44 in the USA, it was reported that 4% of pregnant women were current illicit drug users[442].

Understanding women's attitudes, perceptions and experiences of substance use is essential to obtaining insight into the facilitators and barriers that modulate these behaviours during pregnancy. Qualitative research is a useful approach in this regard to explore and understand the nature and interaction of the different layers of the studied phenomenon while maintaining the particular complexities of human

behaviour[443]. To date a number of primary qualitative studies have been conducted to understand women's prenatal smoking, alcohol and illicit substance consumption[444–453] and we found three meta-synthesis exploring experiences of women who commence pregnancy as smokers[454–456]. However, to date, no prior meta-synthesis has been conducted related to the other types of substances used during pregnancy.

The aim of this meta-synthesis is to analyse and synthesise all of the evidence drawn from qualitative research to date in order to identify facilitators and barriers to a substance-free pregnancy in developed countries. This will facilitate developing in-depth insights and understandings of these prenatal health behaviours[457,458]. By identifying barriers and facilitators common to different type of substances, we intend to inform the development of a behaviour change intervention applicable in high-income countries.

The synthesis of qualitative research was informed by meta-ethnography. Meta-ethnography is a methodology originally developed by Noblit and Hare with an interpretative approach[386]. This methodology is utilised to "put together" all the available research through the translation of qualitative studies into one another[386]. In order to do this, the researcher translates the studies into one another's terms, and into their own interpretation of the data and the world, which will result in a synthesis that is partially produced by the author[387]. To facilitate the reporting of this meta-ethnography, we followed The eMERGe reporting guidance[385] (See supplementary Table 5.1). We chose meta-ethnography because of its potential to produce new interpretations, models or theory[459].

5.3 Methodology

5.3.1 Search strategy

A comprehensive systematic search of the literature was performed for all qualitative research that explored women's facilitators and barriers to abstain from substance use during pregnancy. The databases searched were CINHAL, PsychINFO, Pubmed, SOCindex and Web of Science and the searchers were conducted on the 28th and 29th

of March 2019 with no restrictions on publication date. The search was updated on the 25th of November 2020 to identify new published articles relevant for our synthesis. Further, the reference list of each included study was hand-searched for additional studies.

The protocol for this meta-synthesis was registered on Prospero (no. CRD42019120069). Originally this meta-synthesis had the objective to provide insights into different modifiable risk factors for stillbirth. However, due to the high volume and complexity of the qualitative research on antenatal behaviour practices, this meta-synthesis differs from the original protocol in that it focuses specifically on substance use during pregnancy rather than substance use, attendance at antenatal care and weight management as previously planned. Findings in relation to the other two modifiable risk factors will be published elsewhere.

Search terms were selected based on a preliminary scan of the relevant literature. The search terms used were *facilitators, barriers, promoter, benefit, attitude, opportunity, determinant, promotion, intention, education, initiative, prevention, pregnancy, smoking, smoking cessation, nicotine, alcohol, alcohol abuse, alcohol drinking, drug*, drug abuse, illicit drugs, oral drugs, intravenous drugs* (see example of search in Supplementary file 5.2).

5.3.2 Study selection

Three members of the research team (SM, LL, TES) independently reviewed the titles and abstracts of the studies resulting from the database search. Additionally, two authors independently (SM, TES) conducted the full text screening of the eligible studies.

Studies were included for further review if (1) they used a qualitative or mixed methods design, as long as they included primary qualitative data (2) they were written in English, (3) the participants were women interviewed when pregnant or up to 12 months post-partum as long as the data referred to their experiences during their pregnancy, (4) they were conducted in high income countries, and (5) included extractable data about facilitators and barriers to remain abstinent from substance abuse during pregnancy.

Studies were excluded if they did not include any qualitative data or if they were not original research. Studies that included different types of participants (e.g., healthcare professionals and pregnant women, partners and pregnant women) were only included if the data extracted from the pregnant women was differentiated from the rest. In this meta-synthesis, we included studies that explored the views of women who were current substance users during their pregnancies and women who made the choice to remain abstinent during their pregnancy.

5.3.3 Data extraction

Study characteristics

A data extraction sheet was used to extract the characteristics of the studies by one author (TES). The following data were extracted from each study: country of publication, year of publication, aims, design, data collection method, sampling or recruitment strategy, consent process, number of participants, age of participants, pregnancy status, timing of data collection and method of data analysis.

Quality Assessment

The *Critical Appraisal Skills Program (CASP)* for qualitative studies was used independently by two authors (TES, SM) to assess the quality of the studies. Previous research in the area of quality appraisal has concluded that there is a correlation between the quality of reporting of a study and its value as a source for the final synthesis, and therefore it is appropriate to exclude inadequately reported studies[383]. As a result, it was decided that only the studies with the highest quality (CASP ≥ 15) would be part of the synthesis.

To assess our individual review findings, we used the GRADE-CERQual approach. This approach facilitates assessment of how much confidence can be places on individual review findings from a synthesis of qualitative research[384].

Synthesis informed by meta-ethnography

Noblit and Hare (1988)[386] proposed a series of phases that overlap and repeat along with the conduction of the synthesis (see Supplementary File 4.4).

Phase 1 and 2 – Selecting meta-ethnography and deciding what is relevant

The first two phases of meta-ethnography involve identifying a research gap which has the potential to be filled by meta-ethnography, explaining the rational for using meta-ethnography and stating its purpose and focus, which we have done in the sections above. Phase 2 was completed by conducting a systematic search of the databases as outlined above, and importing the resulting studies into NVivo12 for analysis (see Figure 1).

Phase 3 - Reading the studies

During Phase 3, the papers were read in depth several times and the characteristics and details of the papers were extracted and tabulated into the data extraction sheet (Supplementary Table 5.3). In this phase, each paper was read carefully and notes were used to identify the data that had to be extracted.

Phase 4 - Determining how the studies are related

Phase 4 involved the line-by-line coding of each selected study by one author (TES) using Nvivo12 in order to identify common metaphors and concepts, consulting with the rest of the team when in doubt. We refined the codes in our list and created new ones as the coding progressed in each new study. Every sentence of the studies had to be coded at least once. A second author (SM) coded a sample of the papers to facilitate reliability and validity in the coding process. Both first and second order constructs were extracted for analysis. We utilised second-order constructs to complement the primary data, which offered additional insights, context and explanations. Most of the studies had poor reflexive accounts, which hindered our assessment of the author´s background influence over their interpretations. The key concepts explored and compared in the analysis were related to factors that either facilitated or hindered access to antenatal care for pregnant women in high-income countries.

Once the initial coding was completed, we examined the text added to each code for consistency of interpretation and additional coding was performed when necessary. We obtained a list of concepts that were grouped into themes and categories using thematic analysis.

We used tables to display the concepts and themes across all studies, classifying them depending on the substance they were exploring, this helped us see which concepts and themes juxtaposed across the different studies and substances explored (See supplementary tables 5.4a and 5.4b). Classifying the studies based on the concepts and themes and grouping them by substance helped us understand how the studies were related and in which aspects they were different. Then concept maps were used to establish and discuss the influences of each concept over the other. After this phase, it was understood that most studies related reciprocally, except those that explore different aspects of the topic.

Phase 5 – Translating studies into one another

Phase 5 involved translating the studies into one another. In this phase, the themes and concepts were further refined to ensure that the themes reflected the meaning of each individual study. The initial codes and themes were examined and combined thematically when describing similar findings.

The influence of each study over each concept obtained is documented using references and quotes. Quotes were obtained from primary study participants and by primary author's explanations and interpretations. Since the context of the studies were very similar, the studies were organised in sub-groups depending on the substance they were exploring (either alcohol, smoking or illicit drug use, which included Cannabis). In our study, similar contexts are beneficial as they allow us to identify specific facilitators and barriers. This would then better inform the development of a context-specific intervention.

In this article we are presenting first, second and third level interpretations, based on the women's experiences.

Phase 6 - Synthesizing translations

The result of the translated concepts, their relationships and the primary data were used to create a textual line of argument, which is presented here. Three authors were involved in the synthesis (TES, KMS, SM) and the additional authors provided feedback and insights when necessary (KOD, MB, LL). The authors are from different

disciplinary backgrounds including psychology, sociology, medicine, public health, epidemiology and behavioural science, which promoted discussion of potential different interpretations.

Phase 7 - Expressing the synthesis

5.4 Results

5.4.1 Search outcome

Figure 1 shows the process of inclusion of studies. The first database search was conducted in March 2019 and identified 25,508 studies. Of these, 18,159 remained after duplicate removal. After screening for titles and abstracts, 85 studies remained potentially eligible for full text review. Following full text screening of the remaining 85 studies, 18 studies met criteria for inclusion in the meta-synthesis. Twelve more studies were examined for potential inclusion after hand-searching the reference list of the included studies, and 2 additional studies were included. The search conducted in November 2020 identified 152 potential studies. Four studies remained potentially eligible after screening for title and abstract, of those, two studies were included in our synthesis. The final number of studies included for synthesis was 22. Several facilitators and barriers were identified during our analysis that have an influence on women's' prenatal health behaviours. We classified these factors as internal factors or external factors, with internal factors being those that the woman might have some degree of control over and relate to her own beliefs, knowledge and intrinsic motivation to decide whether to use or not substances during their pregnancy. External factors are those which will have an influence on the woman's behaviour but are elements over which women have very limited control and relate mostly to their social environment or the healthcare system.

5.4.2 Study characteristics

The characteristics of the studies included are shown in Supplementary Table 5.3. Of the 22 studies included for analysis, 14 focused on smoking, 1 on alcohol, 3 on illicit drugs and 4 on different multiple substances. Six were conducted in the UK, 3 in the

USA, 3 in Australia, 1 in Canada, 1 in Sweden, 1 in Finland and 1 in New Zealand. The years of publication ranged from 1998 to 2020.

Twenty-one of the studies were qualitative and one mixed-methods. The studies used different data collection methods; nineteen used semi-structured interviews, three used focus groups. The number of participants in the studies ranged from 6 to 53, with ages ranging from 15 to 49 years. Thirteen of the studies included pregnant women only, one included only postpartum women up to 12 months after birth and eight included both pregnant and postpartum women. Regarding the quality appraisals, most of studies performed poorly with regards to reflexibility and ethical considerations and performed particularly well in the reporting of their aims, qualitative methodology, recruitment strategy and data collection methods justification (See Supplementary Table 5.5). Results from the GRADE-CERQual analysis are presented in Supplementary table 5.6.

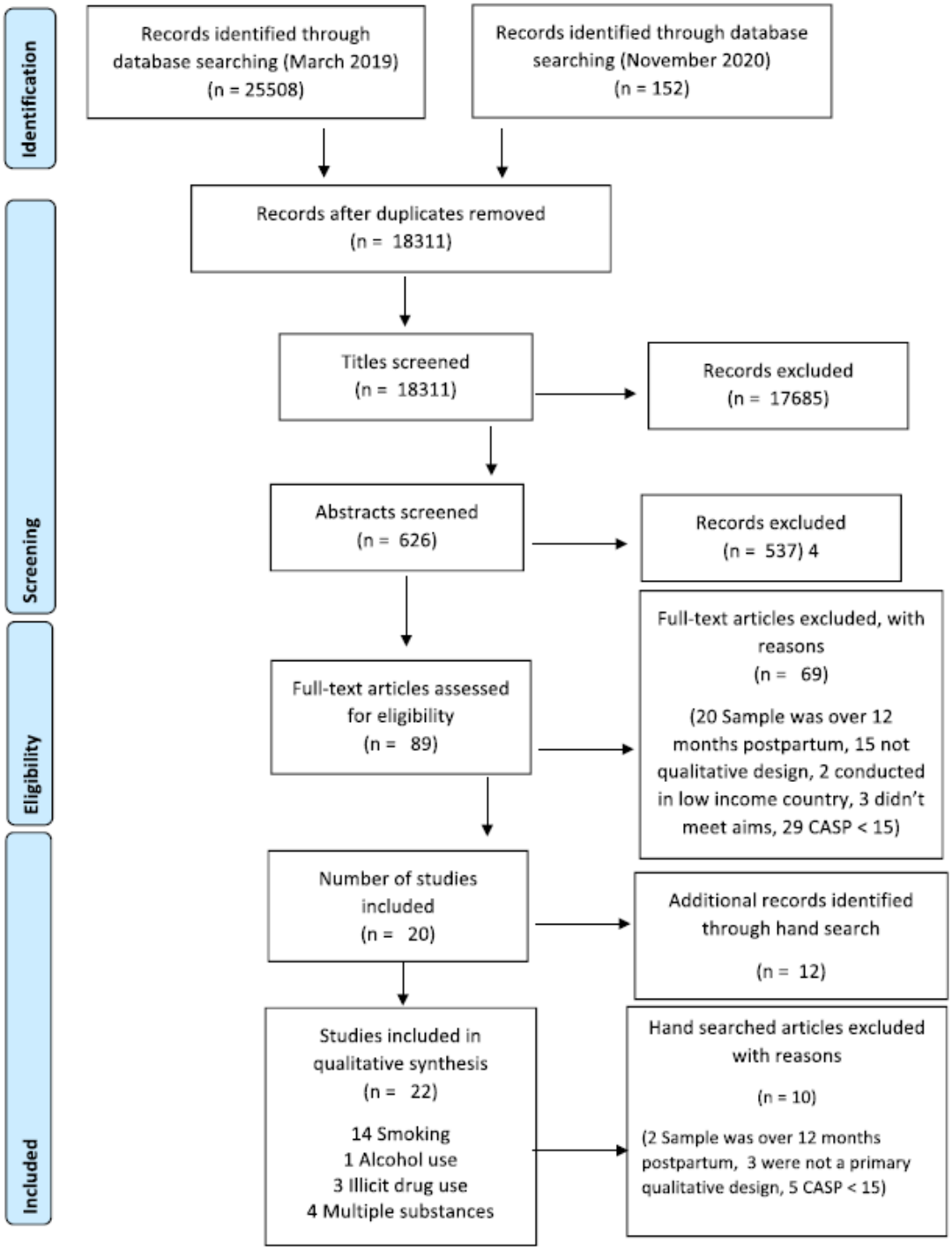


Figure 5. 1: PRISMA Flow Diagram showing the process of inclusion of studies. From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

5.5 Synthesis

Internal factors

Theme 1: Perceived incentives

In this theme we identified internal factors reported by women that might increase or decrease their levels of motivation to use substances or remain abstinent. Women reported perceived psychological and social benefits from their substance use, and additionally, women expressed reasons why interrupting/ceasing their use would have negative consequences upon them. However, some other women were also able to identify benefits of abstaining from substance use.

Category 1.1: Perceived benefits obtained from substance use

Women reported several reasons for continuing to use substances during pregnancy. Lack of motivation to quit[444,446,448,460,461] and boredom[444,446,448,461] were two reasons commonly reported when giving reasons for their substance use.

> *"So you're bored, what are you going to do? Sit down and smoke a cigarette."*[446]
>
> *"The part that wants to quit is smaller than the part that doesn't."*[446] *[Smoking]*

Furthermore, women reported obtaining benefits from the use of the substances that act as barrier to remain substance free. The most commonly reported benefit of substance abuse was that "*It makes me happy; it relaxes me"*[446]*;* hence, many reported that due to this *"total relaxation feeling"*[462] they used their habit as their main coping strategy for stressful situations[3,444,446,447,460–465]. For many women abstaining from the drug meant losing their only coping strategy for stress[3,444,446,460–463]. Taking the time to have a drink or smoke a cigarette gave them a sense of freedom and private time[3,447] whereby it was *"the only time I get a 5-minute break"*[444]*.* In other cases, women also spoke about how using their substance of choice has a positive effect on their mood[460,462], and the substances were perceived as source of enjoyment and socialisation[453,463,466]. In these cases, despite the fact that women might acknowledge the risks of the substance abuse, the benefits obtained

outweighed the risks.

> *"Smoking and drinking were an important part of her social life, and although she always imagined she would quit when she became pregnant, she found she was unable to do so"*[463]

> *"I felt like it was consistently helping me calm down and be able to function enough – well enough to parent my sixyear-old as a single parent and, um, deal with that"*[465] *[Cannabis]*

In the case of Cannabis in particular, women spoke about its therapeutic effects[465]. In one of the studies conducted in an American state where Cannabis use was legalised, women from different socioeconomic backgrounds used Cannabis to control their nausea, increase their appetite and manage stress as a *"more natural alternative than prescribed medications"*[465].

> *"It helps me feel hungry and it takes away my nausea completely. And it helps with the pain, too. And not so much that it takes away all of my pain, but it helps me mentally manage by pain better."*[465]

In addition, some perceived disadvantages of remaining abstinent were identified, including fear of harming the baby due to the consequences of the abstinence[444–447,466,467]; for instance, having increased levels of psychological stress[445,446,461,463,468] which women perceived as more harmful for the baby than the substance use itself[445,461]. The withdrawal symptoms and cravings as a consequence of the addiction to the substance[446,451,467,469] that in some instances lead them to feelings of loss of control[460] were also a source of concern. Additionally, women were concerned about the effect of abstinence on their relationships[446,451,467,469], since using is a common habit within their partner and/or wider social circle, whereby reported that they "*feel left out of some situations with my friends"*[466]

> *''There was a couple of times I just tried stopping, just getting sick. I would get a day into the sickness and it would just get to the point where it's just… I'm running out the door [to get more drugs.]''* [467]

Regarding smoking, women were concerned about gaining too much weight if they attempted quitting[466,468]. Since smoking was their main coping mechanisms for stress[446,463,466,468], women reported that they would probably resort to food as means of getting comfort when not being able to use cigarettes[463,468]. When speaking about illicit drug use, women were especially concerned about their mental health[451] and their perception that their life would feel dull without the effects of the drugs[451].

> *"I either eat or smoke.... I make the choice, I don't want to get big, so I'll go smoke"*[466].

Category 1.2: Perceived benefits obtained from substance abstinence

Within this sub-category, women spoke about practical benefits for their life, such as hygiene and money saving aspects[444,460,461]. But the main benefits identified by the women were related to their own and their baby's health[3,447,453,461,463,464,466]. Additionally, some emotional benefits for the women were also identified. Women felt a sense of relief from the guilt of using substances[3,453,463], and felt that they were giving a good example to their children by remaining substance free[466]. Furthermore, some women spoke about developing an increased perception of willpower and self-efficacy as consequence of their achievements during their process towards abstinence[444,453,463,470].

For some women, being pregnant or having other children was already motivation enough to attempt quitting, engage in harm reducing actions or to abstain from drugs completely[3,447,453,464]. Some women reported that *"No one is more special than my baby"*[463] adopting a moral responsibility towards prioritising their baby's wellbeing which facilitated that they remained substance free[447,448,453,462,463].

> *"There's a baby inside you. You know you've got to do it."*[3] *[Smoking]*

> *"My kid at 5 telling me that he doesn't want me to die, it was heartbreaking"*[444] *[Smoking]*

> *"The only reason why I quit was because I was pregnant, so I mean, if I hadn't fallen pregnant, I'd probably still be smoking cigarettes now"*

Another factor associated with the pregnancy that acted as a facilitator was the pregnancy-related sickness, women found that smoking or drinking alcohol during their pregnancy made them feel sick and resulting in women decreasing their substance use[444,463,464,469].

> *"Look, I don't want to be like a reformed bitch of smoking, but the smoke is just making me sick."*[463]

Theme 2: Feelings of Shame and Guilt

In most of the studies, women referred to feelings of guilt and shame due to their substance abuse[444,447,460,463,466,468,471]. These negative feelings in combination with the external pressure from their social environment acted as barriers to abstaining from substance use for some women whereby *"you feel even worse and in the end it becomes too much"*[460] resulting in women's stress levels increasing which lead to increased use of the substance as a coping strategy[460,468].

> *I just can't stand it. Sometimes I'll even try to turn the guilt off, and it's like it's still in my head and, you know, what I'm doing and I don't know why, it's just so hard for me to quit. Oh, and the baby will kick when I'm smoking, it almost makes me like cry because it's just like, I shouldn't.*[463] *[Smoking]*

External factors

Theme 3: Knowledge and information

Category 3.1: Knowledge about the risks of substance use

Many women discussed a lack of accurate knowledge about the risks women were taking when using substances during pregnancy. One factor that contributes to the generalised lack of accurate knowledge is the use of anecdotal evidence as a source of information and justification for substance use. Regardless of the substance, women often discussed their previous pregnancies or people they knew or heard of that had used substances during their pregnancy who had a healthy baby[3,444,468,446,447,451,453,460,462,463,467]. Generalising these individual cases might be

contributing to the creation of misconceptions about the risks. Furthermore, using these counter examples as justification might also be used as a coping mechanism to reduce the guilt associated with their substance abuse.

> *"I drank a little bit with my first child and I carried on doing that with my second and third pregnancies. My first child is absolutely fine."* [462] *[Alcohol]*

> *''So far from what I've seen, any girl that I know that's done dope throughout their pregnancy, their kids are really overachievers. Which, I'm not trying to say, 'use meth, it'll make your kids smart.' I'm just saying that the ones that I do know, there's nothing wrong with their kids.''*[467] *[Illicit drugs]*

Women reported misconceptions about the risks of substance use, which were likely a consequence of using unreliable sources to obtain information in combination with the anecdotal evidence explained above. For instance, only heavy use was perceived as dangerous by many women[3,447,451,462–465,467–469] and hence *"If you drink in moderation and you're sensible, then I don't think it affects the fetus"*[462]. The distinction between heavy and light use was made by the women in a subjective way, and in many cases, it was not based on medical evidence. For example, light alcohol use was defined by women as *"one sip of wine"*[447], *"one or two [drinks]"*[447], *"drink in moderation"*[462]; whereas women defined light smoking as "*four a day*"[468], "*an odd fag [cigarette] now and then"*[3], "*under ten cigarettes a day*"[463] or women considered illicit drug use safe when used *"in low and steady doses"*[451,467]. Further, certain substances were seen as more dangerous than others[446,447,451,463,465,468] *''alcohol was really bad, but there was no proof that meth did anything to the babies''*[467], and some women thought that the consequences of quitting could be worse than the consequences of using[446,451,463,467]. Some women also believed that the properties of the substance might benefit the baby[447,462,467], with a women reporting that *''babies that have been drug exposed may be more gifted, more creative, and more beautiful.''*[467].Other women thought that the risks are exaggerated[3,444,446,451,460,462,463,467,468,471], believing that the consequences of substance use on the fetus are "*things that are fixable"*[444]once the baby is born[444,446,467] . These misconceptions can act as barriers as women rationalise their

substance use and decrease the feelings of guilt that women report feeling.

> *"I don't want that to sound nasty, but like having a small baby, the baby will grow, he can put on weight . . . and doctors are amazing now like . . . there's more chance of the baby developing a cleft lip as well, that could be fixed with surgery." [Smoking]* [444]

Another barrier to remaining abstinent is that some women believe "*nothing will happen to you"*[468] *and* "*nothing happens to the baby either"* [468] and so they do not feel susceptible to the potential harms of the substances[3,444,446,460,462,463,467,468]. In these cases, despite having some knowledge or awareness of the risks, women share the belief that this cannot happen to them, that *"it only happens to other people".*

On the other hand, having more accurate knowledge about the risks of substance use acted as a facilitator to prevent substance use during pregnancy[3,444,464,466,467,446–448,451,453,460,462,463]. In the case of alcohol, for instance, since the information about its risks can be contradictive and confusing, women tended to *"go for the safety aspect, so because I'm not 100% sure, I just completely abstain to be on the safe side. [Alcohol]"*[462].

In the case of smoking, besides being aware of the risks of the smoking itself[446,448,463,466,468], having knowledge about the risks of passive smoking[444,468] and the influence of smoking on breastmilk[466] also acted as facilitators.

> *"It gets not only into your lungs, but it gets into your bloodstream and everything, so why wouldn't it get into your milk and go to the baby?"*[466] *[Smoking]*

Despite the fact that many women resort to counter examples and misinformation about the risks to rationalise their use and reduce their feelings of guilt, most of them are aware that there are certain risks associated to their behaviour[444,446,465–467,469,447,448,451,453,460,462–464]. This awareness led some women in these studies to take action in order to minimise the harms produced by their use[463,464,466–468]. In some instances, these changes were in line with common recommendations, whereas with others they were in response to their own perception of what was best for the baby.

The most commonly reported strategy to reduce harm was trying to abstain or cutting down the substance use[444,447,461,463,466–468]. Cutting down to levels considered *"not that much."*[468] was perceived as a positive solution to balance the baby's needs with their own needs, and it was advised by healthcare professionals when unable to abstain completely[463,468]. Women also tried to compensate with other health behaviours[447,461,463,466–468] such as *''eating more frequently"*[467] *and "sleep every night''*[467], and keeping themselves busy and distracted whereby *"instead of smoking, doing the kids' sandwiches"*[464] or doing exercise to release stress[464]. Other women limited relationships with other users[467]. Women also reported changing their habit to what they considered safer[444,463,467], for example switching substance[467] or type of administration[467]. Although not always adaptive, these behaviours show that women have some motivation to change their habits and therefore considered them facilitators.

> *''We posted a big sign on the front door saying, 'No Drugs.''*[467]*,*

> *''I was just like, well, ok, in the morning I can just drink coffee instead [of using methamphetamine].'' When this didn't work, she tried switching to drinking alcohol, ''I will just replace that [methamphetamine] with X amount, with alcohol"*[467]

Category 3.2: Sources and quality of information

On several occasions, women reported a lack of sources of information regarding the risks of the substance they were using[444,447,451,462,465–467,469]. There was a perceived lack of evidence about the risks reported by women[451,465,467] *"I've actually heard they don't exactly know what is safe and what isn't. Because they don't know they say not to do it [Alcohol]"*[447]*,* and in many cases, women spoke about not having specific information[444,466] about *"what damage does [Smoking] actually do to the placenta, you know, proper sort of medical facts"*[449].

> *"If I was given more information, you know, per scientific research studies, I could maybe make a more informed decision"*[465] *[Cannabis]*

Another reported issue that acts as barrier is the poor communication with healthcare professionals and women about substance use. Women reported only being asked about their substance use at their booking visit *"there was no other conversation about it"*[447] or if there was discussion it was on very limited occasions[444,447,448,465,466]. The discussions about their substance use were very superficial and generalised, and lacked practical advice[444,446,463], leaving women with poor understanding of the risks[444,466,468].

> *"My doctor just tells me that it's really important for me to quit. Well, I know that already, and I want to quit too. If it were so easy, I would have done it already. So when he says that to me, I just say, "Okay," and that's the end of the conversation"* [463] *[Smoking]*

Further, on several occasions women spoke about receiving conflicting information from different sources[445,447–449,462,465,469], including amongst different healthcare professionals[444–446,448,449,461,463,465,466], and also, there were occasions in which family or friends of the woman would act as source of information.[451,471].

> *"So like my GP I didn't really get a definitive answer…Whereas when I went to my midwife, for my antenatal appointment that was clearly communicated from day one…"*[447].

> *"My doctor, he doesn't like that I use marijuana at all. [...] And once I was pregnant, he's like, 'You've got to stop.' And I did stop when we talked about it. But then when I told him my OB's decision to let me linger a bit, at least until I'm like 20 weeks or so to see ifthe morning sickness goes away [...] He doesn't like it, but my OB doctor totally understands."*[465]

Theme 4: Experiences within healthcare system

Category 4.1: Relationship with healthcare professionals

Women expressed feeling judged and stigmatised for their substance use during their antenatal care[3,445,446,448,451,471]. Women spoke about not being treated like 'normal'-expectant mothers due to their substance use[471].

"You just knew that you were not going to be treated equally and like other pregnant women and accepted for who you were. I mean after all I was at the clinic because of drug problems, and that was never forgotten [by the staff]".[471] *[Illicit drugs]*

Women also reported a lack of encouragement or insensitive attitudes from healthcare professionals during antenatal care[446,461,468,471], which together with a perceived prioritising of the baby's needs over their own needs[471], became a barrier to motivate women.

"…Never a hint of praise for coping the best you can. No-one ever asked me a really personal question that might have a happy answer"[471] *[Illicit drug use]*

When discussing their substance use, some women explained that they found that healthcare professionals adopted an authoritarian or coercive communication style[444,448,460,461,471]. As with the general negative consequences of the social pressure, women perceived this communication style like an additional stressor or a challenging attitude.

"They [midwives] just keep telling me to stop . . . it winds me up a little and makes me worse [with smoking]."[464]

Women also reported some issues with regards to the staff working in the cessation treatment centres for illicit drug users similar to those encountered with the antenatal care staff. Women felt judged and stigmatised, and perceived that staff were biased towards the patients[471] perceiving some of them as too challenging or hopeless[471].

Hence, according to these women's experiences, it seems that healthcare professionals' attitudes and behaviours might be influencing the women's process towards abstinence by potentially preventing care-seeking behaviours or adherence to treatment, increasing stress and consequently use of the substance as coping strategy and missing opportunities to educate and inform women accurately about the risks of their substance use.

In other cases, women also spoke about finding supportive and encouraging

healthcare professionals that facilitated their quitting process[444,451,463,464] who were *"like a nagging mom, but in a good way"*[463]. Further, healthcare professionals with an empathetic and non-judgemental attitude were valued by women as well[446,448,449,460,463,464,466,468,471].

> *"When I walk in her room we have a good laugh and a good giggle and we have a good chat, she isn't one of these that just gets straight down to business and lets you blow in the meter and sends you on your way and just asks you questions and questions and questions, she makes sure that you're alright in yourself." [Smoking]*[464]

Category 4.2: Practical limitations of drug cessation treatment centres

Women using illicit drugs also reported several limitations within the **cessation treatment centres** that acted as barriers for them to engage in care. Women reported having to choose only one child to take with them into the residential treatment center[450], being rejected for being pregnant[450], not being able to meet pre-admission requirements or finding the referral system too complex[444,445,450] which led them to perceive these centres as unhelpful and an additional source of stress[444,450].

> *"There was an opening for here [the treatment program]. But the only hardest part was I had to choose one kid…(crying) So…they (child welfare) came and got my ten-month-old, and my two-year old came here…It's not fair…(crying) Because he's only a baby, he didn't deserve all this. I had to do it by myself…just what I thought would be best."*[450] *[Illicit drugs]*

Theme 5: Influence of social environment

Category 5.1: Social disapproval

The social disapproval associated with substance use during pregnancy was experienced as having disadvantages[444,445,447,448,461,464,465,469]. Women spoke about feeling questioned, judged and labelled as *"just a coke head with a 12-year-drug habit"*[471] by other people[444,447,448,451,471] which led them to think that they were being perceived as bad mothers. The external social pressure to which women were

exposed from their family members, partners, healthcare professionals, etc. acted as barrier since it increased their levels of stress[444,446,448,460,461] and sometimes was perceived as a challenge to their individuality[469]. Hence, due to the negative feelings produced by external pressure in addition to their own feeling of guilt led women to hide when using substances or avoid disclosure[3,445,447,450,463,468,469,471,472].

> *"in the end if my partner said anything I kind of just did the opposite just to you know prove a point…this is my baby and my pregnancy"*[469] [Alcohol]

> *"I did smoke with them but it was in my own house, I never walked around out and about with one, it's not the best look"*[469]

However, some women find this perceived social disapproval as a motivator for change[447,466]. Hence, social disapproval also has some benefits. In these cases, women wanted to be perceived as good mothers which acted as a facilitator to reduce or eliminate substance use. In other cases, women perceived their substance use as "*a family decision; it's not just about me"*[447].

> *"Several women felt that the decision not to drink was a family decision as it would have effects on the baby, and that their partner had input on many of the health decisions made during pregnancy."*[447]

However, after doing our CerQual analysis, we concluded that this finding – perceiving social disapproval as a motivation to change- has serious limitations and hence the level of confidence assigned to it is low. This is because only two studies with moderate methodological limitations support this finding.

Category 5.2: Influence of type of support from close environment

A lack of support in close environments, which include people that the woman shares a house with, their family, partner, friends and work environment, has been reported in several occasions as an external barrier to remain substance free. Women reported not having their partners support to remain abstinent as a barrier[3,444,445,447,451,461]. The fact that their partner uses the same substance and *"was smoking around me all the time"*[461] was reported as a barrier by

women[444,445,447,451,463,464]. Further, women also spoke about perceiving inconsistent attitudes towards substance use from their partners that led to frustration[463]. Specifically in the case of smoking, women spoke about their partners not maintaining their habit changes over time, feeling pressured and guilted by their partners to quit smoking while their partners themselves were still smoking[444,461,463], being encouraged by their partner to smoke lightly in times of stress[461,463]

> *"It made it hard (staying smoke-free) because my husband had not tried to give up and…it was constantly in my face and he would say, 'Come on outside and have a smoke with me you won't be so stressed'. I gave in. It was my own fault for giving in."* [461]

For some women, substance use is a normalised event in their family and they have always been exposed to it, especially in the case of smoking[448,463,464,466,469]. Family members showing resistance to change their own smoking habits also act as barrier[468]. Some women need to share their homes with extended family and are exposed to second-hand smoking which makes them feel frustrated[463,464,466,468,470], but they feel powerless to enforce any changes.

> *"My family smokes, my friends smoke ...everyone's smoking....It's gonna be a challenge"*[463]

> *"Well quite a few of my family smoke, so, I mean I'd say that tempts me to smoke when they're around and stuff"*[464]

On the contrary, a supportive close environment can also facilitate women's attempts to remain substance free. Women that reported having supportive partners that are *"probably drinking less…because he's not drinking"*[447] or changed their habits in some ways to avoid exposing the women to the substance were very helpful in their own quitting process[444,446,447,461,464]. Further, environments that discourage substance use and limit people's access to it also act as facilitators[444,463,464].

> *"I asked him to buy cigarettes for me once because I was really losing my mind and he would not do it"*[447].

"Because I'm doing an 8-hour shift on my own, so there's like 9 hours where I'm not smoking"[444]

Category 5.3: Socioeconomic factors

Regarding the wider context, there were some socioeconomic factors that act as barrier to remain substance free[448,450,451,463,470,471]. Some women found that seeking care to reduce their substance use was not their priority, since they have other more immediate needs to attend to[444,445,461]. For instance, women reported having transportation issues[444,445], housing and food insecurity[445], financial issues[444] and domestic violence[450] as barriers. These women experienced chronic high levels of stress and resort to their substance use as source of comfort[463].

One of the commonly reported barriers associated specifically with the use of illegal substances was the concern that women *"could have been arrested"*[450] due to the potential legal consequences of their use . Women actively avoided attending antenatal care or disclosing their substance use due to fear of losing custody of their babies[450,451,453,465,467] or being legally prosecuted[450]. Even in the case where Cannabis use was legalised, women were still confused and concerned about potential legal consequences due to their use during pregnancy[465].

"If you have another [drug-exposed] child within a three-year period, even if you're staying clean and sober, your child will be taken from you …I was really scared of that…that's what kept me from going to prenatal care."[450]

"And I was like, 'Is that like legal? Can you smoke while you're pregnant?' And she said that I can't get in trouble for it, but I still worry a little about it. She said, 'Just be aware that if you use any for labor or anything like that, do not let the nurses know at the hospital because they will report you to CPS.' She said, 'I will not. I would not do that.[...] She said, 'Just be aware the nurses at the hospital have a different view.'"[465] *[Cannabis]*

5.6 Discussion

This meta-synthesis identified factors that can act as facilitators and barriers influencing whether women would try to abstain from substance use (either smoking, alcohol or illicit drug use) during their pregnancy. Importantly, despite the fact that the facilitators and barriers identified in this study have been divided into different categories and themes, these factors can co-occur and must be considered together to be able to understand the complexity of substance use during pregnancy. Women reported perceived psychological and social incentives associated with substance abuse, and for many women, their substance use was their only coping mechanism for stress. However, benefits of remaining abstinent such as improved hygiene and health and money saving were also reported as facilitators. Another barrier identified was related to the shame and guilt that some women feel due to their substance abuse which created negative feelings and increased levels of stress. Poor information, communication and knowledge about the risks together with lack of discussion with the healthcare professionals and the conflicting information available acted as a barrier which made some women resort to anecdotal evidence or unfounded beliefs to justify their substance use. On the contrary, women with some awareness of the risks tended to adopt cautious attitudes or engage in harm-reducing strategies. Women also spoke about the influence of the type of support they received from healthcare professionals. Being exposed to social disapproval made them feel questioned and labelled. However, for other women this social pressure acted as motivation for change. Non-supportive, stigmatising and judgemental healthcare professionals acted as a barrier to remain abstinent, whereas encouraging and empathetic healthcare professionals were highly valued. Further, the social environment of a woman can have a negative influence, including lack of support, and additional socioeconomic factors such as housing and food insecurities, unstable jobs and financial issues were identified as external barriers.

The results of the study show that some women were incentivised by the effects of substance use to continue using. Women utilised substance use as a coping strategy with psychological and social benefits such as stress reduction, relaxation feelings and mood improvement. These results are in concordance with previous studies that have established stress as a risk factors for substance use[473–475]. Consequently, the

balance of perceived benefits versus potential health, psychological and social risks that substance use has for these women will determine their attitudes towards it[476]. According to the Theory of Planned Behaviour, behavioural intentions are a result of attitudes and subjective norms[477]. This model also adds the concept of perceived behavioural control, which relates to individuals' perceived difficulty to perform a behaviour[477]. Behavioural intentions and perceived behavioural control are considered main determinants of behaviour change, which, according to the Theory of Planned Behaviour, means that this change can only happen when there is a change in the beliefs that influence the intentions and the perceived behaviour control[476,477]. Our results show that in some cases the benefits that these women obtained from their substance use were perceived to outweigh the perceived risks, which might lead them to have a positive attitude towards engaging in substance use. However, the results also demonstrate that some women lack accurate knowledge about the risks, which might mean that this process of deliberating benefits and disadvantages might be biased towards the benefits. Previous research has demonstrated that deficits in health literacy can be translated into a poorer health status as it acts as a barrier to healthcare and can also impact people's decision-making[478]. Additionally, some women spoke about feeling a loss of control when having cravings and perceiving themselves as incapable of remaining substance free. These feelings of low self-efficacy and lack of control over the substance use might influence the women's perceived behavioural control in a negative way. This is in concordance with previous studies looking at perceived self-efficacy and behaviour change in different populations, where the authors concluded that the lower the perceived self-efficacy the minimal will be the behaviour change[479–481].

Stress also seemed to be related to substance use in our results. Exposure to stress involves changes affecting nearly every system of the body, influencing how people feel and behave[482]. Lazarus and Folman's Transactional Theory of Stress and Coping explains that, when dealing with a given situation, individuals first assess whether a specific/individual transaction is positive, neutral or stressful[483,484] depending on the consequences it can have on their well-being. When a transaction is considered stressful, the individual will have to evaluate their coping resources (self-efficacy),

the situation variables (level of control), and their coping styles (coping mechanisms used or learnt in previous similar situations)[483], and this copping styles can be directed at tacking the stressor (problem focused) or to regulate the emotion caused by the stressor (emotion-focused copign) [483]. In the context of pregnancy, coping efforts might influence birth outcomes since they can serve as a resource to prevent prenatal stress exposure to the infant[485]. Positive reframing, acceptance and seeking emotional support are coping strategies that have been seen to have beneficial effects on antenatal anxiety, worries and depression[486]. However, results from our study show that the substance use acted as their main coping mechanism for many women. Hence, in the case of these women, not only is the exposure to stress detrimental to the infant, but also the coping mechanism. Since this strategy does not directly manage the stressor, we could classify substance use as a emotion-focused coping strategy within the transactional model[487]. Although emotion-focused coping strategies can be adaptative when the stressful situations provoke intense emotional reactions or distress, the stressor is perceived as uncontrollable or when the person does not have enough resources to use a problem-focused strategy, they have been proven to be ineffective long term[488].

Hamilton and Lovel conducted a study with the aim of determining how women cope with stress across pregnancy[489]. The authors used the Revised Prenatal Coping Inventory (NuPCI factors) to explore the different coping strategies potentially used by women and concluded that higher state anxiety and pregnancy specific distress were the strongest predictors of use of avoidance as coping strategy throughout the pregnancy[489]. These results are in concordance with the relationship found between high levels of stress and use of substance in our study. Avoiding coping style and behaviours during pregnancy such as substance use have also been associated with a wide range of adverse mental health outcomes such as increased distress, higher depressive mood, increased anxiety, higher perceived stress and greater child abuse potential[485,489–491]. Hence, the circle between high levels of stress and substance use might be a partial explanation as to why women maintain their substance use during pregnancy. Further, some women in our study were aware that stress can be detrimental for their unborn child, and considered that the increased stress that

abstinence brought to them would be more detrimental than the substance use itself and hence the substance abuse would have a protective effect for their babies.

Women spoke about feelings of guilt and shame due to their substance use. These feelings could be a consequence of the cognitive dissonance created by their substance use and their awareness of the risks of their substance use. According to the theory of cognitive dissonance, this occurs when a person holds two inconsistent thoughts, in the case of the women in our study, the substance use and the awareness of the risks of these substances[492]. The existence of cognitive dissonance can be psychologically and physically uncomfortable [492], which might explain the feelings of guilt referred by the women. When cognitive dissonance is present, the person might try to reduce it using different strategies: by avoiding situations or information (which in our study translate as using unreliable sources of information such as anecdotal evidence), adding additional thoughts to rationalise the inconsistency (which we observed as accepting unfounded facts as true), or trivialising the inconsistency (like using unrealistic optimism expressed by some women in our analysis)[492]. Behaviour change is needed to reduce cognitive dissonance. Although information and education is not enough to change behaviour, it can be a first step[363]. This generalised lack of knowledge about the risks of substance use during pregnancy might partially be a consequence of the reported lack of accessibility to sources of information, the limited discussion with healthcare professional as well as the conflicting messages from different sources. It is crucial to make sure women are making informed decisions based on current evidence instead of resorting to non-adaptative strategies to reduce their cognitive dissonance.

An additional issue identified within this study was related to the level of stigmatisation that women were subjected to, especially during their pregnancies. Previous research in the field of substance use has shown that stigmatising attitudes towards drug users are common, not only amongst the general public but also amongst non-specialised healthcare professionals[493]. This leads people with substance abuse problems to have ambivalent attitudes towards seeking treatment due to the negative connotations associated with it which with they would not identify themselves[494]. Additionally, the fear of reprimands or legal consequences

also acted as a barrier for care seeking and disclosure of their substance use as shown in our results, even in the cases where the substance use was legalised. Relatedly, our results show that it is crucial for women using substances to find a non-judgemental and non-authoritarian space within their antenatal care in order to feel safe to disclose their use and seek additional care when necessary.

Our results also show that substance use is a common event for many women in their families or close social circle, which not only acts as a social facilitator for substance use but also might have led some women to normalise the use of substances as a coping strategy for stress through a process of observational learning and imitation[495]. Further, the perceived lack of alternatives and lack of control over their environment, might contribute to this phenomenon. These results can be explained by the Social Learning Theory that postulates that adults who present themselves as role models while being involved in addictions, will have a strong effect on reinforcing other people's addictive behaviours, especially if there are social reinforcements associated to this behaviour[496,497]. Women in our study struggled more to remain abstinent when their partners or close environment continued to use the substance in their presence. Stimuli and environmental factors can act as cues for substance use through a process of classical conditioning[496]. Hence, it would be beneficial for the women to involve their partner's or closest social influences into the abstinence process in order to encourage and promote healthier pregnancy habits.

The social environment to which a woman is exposed will have an influence over their substance use habit. The Social Determinants of Health framework developed by Dahlgren and Whitehead[498] could be used to explain this influence. According to this framework, social aspects such as the conditions under which a person grows up, the availability of social support, community network, access to education, housing, employment, health services and water and food supplies will have an impact on the health outcomes of an individual[405,417,418,499,500]. Our results show that certain factors such as housing and food insecurity, unstable jobs, financial issues and domestic violence as barriers to seek care for their substance use. Those factors will have an influence on the women's ability to remain substance free, and hence,

interventions targeting the different levels need to be developed to make sure that the issue is addressed as a whole[405,419].

The National Institute for Health and Care Excellence (NICE) guideline for substance use during pregnancy recommends healthcare professionals refer women who misuse substances to appropriate substance misuse programmes as soon as they disclose the pregnancy[501]. Additional relevant support services should also be recommended and encouraged based on the woman's individual needs[501], this is very relevant as women in the studies explored reported a need for more individualised care. Further, the NICE guidelines also recommend offering women with information about the potential effects of substance use on her baby and what to expect when the baby is born, which matches the need reported in our study for further information and discussion with healthcare professionals. These guidelines also acknowledge the importance of the healthcare professional's attitudes and how these can prevent women from engaging with the services provided[501] which we also identified as a barrier for care seeking and disclosure.

According to Flenady and colleagues, a large proportion of stillbirths occurring in high-income countries are avoidable[87]. Increased clinical and community awareness of the modifiable risk factors associated with stillbirth might improve prevention efforts and reduce stillbirth rates, and we believe that the findings of this study provide additional detail and context that could be valuable for this purpose.

There are some limitations to our study. Firstly, we used a limited number of databases to conduct our systematic review. These databases were selected based on similar studies in the area, previous experience and knowledge about the databases scope.

Secondly, some studies were excluded based on quality assessment. This decision was made to enable the findings of our studies to be of high quality, however, excluding studies based on quality appraisals might result in a loss of relevant input. Thirdly, the findings obtained from this study are based on women's experiences and interpretations and should not be understood as facts. Additionally, our findings are based on the secondary analysis of the primary data, collected and analysed by the

studies' primary authors. Although the findings across the studies were consistent and we conducted a CerQual assessment to evaluate the confidence of our findings, it is important to acknowledge this potential influence over our findings. Finally, the publication dates of studies included in our analysis range from 1997 to 2020. There is a possibility that findings may have been influenced by variations in antenatal care over time. However, similar themes were identified in studies regardless of time of publication.

In conclusion, interventions need to be designed at different levels to address the issue of women using substances during pregnancy. Factors such as misinformation and lack of knowledge about the risks of substance use should be targeted with the women, this could contribute to the creation of more accurate balance of the real risks and perceived benefits of substance use during pregnancy. Hence, women need to be provided with accurate and accessible sources of information, and discussion with healthcare professionals needs to be encouraged. Additionally, women under these circumstances would benefit from interventions targeted at developing new coping mechanisms for stressful situations.

However, there are external factors that also need to be targeted in order to minimise barriers for these women. Non-specialised healthcare professionals should be educated on the complexities of substance use to encourage non-judgemental and sensitive attitudes. The social circle of the woman must also be addressed, involving the women's partners, family and friends within the quitting process might be helpful. Moreover, especial considerations need to be taken into account with women in vulnerable socioeconomic situations.

The facilitators and barriers identified in this study can serve to inform the development of a behaviour change intervention to tackle this issue and help prevention efforts towards reducing the risk of stillbirth.

5.7 Inter-chapter synopses

Chapter 5 aimed to identify the facilitators and barriers to substance-free pregnancies. Some of the facilitatos identified in the study included having social support, access to reliable information, and a positive attitude towards achieving a substance-free pregnancy. On the other hand, the barriers to achieving a substance-free pregnancy included lack of stigma, shame, and lack of support from social environment and healthcare professionals, as well as lack of awareness of the risks. Additionally, the study found that healthcare professionals' attitudes and approaches towards pregnant women who use substances play a crucial role in facilitating or hindering substance-free pregnancies. Chapter 6 had the same objective than Chapters 4 and 5 but with a focus on weight management behaviours. This study was conducted to address the need of further understanding the influences over women's willingness and capability to engage in weight management behaviours, as there is evidence in the literature highlighting the risks associating high BMI and stillbirth[188].

Chapter 6 - Facilitators and barriers influencing weight management behaviours during pregnancy: a meta-synthesis of qualitative research

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Chapter 6 - Facilitators and barriers influencing weight management behaviours during pregnancy: a meta-synthesis of qualitative research

6.1 Abstract

Background

Obesity and overweight are considered risk factors for a range of adverse outcomes, including stillbirth. This study aims to identify factors reported by women influencing weight management behaviours during pregnancy.

Methods

A systematic search was conducted in five databases from inception to 2019 and updated in 2021. Qualitative studies involving pregnant or post-partum women, from high-income countries, examining women's experiences of weight management during pregnancy were included. Meta-ethnography was used to facilitate the meta-synthesis of 17 studies.

Results

Three themes were identified during the analysis: (1) Awareness and beliefs about weight gain and weight management, which included level of awareness and knowledge about dietary and exercise recommendations, risk perception and decision balance, perceived control over health and weight gain and personal insecurities. (2) Antenatal healthcare, women's experiences of their interactions with healthcare professionals during the antenatal period and the quality of the education received had an effect on women's behaviour. Further, our findings highlight the need for clear and direct information, and improved interactions with healthcare professionals, to better support women's weight management behaviours. (3) Social and environmental influence, the social judgement and stigmatization associated with overweight and obesity also acted as a negative influence in womens' engagement in weight management behaviours.

Conclusion

Interventions developed to promote and maintain weight management behaviours during pregnancy should consider all levels of influence over women's behaviours, including women's level of awareness and beliefs, experiences in antenatal care, education provision and social influence.

Keywords: pregnancy, weight management, behaviour change, facilitators, barriers.

6.2 Introduction

According to the World Health Organization (WHO), the prevalence of obesity worldwide has nearly tripled since 1975. Based on 2016 data, 39% of the adult population globally were overweight, and 13% had obesity[502]. A recent study, including 20 different European countries, concluded that 53.1% of their adult sample had overweight or obesity[503]. Increases in overweight and obesity and gestational weight gain trends are also observed amongst women of childbearing age (15 to 44 years old)[504,505]. Overweight and obesity during pregnancy are associated with a wide range of complications including increased risk of gestational hypertension[506], preeclampsia[506], gestational diabetes mellitus[506], caesarean delivery[506], preterm birth[506], delivery of large-for-gestational infants[507], and stillbirth[1,184]. Additionally, excess of gestational weight gain is associated with gestational diabetes, pregnancy-induced hypertension, caesarean delivery, postpartum weight retention, macrosomia and childhood obesity[508].

Weight management during pregnancy is therefore critically important and can involve individual behaviours related to diet and physical activity, as well as information and support from healthcare professionals. For the purpose of this study, we understand weight management as all of those behaviours that might influence women's weight loss, weight maintenance and women's gestational weight gain. Most information and advice that women receive about weight management during pregnancy is related to nutrition and levels of physical activity[509]. The NICE guidelines for example, recommend that pregnant women base meals on starchy and fibre-rich products, fruit and vegetables and eat a low-fat diet[510]. In terms of physical activity, the Royal College of Obstetricians and Gynaecologists (RCOG) and the Royal College of Physicians of Ireland (RCPI) recommend that women engage in a moderate amount of physical activity such as aerobic (e.g swimming or running) or strength conditioning, starting with 10-15 minute sessions for women who were not active before pregnancy, building up to 150 minutes per week[511,512]. Comparison of prenatal physical activity guidelines from multiple high-income countries indicates that the majority recommend moderate physical activity, with recommendations to seek healthcare provider advice before

starting or continuing an exercise program, and advice to not engage in sports that involve risk of falls, trauma or collision[513–515].

Weight management behaviours such as diet and physical activity are important because they are modifiable behaviours that women can engage in to improve outcomes for themselves and their infants. Despite the recognised benefits of a healthy diet and physical activity during pregnancy, previous studies have shown that a low proportion of pregnant women adhere to prenatal dietary guidelines and/or meet physical activity guidelines[516–518]. Caut et al (2020) concluded from their systematic review that most preconceptual and pregnant women did not meet recommendations for vegetable, cereal grain or folate intake, and in half of the studies, women exceeded fat intake recommendations[518]. Regarding adherence to physical activity recommendations, a study conducted in the USA found that only 9.3% of pregnant women met the American College of Obstetricians and Gynaecologist (ACOG) guidelines for physical activity; 41% of women in this study reported engaging in physical activity <1 day/week[516]. Such low levels of adherence to dietary and physical activity guidelines and recommendations highlights the need for greater understanding of women's barriers and facilitators to engaging in these weight management behaviours.

Previous studies have explored experiences of pregnant women in relation to weight during pregnancy. Evidence of barriers and facilitators to gestational weight gain include physical barriers knowledge and beliefs, logistics and social barriers[519]. We believe that our study has a wider scope by exploring weight management which is understood as all of those behaviours that might have an influence on the women's weight loss, weight maintenance or gestational weight. Identifying these facilitators and barriers can inform the development of interventions to better support women's weight management during pregnancy and ultimately improve maternal and child health outcomes. Hence, the aim of this meta-synthesis is to identify and analyse qualitative research published to date in high income countries in order to establish what facilitators and barriers influence pregnant women's weight management behaviours during pregnancy.

6.3 Methods

The protocol for this meta-synthesis is registered on Prospero (no. CRD42019120069). Originally the objective of the registered meta-synthesis was to examine three different maternal behavioural risk factors for stillbirth (weight management behaviours, substance use, attendance at antenatal care). Given the complexity of these health behaviours and the large amount of literature exploring them, a synthesis including all three risk factors was deemed not to be feasible and/or likely to provide coherent findings across all behaviours. Consequently, this meta-synthesis differs from the protocol in that it focuses only on weight management behaviours rather than weight management, substance use and attendance at antenatal care. The findings in relation to the other two modifiable risk factors are published elsewhere[410,520].

6.3.1 Search strategy

We conducted a comprehensive search of the literature to obtain all qualitative studies that explored women's facilitators and barriers to weight management during pregnancy. The databases searched were CINHAL, PsychINFO, Pubmed, SOCindex and Web of Science. Searches were conducted in March 2019 and updated in April 2021, with no restrictions on publication date.

Search terms used were *facilitators, barriers, promoter, benefit, attitude, opportunity, determinant, promotion, intention, education, initiative, prevention, pregnancy, weight management, physical activity, nutrition, overweight* (see example of search in Supplementary file 6.1).

6.3.2 Study selection

The review of all titles and abstracts obtained from the search was completed independently by two members of the research team (TES, LL); three authors (TES, COC, KMS) conducted the full text screening.

Studies were included for further review if (1) they used a qualitative or a mixed methods design, as long as they included primary qualitative data; (2) they were written in English; (3) the participants were pregnant women or women up to 12 months post-partum as long as the data related to their pregnancy period (e.g.

studies exploring post-partum weight management were excluded); (4) they were conducted in high-income countries; (5) they included extractable data about facilitators and barriers to manage weight during pregnancy; (6) they explored weight management behaviours that women were able to engage with personally (e.g. healthcare professionals' weight management practices such as routine weighing were excluded).

Studies are restricted to high-income countries given the differences in care systems between high, middle, and low-income countries, and the different challenges associated with weight management in the different contexts. Furthermore, limiting the findings of this synthesis to high-income countries only will facilitate the development of prevention strategies applicable to high-income countries.

Studies that did not include any qualitative data or were not original research were excluded. Studies that included different types of participants (e.g. healthcare professionals, partners, or family members) were only included if the data obtained from the women was clearly differentiated from the data obtained from the other types of participants. Studies describing behaviours linked to weight management (such as diet and physical activity) but which were not explicitly linked to prenatal weight management in the published manuscript, were excluded.

6.3.3 Data extraction

Study Characteristics

A data extraction sheet was used to extract the characteristics of the studies by two authors (TES, COC). The following data were extracted from each study: country of publication, year of publication, aims, design, data collection method, sampling or recruitment strategy, consent process, number of participants, age of participants, pregnancy status, BMI and/or activity level of participants if applicable, timing of data collection, method of data analysis and study results.

Quality Assessment

To assess study quality, the Critical Appraisal Skills Program for Qualitative studies checklist (CASP)[521] was used independently by three authors (TES, SM, COC). There are a number of different CASP checklist designed to asses different types of studies,

including qualitative studies. The CASP Qualitative Studies Checklist – used in this study- is composed by ten different items exploring clarity of aims, adequacy of methodology and design, appropriateness of the recruitment strategy and data collection process, issues related to reflexivity and ethical considerations and clarity of the analytical process and statement of findings, as well as the value placed in the research. Since CASP does not provide a rating system, it was decided to rate as 0 for "No", 1 for "Can't Tell" and 2 for "Yes", in order to facilitate grading of the studies.
A previous study conducted in the area of quality appraisals concluded that the quality of the reporting of a study correlates with its value towards the final synthesis, and that excluding studies with a low quality in their reporting will have very limited impact in the findings of the meta-synthesis24. Therefore, only studies with the highest quality (CASP ≥ 15) were part of our synthesis. Any discrepancies in ratings of quality appraisal using CASP tool were discussed between the members of the quality appraisal team to reach consensus based on the evidence provided in the papers.

Regarding the quality of our review findings, the GRADE-CERQual (Confidence in Evidence from Reviews of Qualitative research) approach was used. This approach facilitates assessing how much confidence can be placed in each individual review finding by evaluating four different components: methodological limitations, coherence, adequacy and relevance[25]. To conduct the GRACE-CERQual analysis, the iSoQ interactive summary of qualitative findings platform was used in its beta version[522].

Meta-ethnographic synthesis

To inform the synthesis of the included studies, a meta-ethnographic approach was used. Meta-ethnography is a qualitative methodology originally developed by Noblit and Hare with an interpretative approach that facilitates "putting together" the qualitative research available through a process of translating the studies into one another[386] and into the researcher's own interpretation of the data. The use of meta-ethnography gives researchers the potential to produce new interpretations and

models[459]. To facilitate the reporting of this meta-ethnography, we followed the eMERGe reporting guidance[385].

Noblit and Hare (1988) proposed a series of phases that overlap and repeat along with the conduction of the synthesis[386].

Phase 1 and 2 – Selecting meta-ethnography and deciding what is relevant

The first two phases of meta-ethnography involve identifying a research gap that might be fulfilled by meta-ethnography which was done above. Phase 2 was completed by performing a systematic search of the literature as reported above and importing the resulting studies into NVIVO12 for analysis (See Figure 1).

Phase 3 - Reading the studies

Phase 3 involved the repeated reading of the included studies by three reviewers (TES, CN, KMS) and the independent extraction of the characteristics and details of the study into the data extraction sheets (See Table 6.1). In this phase, the studies were read carefully, and notetaking facilitated the identification of the data that needed to be extracted or assessed for quality.

Phase 4 - Determining how the studies are related

In order to identify common metaphors and concepts across the studies, line-by-line coding of the results and discussions section was conducted using NVIVO12 by one author (TES). The codes were refined as the coding progressed and new codes were created as necessary in each study. A second author (KMS) followed and checked the coding process to ensure its reliability and discuss nuances or disagreements.

Both first and second order constructs were extracted for analysis. In meta-ethnography, first order constructs are the participant quotes used by the original study's authors, and second order constructs are the original authors interpretations reached after their own analysis of the data. These concepts are used to differentiate between the research participant's experiences and the author's interpretations of such experiences[30]. The influence of the author's background over their

interpretations done in the different papers was difficult to assess, as the reflexivity accounts of most studies were very poor or missing. The key concepts that we chose to explore and compare in the analysis were related to facilitators or barriers that influenced women's weight management behaviours.

After completing the first coding phase, the content of each code was checked for consistency of interpretation and additional coding was performed when necessary. The resulting list of concepts was grouped into themes and categories through a thematic analysis process. We used tables to display the concepts and themes across all studies, classifying them into themes and categories, and then we used concept maps to establish and discuss the influences of each concept over the others.

Phase 5 – Translating studies into one another

In this phase, we continued to refine the themes and categories to ensure that the meaning of each individual study was reflected. The initial codes were examined, and themes were examined and combined thematically when describing similar findings.

The influence of each study over the different themes, categories and concepts identified in this synthesis is documented using references and quotes. Quotes were obtained from primary study participants and by primary author's explanations and interpretations. Additionally, a table with a summary of the themes, categories and concepts identified is also provided (Supplementary file 6.2). In this article we are presenting first, second and third level interpretations, based on the women's experiences.

Phase 6 - Synthesizing translations

The result of the translated concepts, their relationships and the primary data were used to create a textual line of argument, which is presented in the results section. Two authors were involved in the synthesis (TES, KMS) and the additional authors provided feedback and insights when necessary (SM, CO, KOD, MB, LL). The authors are from different disciplinary backgrounds including psychology, sociology, medicine, public health, epidemiology and behavioural science, which promoted discussion of potential different interpretations.

Phase 7 - Expressing the synthesis

The findings of this meta-ethnography are narratively presented in this article, additionally, a summarised version of the findings can be found in our CerQual assessment and summary tables (Supplementary file 6.2 and Supplementary file 6.3).

6.4 Results

6.4.1 Search outcomes

The first database search in March 2019, identified 4307 studies; the second search in January 2021 identified 80 additional records. Of these, 4030 remained after duplicate removal. After screening for titles and abstracts, 75 studies remained eligible for full text review. Following full text screening of the remaining 75 studies, 17 studies met the criteria for inclusion in the meta-synthesis (See figure 1).

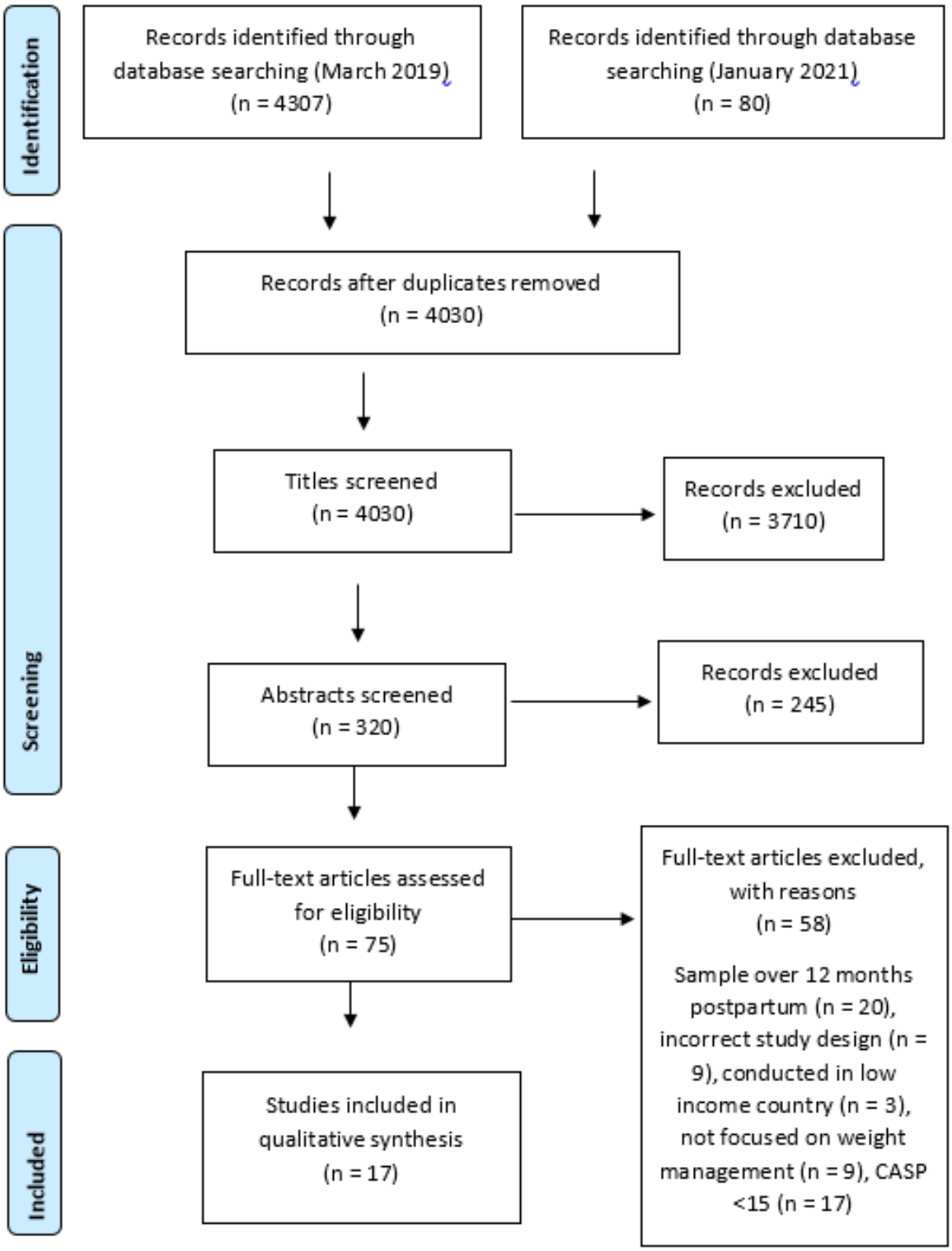


Figure 6. 1: PRISMA Flow Diagram showing the process of inclusion of studies. From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097

6.4.2 Study characteristics

Characteristics of included studies are shown in Table 6.1. Of the 17 studies included for analysis, four were conducted in the UK, six in the USA, three in Australia, three in Ireland, and one in Norway. The years of publication ranged from 2009 to 2020. The number of participants in the studies ranged from 11 to 58, with ages ranging from 17 to 46 years. Sixteen of the studies included pregnant women only, and one included both pregnant and postpartum women up to 12 months[31]. Fifteen studies used qualitative designs and two used mixed-methods designs. Twelve studies collected data using semi-structured interviews and five used focus groups. Regarding study quality, almost all studies performed poorly with regards to reflexivity, ethical considerations and reporting of their recruitment strategy. However, study quality was high due to clear aims, use of qualitative methodology, quality of report of findings and value of the research (Supplementary file 6.4). Results from the GRADE-CERQual analysis are presented in Supplementary file 6.3.

Table 6. 1: Study characteristics

Authors	**Denison et al. (2015)**	**Ferrari et al. (2013)**	**Faucher et al. (2020)**	**Flannery et al. (2020)**	**Flannery et al. (2018)**
Country	UK	USA	UK	Ireland	Ireland
Aims	To explore the barriers and facilitators to physical activity and lifestyle interventions in pregnant women with Class III obesity (BMI >40kg/m2)	Elicit from pregnant women their perceptions about provider advice regarding diet and physical activity in pregnancy.	Identify beliefs and attitudes about GWG, exercise, and proposed intervention	Explore overweight and obese women's experience and perception of dietary behaviours and weight management during pregnancy	Systematically identify the barriers and enablers to physical activity for women who are overweight and obese in pregnancy using the TDF and COM-B model.
Data collection method	Semi-structured interviews	Focus groups	Focus groups	Semi-structured interviews	Semi-structured interviews
Recruitment	Purposive-sampling from a specialist-led clinic providing care to pregnant women with class III obesity.	Sample was part of a larger study. Recruited through newspaper ads, posted flyers and prenatal clinics.	Women were recruited with flyers and HCP approaches in their birth centres	Women with a BMI of ≥25kg/m^2 were identified and recruited in a maternity hospital	Purposive-sampling of women with BMI of ≥25kg/m^2 in the public antenatal clinic of one maternity hospital.

Number of participants	13	58	17	30	30
Age	25-34 years	18-35 years	17-35	20-40+	
Pregnancy status	Pregnant	Pregnant	Pregnant	Pregnant	Pregnant
BMI required for participation	Class III obesity (>40kg/m2)	N/A	Pre-pregnancy BMI of 30 or greater	Between 20 and 40km/m2	BMI ≥25kg/m2
Timing of data collection	17-37 weeks gestation	27-30 weeks gestation	<14 to 38 weeks gestation	Across all trimesters but most women between 27 and 40 weeks gestation.	Across all trimesters
Method of data analysis	Framework approach	Thematic analysis	Content Analysis	Thematic analysis	Framework approach with inductive thematic analysis
CASP score	17	16	15	19	16

Authors	Garnweidner et al. (2013)	Groth et al. (2013)	Holton et al. (2017)	Keely et al. (2017)	Kominiarek et al. (2015)	Lee et al. (2018)*
Country	Norway	USA	Australia	UK	USA	Australia
Aims	To explore experiences with nutrition related information in routine antenatal care of an ethnically and socially diverse study population.	Understand how urban, low income, pregnant African American women view physical activity and how they approach nutrition while pregnant.	Describe women's experiences and perspectives of care for weight management during pregnancy in Melbourne, Australia.	To explore the experiences, attitudes and health-related behaviours of pregnant women with a BMI >40kg/m2	To examine and describe obese racial-ethnic minority women's knowledge, beliefs and attitudes about nutrition, exercise, and healthy lifestyles during pregnancy.	To assess and compare pregnancy nutrition recommendation knowledge and to explore how nutrition knowledge impacts on food choices in pregnant women and nutrition education practices of antenatal care providers.
Data collection method	Semi-structured interviews	Focus groups	Semi-structured interviews	Semi-structured interviews	Focus-groups	Semi-structured interviews

Recruitment	Recruited by midwives in eight mother and child health centres in Oslo.	Recruited from the Special Supplemental Nutrition Program for Women, Infants and Children (WIC) services and prenatal clinic waiting rooms	Recruited from Medical Centre using purposive recruitment strategies inc. Flyers and direct approaches from researchers	Purposeful sampling in a specialist antenatal clinic for women with BMI >40kg/m2.	Recruited at Women's health clinic via recruitment flyers or recruited by study personnel.	Convenient sample from eligible pool of pregnant women attending the hospital for antenatal care. .
n of women Participating	17	26	17	11	16	19
Age	average 28 years old	18-39 years	24-43 years (32.6 average)	26-40 years	21-39	<30 to 40+
Pregnant	Pregnant and postpartum	Pregnant	Pregnant and postnatal	Pregnant and postnatal	Pregnant	Pregnant
BMI required for participation	Pre-pregnancy BMI ≥25kg/m2	N/A	N/A	Pregnant women with BMI ≥40kg/m2	Prepregnancy BMI ≥30kg/m2	N/A

Timing of data collection	Before the 30th week of pregnancy and two months postpartum	Over 60% in their first 20 weeks of pregnancy	Late pregnancy (≥28 weeks gestation) and 4-6 weeks after giving birth.	2 interviews during pregnancy and 1 postnatally	Across all trimesters	Across all trimesters, 45% women last trimester
Method of data analysis	Interpretative phenomenological analysis	Content analysis	Thematic analysis	Thematic content analysis	Conventional qualitative data analysis (Thematic analysis)	Thematic analysis
CASP score	16	15	15	15	17	15

*Mixed methods study

Authors	Leiferman et al. (2011)	Marquez et al. (2009)	O'Brien et al. (2017)	Padmanabhan et al. (2015)	Reyes et al. (2013)	Sui et al. (2013)*
Country	USA	USA	Ireland	UK	USA	Australia
Aims	To elucidate unique barriers and facilitators to antenatal physical activity engagement among women of low socioeconomic status.	To understand how Latina women perceived, understood, and valued exercise to inform a physical activity intervention designed.	To explore the various factors within the life course that overweight and obese pregnant women perceive to influence their food choice and physical activity behaviours.	Examine pregnant women's weight-related attitudes and beliefs (including the weight-related behaviours of diet and physical activity during pregnancy).	Understand the perceptions of low-income, overweight, and obese, African-American mothers about diet quality in pregnancy, specifically focused on what facilitators and barriers exist to eating healthy.	Describe overweight and obese pregnant women's views about making healthy behavioural changes during pregnancy.
Data collection method	Individual and paired interviews	Focus groups	Semi-structured in-depth interviews	Semi-structured face to face interviews	Semi-structured, individual interviews	Face-to-face interviews

Recruitment	Passive print advertisement Via health care clinics and community organisations	Purposive sampling waiting room at the time of patients pre-natal appointments by bilingual/bicultural interviewers.	Purposive sampling from patients attending the ODP	All participants were previously recruited to a prospective quantitative longitudinal study via invitation letters and participation information sheets.	Waiting room of a single university-affiliated outpatient prenatal care clinic, serving primarily Medicaid-insured patients.	Purposeful sample of women participating in larger study.
Number of participants	25	20	22	19	21	26
Age	18-46	18 to 40	Mean 32.3	19-38 years	≥18	not specified
Pregnant	Yes	Yes	Yes	Yes	Yes	Yes
BMI required for participation	N/A	N/A	BMI ranging between 25kg/m2 and 39.9kg/m2	N/A	Excluded in BMI <25	Criteria of larger study.
Timing of data collection	Second and third trimester	<28 weeks gestation	34th week of pregnancy	Third trimester	All trimesters in pregnancy	28 weeks gestation

Method of data analysis	Thematic analysis	Content analysis	Inductive thematic analysis	systematic thematic content analysis	Principles of grounded theory	Framework analytical approach
CASP score	15	15	16	16	16	16

*Mixed methods study

6.5 Synthesis

Synthesis of the included papers led to three themes being generated: (1) Awareness, beliefs and emotions about weight management; (2) Antenatal healthcare; (3) Social and environmental influence (See table 6.2).

Table 6. 2: Summary of themes and categories

Themes	Categories
Theme 1: Awareness, beliefs and emotions about weight gain and weight management	1.1 Knowledge and awareness
	1.2 Risk perception and decision balance
	1.3 Perceived control over health and weight gain
	1.4 Personal insecurities and sensitive nature of the topic
Theme 2: Antenatal healthcare	2.1 Interactions with healthcare professionals
	2.2 Antenatal education and sources of information
Theme 3: Social and environmental influence	3.1 Influence of others, support and social norms
	3.2 Social judgement and stigmatization
	3.3 Environmental and sociodemographic factors

Theme 1: Awareness and beliefs about weight gain and weight management

Category 1.1: Knowledge and awareness

Women included in ten reviewed studies discussed a general lack of knowledge about how to manage their weight, and many women were not aware of the appropriate range of gestational weight gain[523–525]. Women in nine studies demonstrated a general lack of awareness of the specific risks of living with overweight or obesity during pregnancy[523,525–531].

"I don't think there is a risk to me and my baby just yet. It's just, I think, the

number . . . I've gained 40 [pounds], so it's not like a lot . . . I mean, even like you said, you gained 70 [pounds] but you were fine. So, the number I feel like is tricky."[32] *[First order construct]*

Some women were not aware of the benefits of exercise[531–533] and healthy nutrition during pregnancy[531,534,535], with many women not aware what types or duration of physical activity is appropriate during pregnancy[523,526,527,536–539] or what types of food were recommended or considered healthy[531,534,535].

"I mean I don't know can you do certain exercises so I would be worried that I could pull a muscle so I would be extra cautious I suppose at the gym cause I'm afraid and I wouldn't really know"[537] *[First order construct]*

"I don't know what contains iron in food."[536] *[First order construct]*

Lack of knowledge and awareness was not universal and some women in two studies were aware of risks associated with being overweight pre-conception[531,534], and knew recommendations for physical activity and nutritional during pregnancy in three studies[526,538,539].

"I've been warned by the midwife that I've been overweight and it's so important that I should try to keep healthy."[531] *[First order construct]*

Women's knowledge and awareness about appropriate diet and physical activity during pregnancy[533,535] was seen as linked to a range of benefits in thirteen studies[525,526,537–539,527,529–534,536]. For instance, women reported that a healthy diet and physical activity could facilitate labour[523,526,530,531,538,539], enhance feelings of control over their body[526,538], improve baby's well-being[531,533,535,538], increase levels of energy[538], have general physical and mental health benefits[526,530,531,533,538,539], and facilitate social engagement with others[526,538].

'Someone was comparing giving birth to a marathon…the more active you are and the more limber you are, then the easier it is to give birth.'[526] *[First order construct]*

'They are both as important… diet because you are directly feeding the baby and you can control weight by eating the rights things. However, exercise…it's not only about weight control, it's about keeping your body moving and all sorts of other pregnancy things…'[526] *[First order construct]*

Category 1.2: Risk perception and decision balance

Our analysis suggests that women sometimes based their weight management behaviour decisions by weighing up advantages and disadvantages to the behaviours and assessing potential risks based on their knowledge. For instance, some women in two studies thought that risks associated with overweight or obesity during pregnancy are exaggerated[523,531] and considered that the risks associated with other factors such as smoking or drinking alcohol are much higher[529,530]. In addition, some women felt that eating in moderation justified their food choices[530] or that unhealthy choices can be compensated[529,537]. Whereas others perceived the pregnancy period as an excuse to have a treat[530] or overeat[523,525,526,530–532,537–539] as there was a perceived decrease in the pressure to lose weight and social perceptions such as the idea that women are "eating for two". Some women also reported associating hunger with baby's movements[530], as they believed that is the body's way to express the baby's nutritional needs[523,525,529,530,535]. Additionally, household work or active life was perceived by many women as enough activity to meet the physical activity recommendations in five of the studies[523,525,526,532,539].

"Oh, I think it feels like it gives you a free pass… I just think, 'Well, I'm gonna.'…this is likely to be my last baby, I can lose the weight once I'm done"[529] *[First order construct]*

"I eat quite a lot of salad but I also eat quite a lot of chips and I know that chips are not healthy but I like them (laughs), in my mum's house we eat a lot of veg and fruits, so I thought that was just enough really"[530] *[First order construct]*

"If she's moving, then it's like, 'okay, well maybe she wants something'. When she moves, it's like, 'maybe I'm hungrier than I feel. Maybe she needs something special."[535] *[First order construct]*

"I know I don't really exercise but, like I said, getting the kids ready and walking to the car—I feel like that's sufficient enough for me to exercise."[525] *[First order construct]*

Perceived negative outcomes of behaviours were discussed by women in ten studies who expressed fears of harming the baby, having a premature birth or having a pregnancy loss while doing exercise[523,525,526,530–532,537–539]. For many women, feeling their body physically challenged was deemed as a risk[525,530,531] and so they decided to avoid exercise as their fears were stronger than the perceived benefits[530]. In two studies, women with previous fertility issues or history of pregnancy loss expressed that they were especially concerned about the safety of physical activity during pregnancy[531,537].

'I've seen pregnant people there and they're on the treadmill and I think 'cool yourself'…you've got your baby bouncing up and down and then you've got your fat on top of the baby and it's just, you know, you could give them brain damage.' [526] *[First order construct]*

However, for women in three studies, while weight gain was acceptable during pregnancy, weight retention afterwards was not[525,530,531], which led to them engaging in weight management behaviours. Some women with more than one child discussed learning from their previous pregnancies and changing weight management habits in their current pregnancies to ensure they would lose any excess weight after birth[524,538]. A feeling of satisfaction was also expressed by those women who saw the results of their efforts to manage their weight in their own body[530]. Some women in five studies felt a sense of responsibility for the health of their baby[524,530,531,533,536] which in some cases influenced their decision to prioritise healthy eating[523,524,530,533,535].

"Now because I only have a short time to go, I look at the scales and it's a big achievement, and that's brilliant, as what I have gained has been sufficient for the baby, but not to put on myself if you like, so I'm actually quite proud of myself[530]

"Now I'm not eating a lot of greasy foods… (Before pregnancy) I wouldn't say

I didn't care, but I got another life growing in me, so I don't want to jeopardize my life and the baby's life."[535] *[First order construct]*

Category 1.3: Perceived control over health and weight gain

The findings show that women in three studies perceived limited control over their own health[533,536], that pregnancy is risky by nature, and that complications occur randomly[529]. Some women also felt they did not have any control over their weight gain[523,525,530], which some women felt was a justification to "indulge"[524–526,531]. Additionally, some women reported that their food choices were driven by cravings, appetite and taste[524,530,532,533,535].

"You can't control it, cuz that baby controlling it for you."[525] *[First order construct]*

"According to our participants, biology and environment can conspire to make managing dietary intake feel beyond their control."[523] *[Second order construct]*

Many of the women in fourteen studies experienced physical symptoms during pregnancy that were beyond their control, which acted as barriers to weight management[523,524,536–539,525,526,528,530–533,535]. Barriers to physical activity included having a higher risk pregnancy[531,537,539], lack of energy and tiredness[526,530–533,535–539], reduced mobility[537–539], nausea, vomiting and other pregnancy related pains and discomforts[526,528,530–532,537–539]. Barriers to a healthy diet included nausea, vomiting and aversion to certain foods[523,524], lack of energy to cook[535] or making food choices based on appetite and taste[524,530,532,533,535].

'I must admit a few weeks ago I tried [exercise] and I ended up in big trouble, I felt so sick, my head was swimming and my pelvis was killing and the baby wasn't happy and I thought no I've pushed, you know, when you've pushed it too far'[526] *[First order construct]*

"I think I gained weight due to severe morning sickness. The only thing I could eat was bread which helped to stop the nausea and heartburn. I ate bread even when I wasn't hungry as it alleviated the alkaline taste on my tongue."[528] *[First order construct]*

While women in four studies indicated they were aware of behavioural changes they were recommended to engage in, translating that awareness into action was experienced as a challenge[526,529,531,532]. Women who lacked an established exercise routine[532,537–539] or had poor pre-pregnancy dietary habits[525,529,532,533,535], experienced greater difficulties in changing their behaviours when becoming pregnant. This is also tied to motivation, and women in four different studies discussed lack of motivation as a barrier[526,532,538,539] mostly for physical activity but also for healthy eating. Some women expressed that they disliked exercise[531,539] or cooking[531], they felt too tired or 'lazy'[532,538,539] or that they needed an external motivation to remain active[532]. Conversely, women in six studies expressed that having established positive pre-pregnancy healthy habits acted as a facilitator to maintain these habits during pregnancy[529,531,535,537,539].

" […] Like, I can't do anything about it. I mean, I know all this… I mean I've studied this so much… like… I could be a dietitian probably! I just can't implement it, for whatever reason, like… know what I mean?"[529] *[First order construct]*

"If I've already got an exercise routine, then stick with it… and just, if you are eating healthy, keep up with that."[531] *[First order construct]*

Not all women experience lack of control, some women in two studies had a perceived high level of control over their weight gain in terms of their dietary intake and physical activity.

"I think it's more because of my diet and everything and the way I am eating and I can actually see that you know it is working, not eating too much […] so I think that helps you know, that you can physically see that I am in control"[530] *[First order construct].*

Theme 2: Antenatal healthcare

Category 2.1: Interactions with healthcare professionals

In this theme, we identified factors related to healthcare professionals' attitudes that have an influence on women´s capacity and motivation to engage in weight management behaviours as expressed in five different studies[523,524,529,530,537]. Some women felt embarrassed and judged when dealing with healthcare professionals, especially with interactions regarding weight[524,529]. Women reported insensitive and judgmental attitudes[529] and communication[524] from healthcare professionals, including in specialised clinics[523,529]. Women felt they were assumed to have bad habits because of their overweight[529] or that they already had enough knowledge about weight management as they had children previously[530,537].

> *I had a very bad experience during my first pregnancy. I was 29 weeks and I went in to see my consultant and asked him if I could find out the sex of the baby but he just pinched my stomach … I felt very upset. I think they turn off when you are a little bit overweight. And they think oh she's after letting herself go."*[524] *[First order construct]*

> *'…what I found different was when they know that you have children already they kind of thinking that you know everything which is not true…you may forget […] but they seem to assume because you have had other children you know already what to do'*[537]

Some women from four of the studies expressed defensive or avoidant attitudes that might potentially be a consequence of the negative feelings associated with overweight and the perception of weight management as a sensitive topic to address[523,527,532,536]. This resulted in some women feeling challenged when receiving advice regarding their dietary behaviours. Some women in 3 studies considered BMI charts and ranges of weight gain as "a lie"[525], rejected standardised GWG goals[523], or expressed relief when the conversation about weight management with their providers was avoided[527].

> *"I was very frustrated my first pregnancy because my midwife was very keen*

on nutrition and, "Don't gain too much weight," and "We don't want to have a really big baby." And I wasn't gaining any weight at all. And, so the fact that she was harping on it to me made me very angry […] It made me almost want to neglect the nutrition aspect, because I felt like she wasn't listening to me personally."[523] *[First order construct]*

"I don't feel like I'm the typical obese person, you know. They say I'm obese, and I'm like, well, I don't know how you figure I'm obese. How do you classify obesity? I don't like that word because I don't feel—I know I'm big, but I'm not as big as most."[525] *[First order construct]*

The feelings described by the women in this section, together with a perceived societal influence, were reflected in women's perception that weight and weight management are sensitive topics to discuss in their antenatal care[524,525,527–529,531]. As observed in six studies this acted as a barrier that minimised discussion opportunities and hindered knowledge exchange[524,525,527–529,531]. However, some women from two studies felt that, even though it can be a difficult conversation, it is important to discuss weight and weight management with their healthcare professionals[525,531].

"I feel very insecure about my weight … [the midwives] really tried not to mention it or make me feel uncomfortable"[37,48] *[First order construct]*

"They reported that although it "hurt" when providers discussed their weight, they knew they were being truthful and ultimately it helped them."[34] *[Second order construct]*

However, finding supportive and non-judgemental healthcare professionals was also expressed by some women as important[527]. These women reported that sometimes they found it easier to speak to healthcare professionals about their weight management than speaking with the people in their close social context[527].

Category 2.2: Antenatal education and sources of information

Women included in eight studies discussed complaints about the availability of sources of information, or were not satisfied with the education and advice they received during their antenatal care[524,527,528,530,531,534,536,537]. Women perceived that there was a lack of time to discuss weight management advice[528,536,537] or appropriate weight gain[523–525,527–530,537] during their antenatal care, and that clinical aspects were prioritised[524,527,537]. The information received about nutrition and diet while pregnant was perceived as scarce[527,534], too generalised[524,527,530,534,536], and not tailored to individual needs[527,531,536], leaving women dissatisfied[524,527,528,530,531,534,536,537]. Additionally, women reported that nutritional advice was focused on food safety only (i.e., preventing food-borne diseases), rather than weight management in four of the studies[524,527,528,534]. Regarding physical activity during pregnancy, women also expressed dissatisfaction in seven of the studies[526,528,530,531,536–538]. The support received in relation to becoming more active was perceived as very limited[531,538], and advice tended to be perceived as too conservative[530,536], hesitant or unclear in recommendations[536,537].

> *"They don't tend to offer any advice good or bad in terms of weight management and activity and stuff like that. It's more the blood pressure, checking the baby and stuff like that"*[537] *[First order construct]*

> *"It was about the food I couldn't eat. Like some types of raw fish and pasteurized milk and cheese, as I recall it?"* [527] *[First order construct]*

> *"Nobody told me nothing [about physical activity]. They gave me some brochures [chuckling] and that's it."*[536] *[First order construct]*

Women in five studies felt that they were provided with useful nutrition and physical activity information during pregnancy, which acted as a facilitator to actively engaging in weight management[524,527,528,534,538]. Valuing and trusting providers opinions and advice was seen as beneficial in three studies[527,528,536].

The mentioned lack of discussion with healthcare professionals led some women to seek out other sources of information[524,527,534]. Some women found that having

access to different sources of information was useful[526,527,534]. However, some women considered that the amount of information from the multitude of sources available was overwhelming[527,530,536,539], especially in the cases where they would find conflicting or contradicting information[525,531,534,536,537].

> *"I saw a dietician at the pre-pregnancy clinic [due to diabetes]. She gave me useful information about food groups and healthy eating during pregnancy. I think other women would benefit from similar information."*[528] *[First order construct]*

> *"You know, it doesn't say that much to me. It's very, very much information you have to absorb during few consultations. I honestly have to admit that not all information is processed."*[527] *[First order construct]*

Theme 3: Social and environmental influence

Category 3.1: Influence of others, support and social norms

In eight studies, women discussed how the dietary habits of their social circle influenced their beliefs and behaviours. In some instances, women reported that they had no decision power over shopping or cooking choices in their families[523,525,535], and felt the need to adapt their own nutrition habits to other people[525,533]. Women in four studies also struggled when their family or partner encouraged them to overeat[523–525,535] with some women describing their partners as 'a feeder'[524]. The misconception that women "need to eat for two" was shared amongst their wider circle. Some of the women in four studies reported that they relied on their family and friends for advice regarding weight management[524,525,527,538]. This advice sometimes reinforced women's own misconceptions, while some disagreed with advice they received[525,527].

> *"The food that my mother buy [gets in the way of me reaching my GWG goal]. She don't buy healthy food, she, cuz my brother, and they all like fried chicken... She don't buy plain chicken."* [525] *[First order construct]*

> *"My family says eat as much as I want. And just keep eating, 'Cause it's good for the baby."*[535] *[First order construct]*

"I don't know what to follow. I don't know whether to obey what the nurse says about lying down or her [my husband's aunt]."[536] *[First order construct]*

For some women in seven studies, the lack of support and the lack of role models acted as a barrier to engage in physical activity and healthy eating[525,526,530,531,537–539]. Some of these women explained that their family members encouraged them to rest[539], and even disapproved of their exercise[525]. However, availing of social support acted as a facilitator to engage in weight management behaviours for women in nine different studies[524,526,531–534,537–539]. Having positive role models in their family who embedded positive physical activity and dietary habits in their childhood influenced their choices as adults[533].

"Lot of people have constantly said to me throughout my pregnancy, you need to rest, you need to rest, you need to rest. I don't really understand why I need to rest. If my body's not telling me that I need to rest you know, then why do I need to rest? … so it's been quite difficult."[526] *[First order construct]*

"Well, just my husband and both my brothers, they all work out together, and me and my cousin work out, so everybody around me works out, so that kind of helped me."[538] *[First order construct]*

Some cultural differences also acted as a barrier in three different studies[525,527,533]. In these studies, migrant women expressed how their cultural dietary habits or lay food beliefs did not match official health recommendations. Thus, advice was perceived as contradictory and challenging in some instances, especially when it involved additional family members with authority over the women[525,527,533].

"You know, that's really weird, because in Norway you are told to eat eggs and fish, however in Pakistan you should stay away from it in the first three months of pregnancy."[527] *[First order construct]*

"You are in trouble when the elders say something and the midwife says something else. Especially your mother in law. She has much influence, especially during the first pregnancy. It is really difficult sometimes to decide

what I should eat."[527] *[First order construct]*

Category 3.2: Social judgement and stigmatization

Many of the women that participated in ten studies had negative feelings about being overweight[525,526,528–531,534,537–539]. Women expressed feelings of shame, guilt and regret for not being active[529,537] or making food choices that they perceived as unhealthy[534]. For women in five studies, these feelings led them to have problems with their body-image and self-esteem[526,528,531,538,539].

> *"I beat myself up for it [weight gain]. I'm like, oh, I'm just so depressed, and I don't wanna eat no more."*[525]

> *"It's going in like a swimming costume, it's definitely… Yeah, that's what puts me off 100%. It's nothing else… I'm like 'oh my God,' the thought of going swimming and people seeing me."*[526 539] *[First order construct]*

Women discussed feeling social judgement and stigmatization in five studies[526,528,529,531,533], which acted as a barrier to engaging in weight management activities. Women felt they were treated differently if they were overweight[523,528]; for instance, they felt questioned and judged as "greedy" [529,530] or "lazy"[530], and reported that they had to hear to everyone's opinion about their weight which was sometimes accompanied by derogatory language[530].

> *"When I was in the [hospital] waiting room, people looked at me differently … people think that you don't look after yourself or take care of yourself when you are overweight."* [528] *[First order construct]*

> *"I think the thing as well…they think you're just sitting here stuffing pints of Ben & Jerry's, like… that's not what my life is like…" [tearful].*[529] *[First order construct]*

Category 3.4: Environmental and sociodemographic factors

Regarding healthy eating, women in three studies reported that a lack of access to healthy food in their communities or workplaces acted as a barrier to engaging in dietary weight management[525,531,535]. Another barrier to healthy eating was the

affordability and easy access to fast food in the women's communities as reported in five studies[523,525,530,532,533], which led them to perceive healthy foods are more expensive and hard to access[533,535]. Additionally, some women perceived that it was more convenient to buy ready meals than cooking at home[525,532].

> *"I don't drive. So, when I get to the store, I have to go shopping for the entire month, because getting on the bus to go get groceries, it's too much."*[535] *[First order construct]*

> *"I think city life is probably not good for me... Takeaways and stuff, everything is delivered, you find stuff to do that's not even that active, like go to the cinema... it's just too easy to be bold"*[533] *[First order construct]*

> *"It used to be expensive for Cornflakes and so cheap for porridge and it was always if you had less money you made you already made better choices... Now I don't know how they are doing chocolate and doughnuts so cheap... The people eating free--range eggs and wholegrain bread are middle class... They are educated people"*[533] *[First order construct]*

Women also reported environmental barriers that influenced their capability to remain physically active during their pregnancies. Urban environments were considered unsuitable for outdoor exercise[533], especially in neighbourhoods that were perceived as unsafe[538,539] or where the weather conditions were not suitable for outdoor sports[530,537–539]. Women from five different studies expressed that they had very limited access to sports facilities[526,530,533,538,539], and that the affordability of such facilities was a big barrier to exercise[526,530,531,533,537–539]. Additionally, women reported that not having programmes tailored to pregnant women was an issue for them to engage in physical activity[526,530,537,539]

> *"If you live in a bad neighborhood… it's not safe to exercise…You don't do that…"*[539]

> *"I was looking for swimming baths…that I could afford [as unemployed]…I do like going to the gym. I have had to give up my gym membership because I'm*

not working and I can't afford it."[526] *[First order construct]*

Many women from eight different studies expressed that they were not able to engage in weight management behaviours due to a lack of time[525,526,530,532,533,537–539]. For some women, their family and/or life acted as a barrier because it gave them limited time to cook and exercise[530,531,534,537,539].

"We tend to eat a lot of convenience food because I'm working full time and more things like fish fingers, chicken nuggets […] its always just whatever is in the freezer type of things"[530] *[First order construct]*

"I do find it, I would find it difficult to go out and do a proper exercise routine, because I just physically don't have the time I don't get in much until 6pm and I leave the house at 6am"[530] *[First order construct]*

Some socioeconomic factors were also associated with the barriers and facilitators that women reported regarding their weight management. In four studies reviewed, women living in low-income environments found additional barriers to manage their weight[525,533,538,539], whereas women with higher educational level[527], higher socioeconomic status[533], multiparity and older age[533] reported more facilitators.

"I suppose just as the years progress, you know, you have to think for the future, because what you do right now will definitely benefit you for the future. And that's the way I look on it, you know. I think in your early twenties, you're out and what not and you know, your whole perspective on life completely changes"[533] *[First order construct]*

Some environment facilitators to exercise were discussed. Women reported that affordable facilities[537,539], transportation and/or built environments suitable for physical activity, and having the right weather conditions acted as facilitators[538].

"I probably would be less active if they (trails, parks) weren't there. I do walk up and down the neighbourhoods […] when I am really motivated I do the trail because the trail is long. But, yeah, I do think I would be less active if I didn't have that (trail)."[538] *[First order construct]*

6.6 Discussion

In this synthesis, we identified factors that act as barriers and facilitators to women's willingness and ability to engage in weight management behaviours during pregnancy, which in this review were mostly related to physical activity and following dietary advice. The main themes identified were: (1) Awareness and beliefs about weight management, (2) Antenatal healthcare, and (3) Social and environmental influence. The main barriers identified were a lack of knowledge about the risks of overweight and obesity during pregnancy and lack of awareness about the recommendations regarding physical activity and diet. Women who had pre-established unhealthy habits experienced additional challenges to adopting recommendations during pregnancy, whereas women who already had healthy habits found it easier to maintain these habits. Antenatal healthcare experiences also influenced women's behaviour. Women who had negative interactions with healthcare professionals felt shamed and stigmatised; this made weight management discussions with their healthcare professionals problematic and acted as a barrier to engaging in weight management behaviours. Further, women reported a perceived lack of prioritisation of weight management during their antenatal care from their healthcare professionals, which led them to feel like the discussions were rushed and the advice very scarce or limited. On the other hand, having adequate awareness of the recommendations regarding physical activity and diet during pregnancy, as well as how to engage in these behaviours safely acted as facilitators. Women who were aware of the potential risks of being overweight during pregnancy and that had a high level of perceived control over their health were also more keen to engage in weight management behaviours. Additionally, encountering supportive healthcare professional and having access to sources of information and discussion was also positive for women. Additionally, women's social contexts also played an important role in their engagement in weight management behaviours.

Our findings suggest that women's level of knowledge and awareness regarding weight management recommendations influences their engagement and this is in line with previous findings that found women who were informed about physical

activity held more favourable attitudes towards it[540]. This lack of knowledge might be contributing to misconceptions about the risks of engaging in weight management behaviours during pregnancy, which together with the collective perceived fragility of pregnancies, led women to have increased concerns and fears regarding the safety of their pregnancies.

Lack of knowledge and awareness about the recommendations regarding healthy nutrition and physical activity during pregnancy might also contribute to women's perceived low self-efficacy. This is in concordance with previous research conducted in the Netherlands examining socio-cognitive determinants of physical activity, which concluded that possessing knowledge and skills to engage in healthy behaviours improves confidence to overcome other types of barriers, and increases the perceived benefits of changing behaviours[541]. Additionally, our findings suggest that women perceive dietary advice to be predominantly focused on food-borne illness prevention rather than weight management. This might be related to healthcare professionals need to prioritise safety when giving nutritional advice to pregnant women. Although it has been shown in previous research that information provision alone is insufficient to produce behaviour change[363], providing women with clear, concise and practical information regarding physical activity and healthy eating during pregnancy might not only contribute to temper such misconceptions and highlight the benefits of these behaviours, but it might also increase the women's perceived control over their own health, as well as their levels of self-efficacy and motivation.

Findings of our review indicate that social stigma associated with overweight and obesity in general, and especially during pregnancy, led some women and healthcare professionals to feel that weight management is a sensitive topic to discuss. Consequently, some women responded with avoidant attitudes and some others adopted a defensive response. The NICE guidelines for weight management during pregnancy suggest that healthcare professionals discuss dietary and physical activity habits with women at the first opportunity to address concerns and myths, and advise women about the benefits of physical activity and healthy nutrition during pregnancy[510]. However, a previous study exploring healthcare professionals

approach to weight management identified that their perceived stigma and consequent "cautious approach"[542] were interfering in the care provided by healthcare professionals[542] in concordance with our review findings. These findings are also supported by previous examinations indicating that professionals from a range of specialities reported that weight is a difficult topic to discuss with potential to damage their relationship with their patients, and sometimes represents an inappropriate use of their time[543,544]. Such attitudes might influence the quality of information that women are receiving, as the conversation with HCP regarding weight management in our findings has been shown to be quite limited. This lack of discussion with healthcare professionals about weight management during pregnancy also likely contributes to women's lack of knowledge about aspects of weight management. This in turn led some women to resort to other sources of information, which women reported to include conflicting information. Providing trustworthy sources of information and encouraging discussion about weight management during pregnancy with healthcare professionals is important to overcome this barrier.

Previous research demonstrates that unhealthy social eating environments are a risk factor for obesity[545]. Based on our findings, issues such as the influence of the family context, the lack of support and role models, or the influence of social norms and stigmatisation of overweight during pregnancy will have an influence on women's engagement in weight management behaviours. Additionally, physical environments play a role in women's engagement in weight management behaviours during pregnancy, as shown in our findings. The quick and easy access to fast food and lack of access to affordable exercise facilities reported by women were probably contributing to women's low engagement in weight management behaviours, as also shown in previous research[546]. Previous studies have shown how altering elements of people's environments helped modulate their behaviour[547]. Decisions at policy level regarding free access to exercise facilities for pregnant women and/or promoting barriers to accessing fast food such as price increases or location regulation would be very beneficial. As such, interventions at individual and

population levels may enhance women's engagement in health promoting and weight management behaviours during pregnancy.

Interventions need to be designed taking into consideration all the different levels of influence over women's weight management behaviours. The factors identified in this synthesis speak to inter-related influences and levels, which can be understood using the socioecological model. This model states that there is no one single factor influencing people's health, and it sees health levels as the interaction among many factors at five different levels[548]. The five different levels are 1) individual, including knowledge, attitude and skills; 2) interpersonal, including family, friends and social networks; 3) organizational, including social institutions; 4) community, including relationship between organizations and 5) public policy, including national, state, local and legal regulations[548]. Hence, this framework highlights the importance of including all these levels in intervention design in order to address all the social determinants of health involved in a certain issue.

Our themes are similar to findings exposed in the literature[549,550]. However, we believe that our analysis has been able to identify further nuances into the facilitators and barriers. Additionally, our study differs from the ones conducted before in that we are not only interested in women's experiences of gestational weight gain, but in pregnant women's experiences of engaging with weight management behaviours to either lose excessive weight or manage their gestational weight gain, and also, it is not focused on a specific type of population (e.g: ethnic group, etc.).

There are limitations to this study. Firstly, the number of databases used for this study was limited; however we chose the selected databases based on appropriateness with the topic and similar studies in the area. Secondly, to ensure our findings were derived from robust, high-quality studies, we excluded some studies based on quality standards. While this might have contributed to losing some relevant input, we are confident that the findings presented are from high quality primary evidence. Thirdly, the findings resulting from this synthesis are based on women's experiences and opinions, and do not include partner's, healthcare professionals' or policy makers' perspectives on the issue; further research should be

conducted to explore facilitators and barriers from other stakeholder's points of view on this issue. Additionally, our findings are a result of a secondary analysis of primary data, which was collected and analysed by the studies' primary authors. This means that our own background might potentially have an influence on the review findings. To address this potential limitation, we have tried to remain transparent about the influence of each study over the different findings and conducted a GRADE CERQual assessment of confidence in our review findings.

6.7 Conclusion

Our review findings show that women's weight management behaviours are influenced at multiple levels during pregnancy. Pre-established habits were shown to be important predictors of whether women adopt recommendations during pregnancy, hence, interventions to reduce obesogenic environments and generally promote healthy habits in relation to physical activity and diet at a population level have the potential to also benefit women during pregnancy. Additionally, women would benefit from clear and direct sources of information and improved discussion with healthcare professionals. Development of interventions to foster and maintain weight management behaviours during pregnancy should consider women's awareness and beliefs, habits and motivation levels, social influence and their experiences in antenatal care. Such interventions should also include family members or other people in women's social circles to facilitate engagement and adherence. Provision of additional training regarding the complexities of weight management during pregnancy for healthcare professionals would also help them to further develop key practical and emotional skills to better support the pregnant women under their care to engage in appropriate weight management behaviours. Such approaches are needed to minimise adverse pregnancy outcomes of high gestational weight gain for women and infants.

6.8 Inter-chapter synopses

Chapter 6 aimed to aimed to identify the facilitators and barriers to weight management behaviors during pregnancy. Findings highlight that the facilitators of weight management behaviors included having social support, access to reliable information, and positive attitudes towards weight management. On the other hand, the barriers to weight management behaviors included a lack of knowledge, social pressure, and cultural beliefs. The study also identified healthcare professionals' attitudes and approaches towards weight management as an important factor in supporting or hindering weight management behaviors during pregnancy. The main learning points of Chapter 4 to 6 are that understanding the facilitators and barriers to the different modifiable/behavioural risk factors for stillbirth and their complexities during pregnancy is essential to develop effective interventions and support pregnant women in managing their behaviours during pregnancy. Chapter 7 aimed to explore the experiences and knowledge of first-time mothers about behavioral risk factors for stillbirth. The studies conducted so far in this Thesis had highlighted that there was a need for better education and so in this study, one of the aims was to identify whether that findings applied to our research context and if so, what were the bests approaches to information provision according to their attitudes and experiences.

Chapter 7 – Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth

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Chapter 7 – Exploring first-time mothers' experiences and knowledge about behavioural risk factors for stillbirth

7.1 Abstract

Background

Modifiable factors such as substance use, lack of attendance at antenatal care, overweight or obesity, and sleeping position are associated with a higher risk of stillbirth. This qualitative study aimed to explore women's experiences of modifiable factors during pregnancy and their awareness of stillbirth.

Methods

Purposive sampling was implemented by hospital staff in a large tertiary maternity hospital in Ireland between November 2020 and March 2021. Women were approached during their stay in the hospital and were invited to participate in a semi-structured interview 3 to 5 months later. Eligible women were primiparous, >18 years of age, and had an uncomplicated pregnancy and delivery. Eighteen women who consented to be followed up were interviewed at 3 to 5 months postpartum. Thematic analysis was used to analyse the data.

Results

Four themes were identified: attitudes towards behaviour change, awareness regarding stillbirth and risk factors, the silence around stillbirth and risks, and attitudes towards receiving information about stillbirth. Women spoke about behaviour change in terms of outcomes, and most changes (e.g., ceasing alcohol consumption) were perceived as easy to manage. Awareness of stillbirth was limited among the women interviewed, and the association between risk behaviours and stillbirth was not known by any woman. Results suggest that there is a silence around stillbirth, including in antenatal care, which hinders information provision. However, most women highlighted the value of receiving information and extra education about modifiable risk factors and stillbirth.

Conclusion

There is a general lack of understanding of the link between behavioural risk factors and potential pregnancy outcomes such as stillbirth. Providing further information to women about stillbirth and providing additional support with behaviour change might contribute to enhancing preventive efforts.

Patient or Public Contribution

Patients were involved in this study by providing their experiences of antenatal care which were used as primary data.

Keywords: stillbirth awareness, risk factors, antenatal education, stillbirth prevention

7.2 Introduction

Stillbirth is one of the worst outcomes pregnant women and their families can experience[551,552]. Worldwide, the estimated average rate of stillbirth (defined as a baby being born with no signs of life at 28 weeks gestation or more) in 49 high-income countries is 3.5 per 1000 total births, with rates varying from 1.3 (Iceland) to 8.8 (Ukraine) depending on the country[6]. In Ireland, according to the latest report published by the National Perinatal Epidemiology Centre, the stillbirth rate was 4.06 per 1000 births in 2019, which reflected a slight increase compared with 2018 data[8]. The overall perinatal mortality rate has remained flat in Ireland for several years, as opposed to the decrease observed in the decade before 2012[8]. Although the rates of stillbirth in high-income countries have reduced in the last 20 years, the variation between rates in high-income countries shows that a further reduction of stillbirth incidence is possible and needed[47].

Previous research has associated different types of risk factors with an increased risk of stillbirth, including medical[70–72], behavioural factors[185,254,310] and sociodemographic factors[1,60,61,69]. Some of these risk factors are potentially modifiable and addressing them could contribute to reducing the rates of stillbirth[21,341]. Previous research has associated substance use including smoking, alcohol use, and illicit drug use with an increased risk of stillbirth. Flenady et al. and Marufu et al. conducted two separate meta-analyses examining the influence of smoking on the risk of stillbirth, and both concluded that smoking was associated with a 36% and 47% increase respectively in the risk of stillbirth[87,134]. Regarding alcohol consumption, Aliyu et al concluded that mothers who consumed alcohol while pregnant were 40% more likely to experience stillbirth as compared with non-drinking mothers[310]. Additionally, a recently-published study exploring the risk of stillbirth in women who consumed alcohol and smoked in the antenatal period concluded that the adjusted relative risk for all stillbirths was 1.75 (98.3% CI, 0.96-3.18) for double exposure, 1.26 (98.3% CI, 0.58-2.74) for drinking only, and 1.27 (98.3% CI, 0.69-2.35) for smoking only compared with the reference group[138]. Being overweight and obese has also been associated with an increased risk of stillbirth. A recent study exploring prognostic variables for stillbirth found that the most

frequently reported maternal characteristic associated with stillbirth was high BMI and other measures of obesity, and these findings were supported by highly convincing evidence in most of the studies included in their review [553]. Engagement with and attendance at antenatal care has also been linked to the risk of stillbirth. Stacey et al. concluded in their study that attending less than 50% of the recommended visits was associated with an almost 3 times increased risk of stillbirth, and this risk increased as the number of visits attended decreased[206]. Lastly, women's sleeping habits have also been studied in association with stillbirth risk and research has specifically focussed on the maternal sleep position. Cronin et al. conducted a meta-analysis which concluded that the supine going-to-sleep position is independently associated with late stillbirth[554].

Even though there is evidence in the literature associating these modifiable risk factors with stillbirth[341], information about the role of these behaviours in stillbirth is not widely available amongst the public in Ireland, as demonstrated by Nuzum et al. in their survey study[258]. These authors, after surveying 999 members of the Irish population, reported that there is a lack of public awareness of the incidence, causes and risk factors for stillbirth, although over 50% of respondents personally knew someone who had experienced a stillbirth[258]. These findings are similar to those obtained in a survey study conducted amongst members of the American College of Obstetrics and Gynaecology[555]. The authors concluded that knowledge regarding epidemiology, risk factors, and effective interventions to reduce stillbirth amongst participants was only fair. In the same study, only 30% of respondents were aware that preeclampsia, advanced maternal age, elevated α-fetoprotein, multiple gestations, cigarette smoking, illicit drug use, and being post-term increased risk[555]. Regarding knowledge of behavioural risk factors, previous studies have also demonstrated that some women held serious misconceptions about the risks of substance use or engaging in weight management behaviours, as well as having limited reproductive knowledge, which interferes with their care-seeking behaviours and increases their risk of stillbirth[410,556,557].

Previous research has also shown that stillbirth is still a taboo subject in society[27]. Hence, further efforts to increase women's awareness about stillbirth and its risk

factors through routine antenatal care and public health campaigns are necessary to support stillbirth prevention efforts.

Involving patients' experiences in designing and developing public health campaigns and behaviour change interventions is vital to ensure that they are tailored to users' needs. Previous research has established that women engage in behaviour change during pregnancy due to different motivations, but the volume of expectations placed on them and the complexities of those changes are rarely acknowledged[558]. Understanding women's experiences of behaviour change and information provision during antenatal care through their perspective is essential to inform the development of such interventions and public health campaigns. The main objective of this study was to explore women's experiences of modifiable factors during pregnancy and knowledge and beliefs regarding behavioural risk factors related to stillbirth. Additionally, we aimed to examine women's experiences, if any, of being informed about such risk factors during their antenatal care.

7.3 Methods

To enhance the reporting of this study, the Standards for Reporting Qualitative Research (SRQR) checklist has been used[559] (See supplementary file 7.1).

7.3.1 Design

A qualitative semi-structured interview study was conducted using a Reflexive Thematic Analysis approach (See supplementary file 7.2). Qualitative research allows the researcher to explore and understand social phenomena and psychological concepts such as experiences, beliefs, motivations and attitudes[380]. For this study, a constructivist paradigm was utilised.

Constructivism is based on the idea that knowledge is built from an interaction between the subject and reality, and hence, the same situation experienced by different people might result in different interpretations, depending on each individual's perception of such situation[380]. This epistemological approach refuses the idea that there is only one truth but instead supports the importance of taking into consideration the subjective meaning given to a phenomenon and the

understanding of the context of the same[380]. In constructivism, the researcher and the study participants are equally involved in the process to generate knowledge, hence, the researcher has an active role in the research processes by creating -rather than revealing- something which has to make sense within existing frameworks of meaning[380].

This paradigm allowed us to understand the complexity of our participants' lived experiences from their point of view.

7.3.2 Recruitment

Women were recruited using a purposive sampling approach implemented by hospital staff in Cork University Maternity Hospital (CUMH), Ireland. Inclusion criteria included primiparous women, with a low-risk pregnancy and uncomplicated delivery, who had a healthy baby and were 18 years old or older and consented to participate in a one-to-one interview 3 to 5 months postpartum. The follow-up timeframe of 3 to 5 months post-partum for interviews was chosen to facilitate women adapting to their new routine with their babies before taking up their time to participate in the interviews.

Exclusion criteria included multiparous women, women currently pregnant, women with a history of pregnancy loss, women attending high-risk antenatal clinics, congenital anomalies, cases of maternal morbidity, and cases of major complications during pregnancy or birth for both woman and baby. Given the aims of our study, it was decided that obtaining the experiences of primiparous women with uncomplicated pregnancies and deliveries would reflect the common practices regarding information provision and knowledge acquisition during antenatal care. It was assumed that including women with additional care needs during their pregnancy or delivery would have resulted in a biased picture of the normal day-to-day practices of the antenatal care services, as these women require additional education and are exposed to different types of healthcare professionals and levels of care.

Women were invited to participate in the study during their stay in the maternity hospital after delivering their babies from November 2020 to March 2021. If eligible, an obstetrician affiliated with our research group explained the study to the woman and provided them with the patient information leaflet and consent form. After providing women time to read the information and ask questions, their written consent was obtained by the obstetrician. The woman's contact details were obtained at this first encounter, and women were advised that a researcher would contact them three to five months later to participate in a one-to-one interview. Following that period of time, one researcher (TES) contacted all women who had given consent and reminded them of the study. In that brief conversation, the sensitive nature of the topic was again discussed with women. Before starting the interview, women were assured that they could interrupt, postpone or cancel the interview at any moment. During both conversations, verbal consent was obtained to supplement the previously obtained written consent.

7.3.3 Setting and sample

CUMH is one of 19 maternity units in the Republic of Ireland and is situated within the south/southwest hospital group. In 2020, 7040 babies were delivered in the hospital of which 42.2% were born to first-time mothers [560]. Forty-four women consented to be followed up 3 to 5 months postpartum in the current study. One researcher (TES) contacted all women who agreed to participate in the study between 3 to 5 months postpartum by phone or e-mail. Of the 44 women, 4 subsequently declined to participate and 22 were unreachable or did not attend the scheduled online interviews. Eighteen women subsequently participated in the individual semi-structured online interviews between February and July 2021.

In Ireland, even though access to maternity services is free, the option of choosing private or semi-private care also exists. All women participating in this study gave birth in CUMH. Some participants used the service as a public patient, meaning all their antenatal care was provided by CUMH staff; other women used the service as private patients, meaning that their antenatal care was mostly provided by a specific clinician's private practice staff.

7.3.4 Data collection

Women were interviewed about their experiences of behaviour change on modifiable factors during pregnancy and information provision regarding stillbirth and modifiable risk factors for stillbirth during their antenatal care between February and July 2021. Online one-to-one interviews using Google Meet were used as the data collection method due to the restrictions imposed on in-person data collection during the COVID-19 pandemic. The women participating in the study were sent a link to a scheduled online Google Meet at the date and time of their choice, with instructions on how to access the online meeting. The researcher's contact details were also provided to be used in case of technical difficulties.

All online one-to-one interviews were conducted by TES. The semi-structured interviews were guided by a pre-agreed topic guide developed by the multidisciplinary team and informed by previous work of the research group[410,557]. The topic guide included the following areas: history and health habits, awareness of risk factors for stillbirth, feelings and opinions about receiving education on stillbirth, information sources and interventions (See supplementary file 7.2). Some of the questions in the topic guide were designed to elicit information about specific topics such as knowledge about stillbirth and risk factors associated with stillbirth. The use of semi-structured interviews allowed the women to introduce or discuss topics that were not strictly pre-defined in the interview topic guide. As the interviewing process progressed reaching the 14th and 15th interviews, it was clear that the depth and quality of the interviews indicated that it was not necessary to continue actively pursuing the non-responders to the invitation following their initial consent. It was then considered that enough information power was achieved. The concept of information power is proposed by Braun and Clarke as an alternative to the concept of data saturation which they deem incompatible with Reflexive Thematic Analysis. The concept of information power relies on the fact that rich, relevant data requires fewer participants[561,562], hence the more relevant information a sample holds, the fewer participants are needed[562]. However, all 18 women for whom an interview was scheduled were interviewed to respect the commitment that had been made with them.

Interviews lasted between 30 and 50 minutes, were recorded, transcribed verbatim and imported into NVIVO 12 for analysis.

7.3.5 Data analysis

The data analysis conducted in this study is based on the principles of Reflexive Thematic Analysis as described by Braun and Clarke[380,563]. Reflexive Thematic Analysis is a flexible method that is suited to experiential and critical framings of language, data and meaning, and it can be used in either a deductive or inductive way. Reflexive Thematic Analysis involves an interpretative reflexive process, coding does not follow a framework and themes are the final outcome of the analytical process [564]. We conceptualised reflexive thematic analysis for the purposes of this paper within a constructivist approach (See Design Section).

The different phases of thematic analysis described by Braun and Clarke are as follows: 1) data familiarisation and writing familiarisation notes; 2) systematic data coding; 3) generating initial themes from coded and collated data; 4) developing and reviewing themes; 5) refining, defining and naming themes; and 6) writing the report [380]. The analytical process in the current study began by transcribing the audio of the recorded interviews (data familiarisation), to facilitate this process, Tactiq was used during the interviews, which is an automatic transcription tool. One researcher (TES) read and re-read all of the interview transcripts. Inductive open coding was then initiated which facilitated the identification of units of meaning that related to the research aims. Subsequently, those codes were categorised and grouped into themes and re-labelled where appropriate. A record of the evolvement of the themes and the category names was always kept. Further analysis allowed the researchers (TES, KMS) to group the different categories into themes, by refining their meaning to portray the story the data tells. A second author (KMS) reviewed and followed the coding process at all stages, and discussions were held as necessary.

7.3.6 Ethical considerations

The clinical team who obtained consent from women to participate in the study ensured that the sensitive nature of the interview topic was discussed to anticipate potential distressing factors for the women. Women were informed about their

rights to withdraw from the study at any stage without any potential impact on their care or any other type of consequences. We considered the potential for women to experience distress while participating in the interviews and so planned accordingly to provide support to women who experienced distress.

This involved ensuring that the researcher would remain in the online session with the woman after the interview and until she was happy to end the session. The researcher would then make a follow-up contact if deemed necessary or contact the appropriate required support (e.g., referral to specialist bereavement midwives or perinatal mental health services). However, none of the participating women reported distress or were observed to experience distress and so this support was not provided to any participants.

7.3.7 Reflexivity statement

The research team that conducted this study includes experts in health psychology, epidemiology, behavioural science, public health and obstetrics and maternal-fetal medicine. All interviews were conducted by TES, who is a female PhD student with a background in health psychology. TES received training in qualitative methods and interview techniques before conducting this study.

All three remaining authors are highly experienced researchers who have been involved in multiple qualitative studies.

None of the authors had a previous relationship with any of the participants, and the participant only had contact with the researcher conducting the interviews (TES).

The participants were informed that the interviewer was a PhD student and the overall objectives of the PhD project were exposed to them before commencing the interviews.

All members of the research team interacted throughout all phases of this study, which enhanced the process by providing points of view from different disciplines.

7.4 Results

7.4.1 Sample characteristics

The final sample of 18 women included 16 white Irish women, one white Eastern European woman and one white North American woman. Women's ages ranged from 28 to 37 years old. All women were married or cohabiting with their partners (See Table 7.1).

Table 7. 1: Women's characteristics

Women number	Age	Insurance status	Occupation	Relationship status
W1	31-35	Public	Management	Married
W2	25-30	Private	Healthcare	Married
W3	>35	Public	Education	Cohabiting
W4	>35	Private	Social Care	Married
W5	>35	Public	Retail	Married
W6	31-35	Private	Healthcare	Married
W7	31-35	Public	Social Care	Cohabiting
W8	31-35	Public	Management	Married
W9	31-35	Public	Management	Married
W10	31-35	Private	Management	Married
W11	>35	Private	Education	Married
W12	>35	Public	Hospitality	Cohabiting
W13	25-30	Public	Education	Cohabiting
W14	25-30	Public	Healthcare	Cohabiting
W15	31-35	Public	Healthcare	Married
W16	31-35	Public	Healthcare	Married
W17	25-30	Private	Engineering	Married
W18	31-35	Public	Management	Married

7.4.2 Findings

Several themes and subthemes were identified through the analytic process (See Table 7.2).

Table 7. 2: Themes overview

Themes	**Subthemes**
Attitudes towards behaviour change	
Awareness regarding stillbirth and risk factors	Awareness regarding health advice
	Limited awareness about stillbirth
Silence around stillbirth and risks	Lack of discussion regarding stillbirth and risk factors
	Reliance on own information-seeking behaviours
Attitudes towards receiving information about stillbirth	"Knowledge is key"
	Stillbirth perceived as a difficult topic
	Importance of language and preference for information provision

Theme 1. Attitudes toward behaviour change

All women expressed having made changes in their behaviours because of their pregnancies. In some instances, these changes started pre-conceptually in preparation for pregnancy and were maintained throughout the pregnancy. The preconception preparatory behaviours adopted by women involved attending their General Practitioner (GP) for advice, adopting healthier eating behaviours and having a more active life, monitoring menstrual cycles, pre-conceptual alcohol abstinence, pre-conceptual intake of folic acid and antenatal vitamins, as well as pre-pregnancy weight loss. Women spoke about the changes they made as soon as they learnt they were pregnant, the most common behaviours being ceasing alcohol consumption, avoiding foods not recommended during pregnancy, taking vitamins and folic acid, staying active, moderating the intensity of physical activity, having a nutritious diet and/or increasing rest.

Women discussed newly adopting some of these behaviours to achieve their best health status (e.g., being as fit and eating as healthily as possible, taking folic acid), and also abandoning some previous behaviours in order to prevent illness and adverse outcomes (e.g., avoiding consumption of alcohol, avoid stress, quitting smoking, etc). These results show that women perceive behaviour change during pregnancy to have a dual nature in that it can help to improve positive outcomes and reduce the likelihood of adverse outcomes.

> *I just took vitamins and I did go to the doctor, alright? For advice, because I was on the pill for so long and I was a bit worried [W11]*

> *I used to drink, alcohol, at weekends, but I stopped. And I obviously stayed off as well while I was pregnant. [W6]*

> *I was exercising intermittently and I was kind of prioritizing work and I just said 'no I'm going to prioritize myself a little bit more' so I made it my purpose to just do swims and run more often, that was it. [W15]*

Women discussed their different experiences when engaging in behaviour change during pregnancy. All women in this study expressed a strong and clear opinion about the need to stop consuming alcohol during pregnancy. However, some women decided to stop consuming alcohol as soon as they started planning the pregnancy, while others waited until they had confirmation of pregnancy. Regarding smoking and illicit substance use, women also expressed negative attitudes. Only one of the women was a smoker before her pregnancy, and she quit as soon as she learnt about her pregnancy. Physical activity was perceived as beneficial to achieve an appropriate fitness level and general health status, rather than a tool to manage weight gain. Some women expressed difficulties keeping up with their levels of exercise through their pregnancy, which led them to slowly disengage from these behaviours in response to perceived challenges associated with pregnancy (e.g., lack of energy, nausea). On the other hand, the adopted changes in diet were mostly focused on avoiding foodborne diseases, with some women starting to relax their attitude towards their food consumption as the pregnancy progressed.

I think it was less about weight management and more about just fitness, or like wellbeing or health or whatever, more cardio and all that more so than the weight management thing. [W17]

Women had the perception that they had been able to continue with their normal life during their pregnancy with little interference from their pregnancies. Most of the women perceived their behaviour changes (e.g., stopping alcohol consumption, avoiding dangerous foods, engaging in low-demand physical activity) as easy to manage and natural because they were doing these changes for their babies. However, one woman expressed after the interview that actively thinking about all of the changes made during her pregnancy helped her realise that she had made more changes than she had previously perceived. Hence, it seems that the fact that women perceived all their behaviour changes as natural might have contributed to the fact that they were less consciously aware of the range of different changes they did engage in. Women also discussed behaviour change during pregnancy in terms of consequences for the health of their babies or their health which acted as their drive to change their behaviour. As an example, several women spoke about exercise as useful to facilitate labour in general

I thought that the having no drinks would be harder than it was because I'd never been one to be able to go months and months without drinking. But it was actually easier than I thought because I was doing it for my baby. [W 18]

Yes, I had the day sometimes that I really wanted to smoke but, you know, because I was waiting so long for that baby. I was like, 'no, I'm not gonna put her in any risk no matter what'. [W12]

it's all actually just coming back now talking. I'm like, 'no, I didn't really change much'. Yes, I actually did. [W5]

Theme 2. Awareness regarding stillbirth and risk factors

Subtheme 2.1 Awareness regarding health advice

Most women in this study were aware of the importance of maintaining a healthy diet and adequate levels of physical activity during their pregnancies, with little concern about the safety of the physical activity. Women were also aware of the relevance of antenatal vitamins and pre-pregnancy supplements, especially folic acid.

> *I just always knew that if I did want to fall pregnant that I should be on folic acid. I really started ensuring that I took it for the six months beforehand, but I've kind of really been taking it for years sometimes. [W10]*

Risks associated with substance use were also discussed by many of the women. For instance, women were aware of the recommendations regarding alcohol consumption during pregnancy, and all women in this study decided to abstain from alcohol. All of the women also discussed smoking as a behaviour with risks for the baby. Although alcohol consumption and smoking were issues mentioned by almost every woman, some women also commented on the risks associated with illicit drug use.

> *I was at a party and I was said, like, "you know, one drink is okay for the baby" but I was reluctant. [W15]*

> *Um, obviously smoking is a big no-no or any drugs of any description. [W9]*

Some women were also aware of the risks associated with sleep position and were aware of the importance of monitoring their baby's movements, which for one woman was a source of distress.

Subtheme 2.2 Limited awareness about stillbirth

Women were explicitly asked about their knowledge of stillbirth and the risk factors associated with stillbirth. Our findings suggest limited awareness about stillbirth among the women included in this study. Some women discussed an understanding that stillbirth is a pregnancy loss that can occur later during pregnancy. Further, most women openly expressed that their knowledge about stillbirth and related risk

factors was very limited. When explicitly asked, none of the women reported that information about stillbirth was received from a healthcare professional during their antenatal care.

> *No, I think I have a very limited view of it. You know that baby is born and unfortunately, baby is not born alive but I wouldn't say that I know the reasons why are… you know, obviously it was something that crossed my mind […] but no, I can't say that I know a whole deal about it. [W3]*

> *I suppose what I know about stillbirth is when a baby is born and they're dead. I know that there are stillbirths were you can go through full pregnancy and you can give birth and you don't know that your baby's not going to breathe when they come out, and that's just so sad. Then I do know that there's others where a baby may… their hearts might stop or they might stop breathing during pregnancy and you know that and then you have to give birth to the baby, you know? And they're… that's kind of as much as I know really. [W10]*

Additionally, some women confused the term stillbirth with other adverse outcomes given their lack of knowledge of the definitions of pregnancy/infant loss. Women were not aware of the differences between the concepts of stillbirth and miscarriage regarding the gestational cut-off point that differentiates them, and some women conceptualised stillbirth as an event that can only occur during labour. Further, two women confused the term stillbirth with Sudden Infant Death Syndrome.

> *I'm not sure, can it happen at any stage during pregnancy? can the term stillbirth and miscarriage, you know, can they be used in the same? [W8]*

> *I thought it was kind of random…, that it was most risky before kind of 20 weeks was the highest probability [of experiencing stillbirth] or whatever. Um, that's all I really know. [W17]*

> *I don't know a huge amount about it, but it's if a baby unexplainably passes away and usually, when sleeping before they're six months and the child can*

seem to be perfectly healthy and there doesn't seem to be an explanation for it. [W4]

Most of the women in this study expressed that they were “guessing” or “supposing” when asked what their knowledge was about stillbirth and risk factors. Some women stated their knowledge and awareness were drawn from other people’s experiences of pregnancy loss in their social circles (e.g., friends of friends, distant relatives, and neighbours).

The behaviours that women thought were most likely related to stillbirth were substance use, sleeping on their back or doing exercise lying down on their back, or “knocks on the stomach”. Women also spoke about and identified behaviours such as substance use or consumption of certain foods associated with other adverse pregnancy outcomes (e.g., long-term developmental delay, physical disabilities or extreme prematurity); however the link between such risk behaviours and stillbirth was not present in the majority of women’s discourses. These other potential adverse outcomes were perceived as very relevant, concerning, and more present in women’s minds during pregnancy than the possibility of stillbirth.

I think most women are more concerned with development issues with like the drinking and smoking and things like that. They are more thinking.. "okay, when my baby comes out is it going to have learning issues?" No one actually thinks about stillbirth. [W18]

I didn't actually worry about stillbirth, I worried about early miscarriage, I worried about falling like that kind of stillbirth, maybe an early delivery and extreme prematurity and neonatal units. [W6]

Theme 3. Silence around stillbirth and risks

Subtheme 3.1 Lack of discussion regarding stillbirth and risk factors

Many women felt that they had not received information about stillbirth, health habits or risk factors for stillbirth during their antenatal care. When asked, none of the women could remember a moment during their interactions with healthcare professionals (e.g., midwives, consultants) in which they had received information about stillbirths.

> *It is kind of common knowledge, you know, 'don't drink. Don't…' you know, like a lot of it, I think people kind of know already. [W13]*

Regarding specific health habits, such as physical activity, women perceived that healthcare professionals adopted a very conservative approach towards offering advice, and the general message given to women was to "not overdo it". Regarding weight gain, women also expressed a lack of advice in their interactions with healthcare professionals, and most women reported only being weighed at the time of antenatal booking.

> *No, no, not really [got any advice regarding physical activity]. Of course, that's if you did some like, very hard exercise, you have to stop a bit so they warned me about it. [W12]*

On the other hand, some women reported receiving some information regarding health habits and risk factors (e.g., keeping physically active, avoiding substance use, sleeping on the left side or monitoring baby's movements), even though this information was never associated with the risk of stillbirth. In these cases, the information would normally be provided by the women's GP, the hospital midwives or the midwife at their consultant's private clinic.

> *They were both lovely [Consultant and midwife] and so Midwife would have done a lot of the antenatal education when I went in to see her before I went to see Doctor. [W2]*

That's something I came across in my public health nurse, she said everything. And she did say to me, 'sorry, if I'm being kind of condescending or telling you things, you know, but I need to tell you in case you don't know'. [W17]

The information that women discussed being provided by healthcare professionals regarding risk factors and health habits was mostly focused on sleep position, monitoring fetal movements and preventing food-borne diseases. However, even though some women were provided with this information, in most cases women reported that they were not informed about the reasons to adopt these behaviours. Most women received written information during their care, and even though they valued it and used it to prompt discussions with their providers, they did not feel it could replace a conversation with a healthcare professional.

It was just a brief conversation that was had in the earlier stages [about left-side sleeping]. And I don't even know why. [W15]

I was advised that if I felt any reduced fetal movement […] I presumed that was down to... chances of stillbirth or being at risk of something happened in the baby. This was all after 24 weeks and I know that stillbirth is after 24 weeks. So, I presume, I just put that down to being that without ever mentioning this is a risk of stillbirth. Nobody ever mentioned those words to me in that time. [W15]

According to women's accounts, it seemed that the limited information provided by their healthcare providers might relate to the fact that the participant women were healthy and were not experiencing complicated pregnancies.

I don't feel like they felt the need to have that conversation with me because I wasn't part of the criteria for it perhaps. [W15]

I suppose the fact that I was quite young, the fact that I was low risk and I had no other health concerns... It wasn't the thing that they really discussed with me. [..] So we were happy out and I didn't have any signs or symptoms to suggest that there might be something wrong. [W2]

Subtheme 3.2 Reliance on own information-seeking behaviours

Most of the women who participated in our study reported feeling able and confident when trying to find information about their pregnancies using information sources of their choice (e.g., websites, and books). Our findings show that most of the awareness that women showed about stillbirth, risk factors and health habits during pregnancy was a consequence of the women's autonomous information-seeking behaviours. Information-seeking behaviours sometimes translated into decision-making modulating women's behaviour. However, in other instances, finding conflicting information (for example, in online forums) acted as a source of concern.

> *I was happy with that because I knew myself that I got my information from a variety of sources […] I was kind of happy with my own research. [W16]*

> *I was actually looking and I found like different information because there are some pages were saying that you should sleep on the left side, some people on the right side, so I was wondering, should I sleep on the left side or the right side? [W12]*

Proactive information-seeking behaviour led some women to feel that they already had enough information through their research, and they did not require additional professional advice. However, some women acknowledged that only relying on the information provided by their healthcare professionals would have made them feel uninformed.

> *Like it was fine, I'm well able to kind of Google things and research myself. [W9]*

> *I don't think so [that information provision would have been enough without own research]. Because I have a friend who refused to do any research. She was just totally caught off guard. With lots of different things that happened, yeah, I don't think there was enough information there but I mean maybe they would have given me more information if they got the impression that I didn't know, you know. [W16]*

Women also spoke about some of the characteristics of the sources of information that they used during their pregnancies. Women expressed a strong reliance on official sources, like websites or hospital-provided books, and had a critical attitude towards certain sources of information, especially those found online.

> *I did take on board what was in the HSE websites […] I did find the HSE website quite trustful as well. [W2]*

> *Most of the information I just got from the HSE book, I found the HSE book actually to be excellent. That was probably the best, along with the nurses and, and the consultants, that was one of the best sources of information. I just kind of stuck to that because I it's accurate what was in it. [W4]*

Some of the sources of information named by the women were: hospital-provided books, commercial mobile applications, websites, family and friends, conversations with healthcare professionals, social media, online antenatal classes, formal education, peers and podcasts. Most women engaged with commercial mobile phone applications (apps) to obtain information, and expressed that one of the features that they appreciated the most about apps in particular was the weekly updates and notifications; these features provided women with timely relevant information for their stage in pregnancy.

> *[weekly updates] it helped me along the journey. Kind of nearly makes it easier, you're better with something that you can see, now you know you're getting bigger, but like you can kind of just see the journey and it helps you along, but it was just really simple. [W10]*

Theme 4. Attitudes towards receiving information about stillbirth

Subtheme 4.1 "Knowledge is key"

Eight women in our study had a positive and open disposition towards receiving information about risk factors and stillbirth during their antenatal care from their healthcare professionals. For these women, information was perceived as a tool that might have an influence on preventive efforts, and also facilitate the grieving process of parents who experience stillbirth.

> *I think it is good to know about stillbirth because I think it's a very Irish thing maybe, that we don't talk about things that we don't want to talk about. It should be spoken about. I think we should be told about it. We're told about everything else. Then, you know, if there's risk factors for stillbirth and it's also risk factor, we should be made wherever of it. A lot of people don't talk about it. There's a stigma around it, I understand why. It's, you know, it's sad, it's heart-breaking. [W10]*

> *I think to have the knowledge would be way more beneficial than negative. Even if I did go away with a little bit of concern about it, I think that's very natural because you fear for your baby and you want to do everything that you can. But I think overall it would have been extremely positive. [W15]*

> *It's better to be prepared basically for the unexpected. [W5]*

Some comments made by the women indicate that women's concerns regarding pregnancy loss are focused on the first trimester, and that the probability of loss is almost nil afterwards. This is a popular misconception that might have been supported by the women's family or social context.

> *I suppose with stillbirth it's the same kind of thing, preparing women that this is not a 'You've now reached 12 weeks it's not a 100% guarantee' and I suppose if you have no awareness of it and it happens to you would be so shocking. [W18]*

Some women felt that receiving information about stillbirth and its modifiable risk factors would also work to facilitate women's decision-making, by providing them with enough information to facilitate making informed judgements about their risks.

> *I think information is power. Just to give them as much information as possible and then they eventually choose what they want to do with it. But at least they have the information then. [W9]*

Subtheme 4.2 Stillbirth perceived as a difficult topic

Most of the women participating in this study engaged in a process of balancing the pros and cons of receiving this information, which was evident in their discourses. Some of the women in the study expressed that they would have found stillbirth a difficult topic to discuss, especially during pregnancy. However, some women with this opinion also recognised that stillbirth was a latent concern of theirs during their pregnancies.

> *It was something I didn't want to talk about and I wouldn't have brought up myself, but if they had approached me with this, I would have been absolutely fine to talk about it. [W3]*

> *When you're pregnant, it's something you tend to not want to overly research just because […] you're hypersensitive at that time also. [W8]*

Nine of the women expressed that discussing the topic of stillbirth might have the potential to increase their levels of anxiety during pregnancy. The word stillbirth was perceived as a societal taboo which, for some women, acted as a barrier to both information seeking and information provision. However, all of these women felt that the balance between being empowered with the information, despite having certain negative feelings, was still positive. Women rationalised their decision by considering the information as a positive resource, but they highlighted the importance of tailoring the information based on each woman.

> *I think it would have been counterproductive, but you know… saying that I'm a person who likes to know all the facts. So, you know, I would like to think*

that if I did ask that they would have been very forthcoming and very honest with the information that it wouldn't be a case of "oh you don't need to worry about that". [W8]

I think knowledge is key so I would have absorbed the information and taken on board for sure. However, being and discovering that I'm a bit of a worrier, there's no doubt it would have played in my mind. [W15]

I think it would be beneficial, but I'm not sure everyone would want to hear it. Like if you're quite emotional and it is an emotional time, but I do think it is good to kind of maybe touch on it and maybe not put too much emphasis on it. [W9]

One woman had a negative attitude towards receiving information about stillbirth and risk factors, for this woman, the benefits of obtaining the information did not justify the potential harm.

If someone just says 'don't drink alcohol' fine. You don't do it. If someone says 'don't drink alcohol because you might have a miscarriage' and then you're worrying about it… Does worrying increase the risk? I don't know. No, it's probably better that it's not pointed out maybe, for fear that you do worry more about it and you worry unnecessarily. [W17]

Subtheme 4.3 Importance of language and preferences for information provision

Some of the women that participated in our study insisted on the importance of the language used when providing information about stillbirth and risk factors. Women felt that the best way to provide education about stillbirth and risk factors was to do so in a very sensitive way and avoid the use of 'blaming' vocabulary when talking about the behaviours that might increase the risk of stillbirth.

So while it might invoke some degree of upset. I think if it's done in a sensitive way and in a kind of a positive slant of, you know, "we know there are some things that we can do to reduce this risk", that I would think that most women would be receptive to that. [W6]

Maybe do it without using the word stillbirth: 'avoid these behaviours during pregnancy' or something. Yeah. [W13]

Maybe it's just the people don't like to say the words, you know? [W15]

Regarding women's preferences for information provision, women highlighted the importance of having reliable sources, and providing specific advice or information, with the option for more information on demand. Women also expressed a preference for group educational sessions on behavioural risk factors for stillbirth and best health habits for pregnancy. According to these women's opinions, receiving information about stillbirth and risk factors for stillbirth on a one-to-one basis would have made them feel as if there was something wrong with their pregnancy, whereas receiving the information in a group format would be considered as "just another topic to discuss".

I think maybe groups, because I think if it's one on one, the person might feel it's being directed at them. Whereas if it's in a group, it's just, you're informing them. [W5]

When it comes to the best timing to provide women with information about stillbirth and risk factors, women felt that this information should be provided early in pregnancy. These women were aware of the importance of not engaging in risky behaviours as soon as possible, and so they considered that receiving the information early would help prevent adverse outcomes in pregnancy. Some women expressed that they had a heightened cautious attitude at the beginning of their pregnancies and that this could be taken advantage of in terms of increasing awareness about risk behaviours. Additionally, some women expressed the potential for conversations about stillbirth between women and healthcare providers to become more difficult as the pregnancy progresses, when women's concerns start focusing on labour and birth.

There's a lot of information at the start, but it's really that's when you need to be doing it [changing behaviour], […] I think giving the information earlier is better. [W15]

I think earlier in the pregnancy so they have time to set up for a healthier pregnancy. [W14]

However, other women considered that the information about stillbirth and risk factors should be provided in the second trimester since the first trimester represents a vulnerable period, in which discussion of stillbirth would constitute additional stress and pressure for the woman. Whereas other women, especially those with a background in healthcare, highlighted the importance of the pre-conceptual period as the best time to provide education to women.

Maybe in between, 12 weeks and the anomaly can. I think a lot of people are kind of stressed about getting to the 12 weeks anyway […] I think maybe we should leave people get to that stage rather than stressing them out even more. [W1]

At the beginning I think there's enough stress anyway … after the half, if you know that everything is fine, that kind of information won't actually scare you so much like at the beginning. [W12]

7.5 Discussion

Our findings have shown that information provision and awareness about stillbirth and associated behavioural risk factors are mostly poor amongst most women with uncomplicated pregnancies and births participating in our study. Women showed good awareness regarding health recommendations and behaviours to avoid during pregnancy, however, regardless of the level of knowledge that women expressed about stillbirth or potential behaviours associated with an increased risk of stillbirth, it seemed that there was not a clear link establishing the association between risk behaviours and stillbirth in women's discourses. Further, the awareness that women expressed regarding risk factors and stillbirth was predominantly a result of their information-seeking behaviour. Women expressed that information provision during their antenatal care regarding health behaviours was poor, and information regarding stillbirth was non-existent. Women in this study showed varying attitudes regarding being provided with information about stillbirth and behavioural risk

factors for stillbirth. Whereas some women perceived information as a tool to improve prevention efforts, other women considered that this information was not necessary in all cases and it would only increase anxiety levels for them.

Women's reasons for engaging in behaviour change were commonly associated with obtaining the best possible outcomes for their babies and themselves. This motivation to make healthy choices and a sense of responsibility driven by the desire to improve the baby's health and reduce risk has been described in the extant literature[262,410,556,557]. Further, women's perception of risk, which can only be accurate with appropriate communication and awareness, will also influence their decision-making[565]. This highlights the importance of providing accurate information to women about stillbirth related to different behaviours during pregnancy. Providing women with this information might increase their level of motivation to tackle modifiable behaviours and reduce their risk of stillbirth.

Women in our sample were able and willing to conduct their research and find answers to their concerns, and one of the most mentioned information resources was the internet. However, we know from previous research that the information available online regarding stillbirth and behavioural risk factors for stillbirth in Ireland and the UK is scarce and difficult to access[566]. Further, women in our study were a small number of predominantly highly educated white Irish women. Previous research has demonstrated that sociodemographic characteristics are associated with the choice of a particular source of information, and also with the number of sources used [567], with being female, educated and young the best predictors to engage in health information-seeking behaviours [567]. Therefore, it is possible that the positive attitude towards information-seeking in our sample is not representative of the general population, and further research would be necessary to explore the needs of people from different sociodemographic backgrounds.

Previous research has demonstrated that the levels of knowledge and understanding that women have about advice received during pregnancy will have an impact on their psychological capabilities towards behaviour change[410,557]. Women who are less aware, or who held misconceptions about the consequences of their behaviours will

be less likely to engage in behaviour change [299,410]. The women in our sample had limited knowledge about stillbirth and its associated risk factors; however, when prompted to speak about the advice received during their antenatal care, monitoring of fetal movements and the importance of sleep position was often mentioned. These findings are very similar to those obtained by Stacey et al. in a recently published study, where they explored migrant women's awareness of health messages to reduce stillbirth risk[568]. Our findings also highlight the importance of promoting health education during pregnancy from the healthcare professionals involved in women's care, as well as the value of generating additional written or online resources for women.

Additionally, our results show that women prefer antenatal groups to receive information regarding their health and their pregnancies, and most women found that antenatal classes represent appropriate spaces to share information about stillbirth and risk factors for stillbirth. This is positive considering that previous research has demonstrated that passive transfer of information is not sufficient to prepare women and their partners for birth and parenthood, and hence facilitating educational groups using different types of experiential methods should be recommended [569]. Women in our study were also very reliant on the written information provided to them by their healthcare providers. Hence, health services should produce up-to-date user-friendly online and paper-based resources or materials for women and encourage engagement with such materials from the healthcare professionals' perspective to support discussion with patients. However, given the diversity in which women prefer to receive information, it is important to tailor and utilise diverse types of intervention approaches to maximise engagement and effectiveness of antenatal interventions.

Healthcare professionals should engage in active discussions with women, regarding risk factors for stillbirth. Antenatal education standards and healthcare professionals' training programmes should guide to support healthcare professionals in promoting health with their patients, and also be able to discuss risk factors for stillbirth or other potential adverse outcomes. The National Women and Infant Health Programme developed the National Standards for Antenatal Education in Ireland in 2020[569].

Although this document highlights the importance of including an element of better health and wellbeing (theme 4) into the optimal standards of antenatal education, this theme only refers to supporting healthy habits and addressing mental health concerns generally, without specifying the need to address issues such as the risk factors for stillbirth with women[569].

Previous research has also demonstrated that stillbirth is a taboo topic in society [27,570,571]. However, as seen from our results, most women would welcome information about stillbirth and risk factors as a tool to improve their health and prevent it from happening to them, or even as emotional preparation after experiencing a stillbirth. However, a high number of women also acknowledged that receiving this information would have the potential to increase their levels of anxiety and concerns, which is an issue that should be taken into consideration. Literature in areas such as sexual health or weight management has shown that healthcare professionals are reluctant to discuss topics perceived as taboo or difficult with their patients[572], and stillbirth is presumably a similarly unmentionable topic[570]. We believe that it is important that official bodies regulating antenatal education standards in different high-income countries include specific guidance regarding advising women about health habits, risk factors, and potential adverse outcomes such as stillbirth to support healthcare professionals in providing antenatal education with confidence. Furthermore, as mentioned by multiple women, receiving this information has the potential to increase their anxiety levels or their concerns, and hence, healthcare professionals must be prepared to address women's concerns and resolve their doubts, trying to alleviate their worries by providing accurate and sensitive information. The different types of pregnancy loss have different implications regarding care, and hence, breaking the silence around stillbirth is essential to ensure that women who experience stillbirth received the appropriate support[27].

Our study has some strengths and limitations. The study's first limitation is related to the sample of interviewed women. The study participants were predominantly white, educated, heterosexual women from developed countries, and, likely, their experiences with behaviour change during pregnancy and challenges identified are

not generalisable to people from different sociodemographic backgrounds. Additionally, the rates of smoking and alcohol consumption were also lower in our sample than in the general pregnant population, thus findings related to this modifiable risk factor for stillbirth are likely influenced by this. Further, another limitation that we encountered when conducting the interviews is that in some instances women struggled to recall some parts of their experiences, given the timeframe in which they were interviewed. This might also be explained by the changes in memory that occur during pregnancy and have been described in the literature before, where women reported memory deficits categorised as a general sense of "fogginess"[558]. The number of women approached to be consented to this study was high for a qualitative design. This decision was made to ensure that we were able to obtain an appropriate amount of rich data and anticipate that most women might decline to participate a few months after leaving the hospital. Of the 44 women recruited, only 18 participated in the interviews, indicating that possibly our timeframes did not suit the majority of women and highlighting the challenges of recruiting women during the busy postnatal period. Furthermore, given that only the clinical team has access to the patient's medical record, we are unaware of how many women were approached by the clinical recruiting team and declined to participate as this number was not recorded. The researchers only obtained the contact information of the women who consented to participate in the study. This hinders the assessment of the efficacy of our recruitment method, and it also limits our knowledge about potential differences between women who agreed to participate and women who declined. It is possible that women who chose to take the time to participate in this study were adapting better to the postpartum period than those who could not participate. This might be related to their support networks, their baby's sleep patterns or temperament, their baby's health needs or their own health needs. Hence, it is important to acknowledge that women participating in this study might have been having a more positive experience regarding their pregnancy, birth and the postpartum period, and so these factors also need to be considered when trying to generalise these findings.

Despite these limitations, our study also has several strengths. Our inclusion and exclusion criteria were designed to recruit women with uncomplicated pregnancies and births, enabling the exploration of experiences of women receiving the most "generalised" type of care in our maternity services; this increases the generalisability and transferability of our findings to other low risk/uncomplicated pregnancies in similar maternity systems. Furthermore, the use of online interviews was praised by some of the women as a very comfortable way to engage in research, without causing much interference in their normal lives. To date, there is only one other study exploring women's perception of public health messages to reduce stillbirth, in this case, exploring the views of migrant women in the UK [568]. The findings of this study are similar to our findings in that they highlight the complexities of discussing stillbirth with women during their pregnancies and the importance of developing culturally appropriate resources to secure efficient communication.

7.6 Conclusion

This study aimed to explore women's experiences of modifiable risk factors during pregnancy, awareness of stillbirth and its risk factors, as well as their experience with information provision during their antenatal care. The findings of this study have shown that women with uncomplicated pregnancies receive very poor information about health behaviours, behavioural risk factors or stillbirth during pregnancy. Women have high levels of understanding of how to have a healthy pregnancy, but the link between behavioural risk factors and potential outcomes such as stillbirth is not considered. Information provision during antenatal care was not sufficient, and women had to rely upon their information-seeking behaviour. Most women perceived receiving information about stillbirth during antenatal care to be useful to help preventive efforts, although others acknowledge the potential for this information to raise some concerns highlighting the importance of using sensitive non-judgemental language.

Information provision alone is not sufficient to support behaviour change, however, it might act as a first step to engage in discussions and facilitate women seeking adequate care for their specific needs. Healthcare professionals should break the

silence around stillbirth and incorporate risk factors, health habits and stillbirth in their routine discussions with women, especially in terms of outcomes for their babies, to motivate women to engage in behaviour change. Tackling the modifiable maternal risk factors for stillbirth by providing information and supporting women with behaviour change during pregnancy might contribute to reducing the stillbirth rates in Ireland.

7.7 Inter-chapter synopses

Chapter 7 aimed to explore the experiences and knowledge of first-time mothers about behavioral risk factors for stillbirth. The study found that the participating women had limited knowledge about the behavioral risk factors associated with stillbirth. The women also reported receiving little information about these risk factors from healthcare professionals. However, when asked about their attitudes to receive this information during antenatal care, most of the participating women were able to acknowledge its value to enhance preventive efforts. The study highlights the need for healthcare professionals to provide more in formation and support to first-time mothers regarding behavioral risk factors for stillbirth to reduce the incidence of stillbirth. Chapters 2 to 7 add to the growing body of evidence that highlights the need for better education and support for pregnant women to address their modifiable risk factors and potentially reduce the incidence of stillbirth, and also provide in depth analysis of the influences affecting such behaviours. Chapter 8 aimed to identify which interventions had been produced to date to address the behavioural risk factors for stillbirth and what behaviour change techniques had been used with that purpose. Before conducting Chapter 8, it was well known that several behavioura change interventions had been designed to address some of the behaviours associated with stillbirth individually, and even some of them addressed more than one behaviour at a time, but no other review had analysed the interventions in detail to identify the behaviour change techniques composing them.

Chapter 8 – A systematic review of behaviour change techniques used in the context of stillbirth prevention

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Chapter 8 – A systematic review of behaviour change techniques used in the context of stillbirth prevention

8.1 Abstract

Background

Stillbirth is one of the most devastating pregnancy outcomes that families can experience. Previous research has associated a wide range of risk factors with stillbirth, including maternal behaviours such as substance use, sleep position and attendance and engagement with antenatal care. Hence, some preventive efforts have been focused on tackling the behavioural risk factors for stillbirth. This study aimed to identify the Behaviour Change Techniques (BCTs) used in behaviour change interventions tacking behavioural risk factors for stillbirth such as substance use, sleep position, unattendance to antenatal care and weight management.

Study design

A systematic review of the literature was conducted in June 2021 and updated in November 2022 in five databases: CINHAL, Psyhinfo, SociIndex, PubMed and Web of Science. Studies published in high-income countries describing interventions designed in the context of stillbirth prevention, reporting stillbirth rates and changes in behaviour were eligible for inclusion. BCTs were identified using the Behaviour Change Technique Taxonomy v1.

Results

Nine interventions were included in this review identified in 16 different publications. Of these, 4 interventions focused on more than one behaviour (smoking, monitoring fetal movements, sleep position, care-seeking behaviours), one focused on smoking, three on monitoring fetal movements and one on sleep position. Twenty-seven BCTs were identified across all interventions. The most commonly used was “Information about health consequences” (n=7/9) followed by “Adding objects to the environment” (n=6/9). One of the interventions included in this review has not been assessed for efficacy yet, of the remaining eight, three showed results in the

reduction of stillbirth rates. and four interventions produced behaviour change (smoking reductions, increased knowledge, reduced supine sleeping time).

Conclusions

Our findings suggest that interventions designed to date have limited effects on the rates of stillbirth and utilise a limited number of BCTs which are mostly focused on information provision. Further research is necessary to design evidence base behaviour change interventions with a greater focus to tackle all the other factors influencing behaviour change during pregnancy (e.g.: social influence, environmental barriers).

8.2 Introduction

Stillbirth is a devastating outcome for expectant parents. Stillbirth definitions vary across the world, with some countries using different weight or gestational thresholds, ranging from 350 grams to 1000 grams and from 20 weeks to 28 weeks [12] which leads to discrepancies when comparing prevalence rates. However, in 2015, the worldwide estimates for stillbirth at 28 weeks gestations were 3.5 per 1,000 total births when looking at 49 different high-income countries, with the minimum rate being 1.3 in Iceland and the maximum being 8.8 in the Ukraine [573]. Stillbirth rates have remained steady over many years in high-income countries and even though most stillbirths in high-income countries occur before labour and are associated with placental pathology [574] some others are associated with modifiable risk factors [575].

Ota et al. (2020) conducted a systematic review of 43 Cochrane reviews assessing 61 different stillbirth prevention interventions across low-, middle and high-income countries [576]. The authors concluded that few reviews were able to provide clear evidence, as the settings of the different interventions were too different. Ota et all's review differs from the current review in that it focused on a wide range of interventions (i.e., nutritional interventions, prevention and management of infection, prevention and management of maternal morbidities, screening, and management of fetal growth), however, our review is focused on those interventions conducted in the context of stillbirth prevention with behavioural focus. Stillbirth has been associated with a wide range of different risk factors in the literature, and these include behavioural risk factors. These risk factors include smoking [47,134,298], drinking alcohol [144,310], illicit drug use [87,194,315,341], lack of attendance or engagement with antenatal care [206,577], sleep position [578] and behaviours associated with increased maternal weight [188]. Given that these factors have the potential to be modified, there have been previous efforts to develop and implement behaviour change interventions to address them and attempt to reduce the risk of stillbirth.

Being able to identify the different components of the interventions designed and examined to date in the context of stillbirth prevention is essential. This kind of work allows to build cumulative evidence that can be used by intervention designers to plan effective and replicable behaviour change interventions using systematic

methodologies like, for example, the Behaviour Change Wheel (BCW). The BCW is a systematic methodology to develop behaviour change interventions, which includes the identification of relevant BCTs. Identifying the different components of each intervention, or behaviour change techniques (BCTs) will facilitate this process by providing detailed information about the different components of the intervention, especially given the variability across definitions of stillbirth used, outcomes measured (e.g.: different obstetric outcomes such as aggregated perinatal death vs. specific definition for stillbirth; different behavioural outcomes such as reduction in the number of cigarettes smoked, weight loss, number of baby's movements, etc.) and the different intervention approaches.

A BCT is an observable, replicable and irreducible component of an intervention, an "active ingredient", designed to alter or redirect causal processes that regulate behaviour [302]. Identification of BCTs in interventions allows for accurate replication of interventions and faithful implementation of interventions demonstrating effectiveness [302]. Taxonomies and ontologies can be used to deconstruct intervention components. The BCT Taxonomy V.1 (BCTT.V1) is a commonly used BCT classification and definition system to identify BCTs [579–581]. To our knowledge, there has not been any systematic review focused on behaviour change interventions tackling the behavioural risk factors for stillbirth that were designed to reduce stillbirth rates. Hence, the current review can be valuable to identify which techniques have been used in the past to inform the development of future theoretically informed interventions.

Hence, this systematic review aimed to identify all the Behaviour Change Techniques (BCTs) that have been used to date in behaviour change interventions designed to reduce the rates of stillbirth by addressing modifiable behavioural risk factors such as substance use, attendance at antenatal care, weight management behaviours or maternal monitoring of fetal movements.

8.3 Methods

This systematic review has been informed by the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) guidance for reporting systematic reviews[582]. The protocol for this review was submitted to the PROSPERO, Registration code (ID CRD42021264914) and the protocol has been published[583] (See supplementary file 8.1); hence, the methods will be reported in brief here.

8.4 Eligibility criteria

Exclusion and inclusion criteria were developed using the PICO Framework (See Table 8.1).

Inclusion criteria included (1) interventions with a behavioural focus that included (2) pregnant women or women up to 12 months postpartum, that were conducted in (3) high-income countries, and (4) reported changes in rates of stillbirth after the intervention. Case-control studies, randomised control trials, cross-sectional studies, and quasi-experimental studies were eligible for inclusion. No limiters were used in the search.

While we acknowledge that there is an acute need for interventions in low and middle-income countries, we limited our study to high-income countries due to the differences between care systems and challenges associated with improving pregnancy outcomes amongst low-, middle- and high-income countries. Furthermore, limiting this study to high-income countries might facilitate the use of its findings to develop behaviour change strategies relevant and applicable to such contexts.Interventions with a behavioural focus, or behaviour change interventions, are defined here as those interventions targeting behaviours that have been previously associated with an increased risk of stillbirth including substance use cessation interventions, alternative packages of antenatal care to increase engagement and weight management interventions.

Exclusion criteria included interventions that (1) targeted behavioural risk factors for stillbirth but that were not designed to reduce stillbirth rates or that did not include stillbirth in their measured outcomes and (2) interventions conducted in middle- or low-income countries.

Table 8. 1: PICO Framework

PICO framework	Eligibility criteria
Population or problem	Population: Pregnant women Setting: High-income countries
Intervention or exposure	Any intervention designed to prevent stillbirth that includes a focus on behavioural risk factors.
Comparison	Participants who were not exposed to an intervention, who receive 'standard care' or who acted as an active comparator group.
Outcome	Behaviour change techniques used in the interventions, classified using the Behaviour Change Technique Taxonomy V1[302] Changes in rates of stillbirth after the intervention, no specific definition for stillbirth will be used as they differ internationally.

8.5 Systematic literature search

A systematic search was conducted using CINAHL Complete, SocIndex, Web of Science, PubMed, PsycINFO and Open Grey in September 2021 and updated in November 2022. Keyword searches were used across four different concepts (1) Stillbirth, (2) Intervention, (3) Pregnancy and (4) Study design. The search strategy was developed for PubMed (See supplementary file 8.2) and was adapted for each relevant database.

Two individual reviewers (TES, KMS) screened titles, abstracts and full articles using Rayyan against the inclusion and exclusion criteria to identify relevant articles. A manual check of the reference lists of included articles was also conducted by TES. When the search identified systematic reviews relevant to the topic, these were then manually reviewed for potentially relevant articles. Following the identification of relevant articles, all supplementary materials (e.g., intervention leaflets, trial registrations and/or protocols) or other relevant articles associated with the identified interventions were manually searched to further inform data extraction or quality appraisals.

8.6 Data extraction

8.4.1 Study Characteristics

A standardised data extraction sheet was used to extract data by one reviewer (TES), with extraction reviewed for accuracy by a second reviewer (KSM).

Data extracted included: (1) General information: Authors, title, name of intervention/ approach (if applicable), year of publication, country of origin; (2) Study characteristics: Study aims/objectives, type of study/report, stillbirth definition used, recruitment strategy; (3) Participant details: sample size, number of people in the control arm (if applicable), number of people in the intervention arm (if applicable), mean age, age range, socio-economic status, education level, ethnicity, pregnancy status, relationship status; (4) Intervention details: setting, investigated topic/ target behaviour, description of intervention/approach and control treatments, delivery mode, intervention duration, current stage of implementation, outcomes measured in the intervention (e.g. reduction stillbirth rates, increased awareness, behaviour change), the effectiveness of the intervention.

8.4.2 Quality assessments

Three different quality assessment tools were used depending on the type of study or report identified in the search. The Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria[584] was used to assess randomised controlled trials, the Joanna Briggs Institute Critical Appraisal checklist was used for analytical cross-sectional studies[585], and the ACCORDS checklist for Grey Literature[586] was used to assess reports and descriptive studies that were not suitable to be appraised with the previously stated tools. This approach to quality appraisal differs from the proposed approach in the study protocol[583] and represents a comprehensive approach to ensuring that all study types are evaluated by the most suitable appraisal tool.

To assess the overall quality of the studies, a subjective judgement of the overall quality of each study was made based on the evaluation of each item of the relevant tools.

8.4.3 Behaviour change technique coding

Intervention content was coded using the BCTTv1 Taxonomy[302] and the presence or absence of individual BCTs was recorded in an excel sheet. Intervention content was coded using all the relevant documents for each intervention (e.g., journal articles, protocols, leaflets, reports) One author (TES) conducted the coding of the BCTs providing evidence for each one by selecting the fragments of the text supporting the coding of each BCT. A second author (KMS) reviewed the coded materials and the identified BCTs. Any discrepancies were discussed until an agreement was reached.

8.4.4 Intervention effects

Intervention effects are presented in table format and summarised in the body of the text for stillbirth outcomes when reported in the papers. A meta-analytic approach was not used for intervention effectiveness given the heterogeneity in the stillbirth definition used and in the behavioural outcomes measured. Given that the interventions focus on different behaviours, and that there is only a small number of trials included in the synthesis, trying to group the interventions into categories for inclusion in a meta-analysis would not result in robust, reliable data on intervention effects[587]. This heterogeneity in definitions and outcomes is also what impedes linking individual BCTs to effectiveness.

8.7 Results

8.7.1 Search outcome

The database search identified 8874 studies in September 2021 and 312 studies in November 2022. Of these, 8253 remained after duplicate removal. After screening titles and abstracts, 38 studies remained potentially eligible for full-text review. Sixty-six potentially relevant systematic reviews were also identified when conducting the title screening, so they were manually reviewed to identify potentially relevant interventions. The result of both screening processes left 55 studies eligible for full-text screening. From these, 16 articles, representing 9 interventions, met eligibility criteria and were included in the synthesis (See Figure 1). From the 16 studies included in the synthesis, additional study protocols, trial registrations,

supplementary materials, reports, and brochures were identified when available and included in the evidence synthesis. Further, one relevant study relating to one of the included interventions published in early 2023 was identified after the last search and was included in the review for additional information. As such, 31 documents were included in the analysis representing 9 different interventions (Supplementary file 8.3).

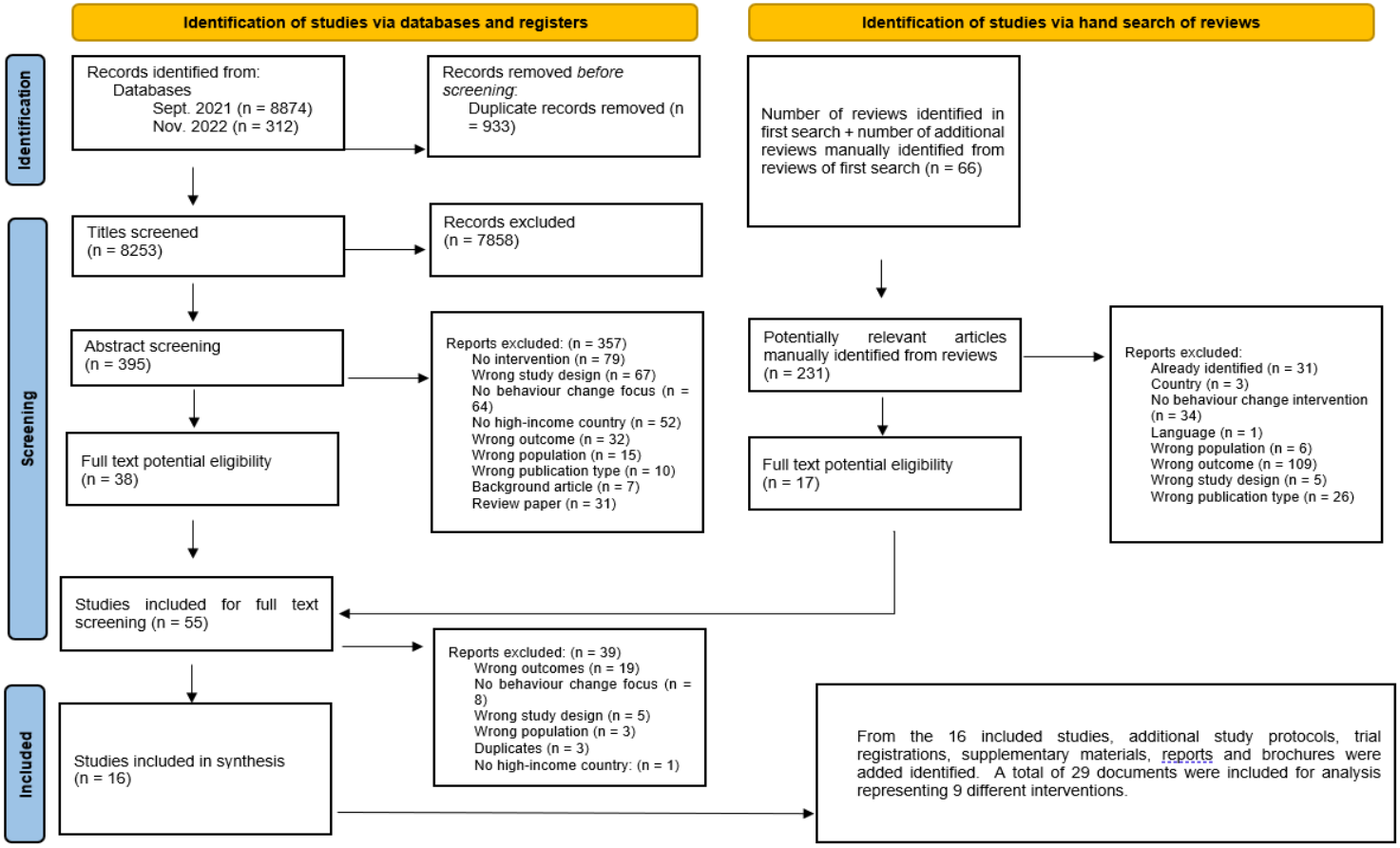


Figure 8. 1: PRISMA 2020 flow diagram for new systematic reviews which included searches of databases, registers and other sources From: Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. BMJ 2021;372:n71. doi: 10.1136/bmj.n71. For more information, visit: http://www.prisma-statement.org/

8.7.2 Study characteristics and quality assessments

Study characteristics are presented in Table 8.2. For information on the number of articles and/or additional documents informing Table 8.2 please Refer to Supplementary file 8.3. Studies were published between 1985 and 2023. Two interventions were conducted in Australia[241,588], one in USA[589], three in the UK[589,590] and one in Norway[591]. One intervention was conducted in Australia and New Zealand[592], another was conducted in Australia, Canada and USA[271,593]. Of the included studies, 8 were randomised controlled trials (RCTs)[271,590–594], two were non-randomised control trials[592], one was a cross-sectional study[595], one was a prospective cohort study[596] and two were pre-post intervention measurements[589,597]. Regarding the behavioural focus of the intervention, one intervention was designed to address smoking[598], three interventions were designed to focus on monitoring reduced fetal movements[591,595,596,598,599], and one intervention was designed to target women's sleep position[271,593]. The four remaining interventions were designed to address multiple aspects[237,238,241,242,589,594,597,598,600], for example, smoking[237,242,594], tobacco exposure[594], depression[594], intimate partner violence[594], medical management of growth restriction[237,242], monitoring of decreased fetal movements[237,242,589], maternal sleep position[242,589] attendance at antenatal care[589], and timing of birth[237,242] (See Table 8.2 for further detail).

Quality appraisals indicated that the overall quality of the included studies was moderate to good, with some methodological limitations. The most common issues in RCTs were related to the blinding of participants and personnel, and the blinding of outcomes. Cross-sectional studies had better quality overall, with the most common issues related to the identification of confounding factors. The two reports[237,242] assessed with the ACCODS criteria[601] presented no methodological limitations (See supplementary file 8.4).

Table 8. 2: Intervention Characteristics.

Author, year and country	Aim and target behaviour	Stillbirth definition used	Pregnancy status of participants	Design	Intervention	Control	Results	Qualitative assessment and risk of bias
Intervention 1. The Safer Baby Bundle[241,242]								
Andrews et al. (2020), Australia.	To compare stillbirth rates at 28 weeks gestation or more across Australian maternity settings. **Behavioural target:** Smoking, monitoring of decreased fetal movements, sleep position. **Other elements:** Timing of delivery and education about fetal growth restriction.	Stillbirth at 28 weeks or more in singleton pregnancies without lethal fetal congenital abnormalities.	Currently recruiting.	Non-randomised trial.	Currently recruiting. The SBB contains five elements: Element 1: Supporting women to stop smoking in pregnancy. Element 2: Improving detection and management of fetal growth restriction Element 3: Raising awareness and improving care for women with decreased fetal movements Element 4: Improving awareness of maternal safe going-to-sleep position in late pregnancy Element 5: Improving decision-making about the timing of birth for women with risk factors for stillbirth	NA. Pre-post intervention measures.	Most jurisdictions in Australia are currently in the implementation phase. Victoria jurisdiction showed a decrease the stillbirth rate by 21%, from an average rate of 0.24% to 0.19% and an increased the days between stillbirths by 131%, from an average of 3.5 days to 8.1 days	JBI Checklist for cross-sectional studies. Moderate quality. ACCODS. High-quality.

Intervention 2: Intervention to Reduce Behavioral and Psychosocial Risks[594,598]								
Subramanian et al. (2011), USA	To address the impact of an intervention on pregnancy and neonatal outcomes and psychosocial risk factors. **Behavioural target:** Smoking, environmental tobacco smoke exposure, depression and intimate partner violence	Perinatal death, no explicit definition	Less than 29 weeks gestation.	RCT	n = 510. Elements from the successful Smoking Cessation or Reduction in Pregnancy Program Treatment (SCRIPT) trial, the transtheoretical model of behaviour change, and the ‘‘pathways to change’’ self-help manual were incorporated. Intervention sessions were provided at each routine prenatal care visit.	n = 515. Usual care group.	Behavioural and psychosocial risks were reduced by the intervention, 58.0% of women assigned to the intervention group resolved some or all of their risks, whereas only 48.2% of those receiving usual care did so (unadjusted P=.019). No variation was detected on their impact on pregnancy and neonatal outcomes.	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. Moderate quality.
Intervention 3: Keeping baby SAFE[589]								
Warland et al., (2011), Australia	To determine women’s baseline knowledge of stillbirth incidence and to identify if giving them an information	No explicit definition	Women in their final trimester of pregnancy	Pre-post intervention study	n = 22. Women were provided with a stillbirth information leaflet.	NA. Pre-post intervention measures.	85.7% of women were aware of the definition and prevalence of stillbirth after the intervention (p-value <0.001), as well as improved understanding of	JBI Checklist for cross-sectional studies. Moderate quality.

	brochure about stillbirth would improve awareness. **Behavioural target:** Awareness about stillbirth and modifiable risk factors including sleep position, attendance and compliance with antenatal care appointments, monitoring baby's movements, and seeking care early.					risk factors associated with stillbirth. No pregnancy outcomes reported.	
Intervention 4: Saving Babies' Lives Care Bundle[237,238,597,600]							
Widdows et al., Tommy's (2018 – 2021), UK	The overall purpose of the study is to determine the impact of the care bundle on UK maternity services and perinatal outcomes.	The death of a baby before or during birth after 24 weeks of gestation expressed as a proportion of live births	Pre-post intervention study	Number of participants varies depending on element of the care bundle and NHS Trust. Smoking monitoring cessation: CO testing for all pregnant women at booking appointment, referral of all women identified as smokers to smoking	NA. Pre-post intervention measures.	Stillbirth rates fell in the participating maternity units by 20% from 4.2 to 3.4 per 1,000 livebirths. This rate of reduction was greater than seen across the whole of England	JBI Checklist for cross-sectional studies. Widdows et al. (2018a). High-quality.

	Behavioural target: Smoking monitoring cessation, Monitoring fetal growth, reduced fetal movements and effective fetal monitoring in labour. Second version of care bundle also includes reducing preterm birth.				cessation services on an opt out basis / Reduce fetal movements: information and advice leaflet on RM to be provided to all women by 24 weeks pregnancy, reduced fetal movements discussed at every antenatal care appointment, use of the provided checklist to manage care of women who report RFM.		over the same period	Widdows et al. (2021). High-quality. Widdows et al. (2018b). Moderate-quality.
Intervention 5: Financial incentives for smoking cessation[590]								
Tappin et al., (2015), UK	To assess acceptability and begin to explore efficacy and cost effectiveness of up to £400 of shopping vouchers added to routine specialist pregnancy NHS "Stop Smoking Services" to help pregnant smokers quit.	Stillbirth is a non-registered outcome. No explicit definition	Mean gestation age at booking 12.62 weeks control group, 12.27 weeks incentives group	RCT	n = 306. The intervention involves offering up to £400 of shopping vouchers to participants if they engaged with services and subsequently quit smoking. Intervention participants received £50 of vouchers if they attended their face-to-face appointment and set a quit date. Confirmed quitters were sent a further £50 voucher. Twelve weeks after stopping smoking,	n = 306. The control group was offered routine specialist pregnancy support by the stop smoking services.	The number of smokers that stopped smoking was significantly higher in the incentives group (22.5% vs. 8.6%). No differences were seen between groups of neonatal outcome or stillbirth rates.	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. High-quality.

	Behavioural target: Smoking				women in the incentives group who were quitters were contacted and, if confirmed to be abstinent, were sent a £100 voucher. Women in the incentives group who were confirmed as abstinent between 34-38 weeks gestation by the carbon monoxide breath test were sent a final £200 voucher.			
Intervention 6: My baby's movements[590]								
Flenady et al. (2019), Australia and New Zealand Flenady et al. (2022), Australia, New Zealand	To evaluate the effectiveness of a reduced fetal movements awareness package and amobile phone application for women combined with an educational program for clinicians. **Behavioural target:** Monitoring fetal movements	Stillbirth at 28 weeks or more gestation	From 28 weeks gestation until birth.	Cluster RCT.	The intervention consisted of the provision of the "My baby's movements" education package including awareness materials for antenatal clinics. Further, it included a mobile phone program consisting on a phone app or short message service. The app sends an alert to prompt woman's awareness of her baby's movements at a time and frequency of her choice. The app	Control period: women will receive standard care which includes provision of the bi-national brochure to women and receiving information about what constitutes normal FM and advice in case of concerns.	The stillbirth rate was lower during the intervention period although no significantly (2.2/1000 versus 2.4/1000 births; aOR 1.18, 95% CI 0.93–1.50; P = 0.18). App users had higher rates of induction of labour (40.5% vs 34.3%) (aOR 1.10; CI 1.06– 1.14; P < 0.01).	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. Moderate quality.

					also provides with a "movement counter". The alternative SMS program sends a series of messages to the woman on a weekly basis after 28 weeks gestation. Women are prompt to seek care if concerned.			
Flenady et al. (2023), Australia, New Zealand	To evaluate the effectiveness of the My Baby's Movements app.	Stillbirth at 28 weeks or more gestation	Women with a singleton pregnancy without major fetal anomaly at ≥28 weeks gestation were	Cluster RCT	The My Baby's Movements app provides information to raise awareness about fetal movements during pregnancy and provides as described above.	As above	Stillbirth rates were not statistically lower in app users (1.67/1000 vs 2.29/1000) (aOR 0.79; 95% CI 0.51–1.23; P = 0.30).	The Cochrane Effective Practice and Organisation of Care
Intervention 7: Count the kicks[591,595,596]								
Saastad et al., (2011), Norway	To assess the effects of increased awareness towards fetal activity on antenatal identification of fetal pathology. **Behavioural target:**	Perinatal death, no explicit definition	17 to 19 weeks gestation	RCT	n = 544. Women in the intervention group received an information brochure, including instructions on how to use a fetal movement chart, and were asked to count fetal movements daily from gestational week 28. The brochure, which included a fetal	Control period. Usual care.	No perinatal deaths occurred in either group. However, the growth-restricted foetuses were more often identified prior to birth in the intervention group than in the control group; 20	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. High-quality.

	Monitoring fetal movements				movement chart (a kick chart), was provided at the ultrasound screening.The brochure covered information on: expected normal fetal activity; differences in perception according to different fetal movements, maternal position, the inter- and intra-individual variation between fetuses, maternal weight, and smoking; interpretation of variation of fetal activity; instructions on how to use the kick chart; and when to contact health professionals if experiencing DFM	of 23 foetuses (87.0%) versus 12 of 20 foetuses (60.0%), respectively, [RR: 1.5 (95% CI 1.0–2.1)].	
Saastad et al. (2010), Norway	To evaluate an intervention of implementation of uniform information on fetal activity to women during the antenatal period. **Behavioural target:** Monitoring fetal movements	No explicit definition	17 to 19 weeks gestation	Cross-sectional study		The stillbirth rates among women with DFM were reduced by almost 50% (OR 0.51; 95% CI 0.32–0.81, p = 0.004)) from 4.2% (n = 50) to 2.4% (n = 73) during the intervention. Stillbirth rates among women in the entire cohort were reduced by one third from 3.0/1000 to 2.0/1000 (OR 0.67; 95% CI 0.48–0.93, p = 0.02). Stillbirth rate in their total cohort of births was comparable to the rest of Norway in the baseline	JBI Checklist for cross-sectional studies. Moderate quality.
Tveit et al. (2009), Norway	To examine two cohorts of women with DFM before and during a quality improvement intervention by implementing guidelines for management of DFM and uniform	Antepartum, intrapartum or neonatal death from 28 completed weeks of gestation	All pregnant	Prospective cohort study			JBI Checklist for cross-sectional studies. High-Quality

	information on fetal activity to women. **Behavioural target:** Monitoring fetal movements.						observation (OR 1.06; 95% CI 0.70–1.65, p = 0.73), and significantly lower during the intervention period (OR 0.64; 95% CI 0.47–0.87, p = 0.005).	
Intervention 8: A controlled study of daily fetal movement counts in the prevention of stillbirths[602]								
Lobb et al., (1985), UK	To assess the impact of maternal fetal counting on stillbirth outcomes. **Behavioural target:** Monitoring fetal movements.	Stillbirths were subdivided into those weighing more than 1500g at birth and those weighing 1500 g or less.	From 28 weeks gestation	Non-randomised trial	n = 6,597. The Cardiff "Count to Ten Kick Chart" was introduced by two consultants to all women booked at the Liverpool Maternity Hospital. The women were instructed to telephone the hospital at 21:00hrs if they ha less than 10 fetal movements for 2 days, or no fetal movements for 12 hours.	n = 13,705. Kick chart is introduced to a selected, high-risk patients and accompanied of usual care.	Daily fetal movement counts failed to reduced the incidence of intra-uterine deaths.	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. Low quality.
Intervention 9: Modifying maternal sleep position in late pregnancy through positional therapy[271,593]								
Warland et al., (2018), Australia and USA	To test the effectiveness of PrenaBelt to reduce time spent sleeping supine in a group of healthy pregnant women	Stillbirth or perinatal death are not outcomes, however ultimate aim of device is to reduce the	From 32 to 38 weeks gestation	RCT	n = 13, Participants are their own control. Women were prompted to use a device called Prenabelt, which causes subtle pressure points on the pregnant user's lower	n = 13, The study lasted for two nights, one night participants receive the intervention whereas the other night participants	No stillbirths occurred in the study participants. The study concludes that reducing time spent in the supine maternal	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria.

	in the third trimester of pregnancy. **Behavioural target:** Sleep position.	rates of stillbirth			back and buttocks when she lies in the supine position and thereby activate her body's natural mechanisms to spontaneously reposition to maintain comfort and reduce time spent sleeping supine .	are treated as control group (no device).	sleep position may ask as a protective factors against stillbirth.	Moderate quality.
Kember et al. 2018 (Canada, USA, Ghana)	To evaluate whether the percentage of time spent supine during sleep in the third trimester of pregnancy could be reduced using a positional therapy device (PrenaBelt) compared with a sham device. **Behavioural target:** Sleep position.	Stillbirth or perinatal death are not outcomes, however ultimate aim of device is to reduce the rates of stillbirth	Between 28 and 37 weeks gestation	Randomised pilot trial	n = 10. Each participant underwent two overnight polysomnograms, one wearing the Prenabelt device and another one wearing a sham. The PrenaBelt is worn at the level of the waist and has four firm plastic balls embedded into foam inserts within the pockets posteriorly. When women are supine, the balls apply pressure points across the woman's lower back, prompting her to change position to a lateral position to remain comfortable.	n = 10. Each participant underwent two overnight polysomnograms, one wearing the Prenabelt device and another one wearing a sham. The sham was identical to the PrenaBelt but had soft foam balls instead of firm plastic balls. It did not provide pressure points.	The percentage of sleep time supine on the PrenaBelt night (n = 20) and sham night (n = 20) was 3.5% (0–16.6) and 16.4% (3.5–25.3), respectively. Use of the PrenaBelt resulted in a reduction in the percentage of sleep time supine in comparison with the sham,	Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria. High-quality.

8.7.3 Behaviour Change Techniques

A total of 27 BCTs were identified across all nine interventions. The most common BCTs were “Information about health consequences” (n = 7, 78%) and “Adding objects to the environment” (n = 7, 78%). The BCTTv1 Taxonomy[302] contains 3 BCTs related to social support which when added together were present in 7 out of the 9 interventions (78%) included in this review, either being “Social support (unspecified)” (n = 5/9, 55.5%), “Social Support (practical)” (n = 1/9, 1.11%), “Social support (emotional)” (n = 1/9, 1.11%). The maximum number of BCTs identified in an intervention was 10[594], and the minimum was two[593]. The mean number of BCTs used in the interventions was 6.11 and the median was 6 (See Table 8.3).

The three interventions focusing on monitoring fetal movements used eight different BCTs in total, being "Problem-solving", "Self-monitoring of behavioural outcomes", "Social Support (unspecified)”, “Instructions on how to perform the behaviour”, “Information about health consequences”, and “Adding objects to the environment” the most common BCTs used (present in 2 out of the 3 interventions). The maximum number of BCTs used in the interventions focusing on monitoring fetal movements was 6[596] and the minimum was 3[599].

Only one intervention looking at smoking and another one looking at sleep position were included in this review. The intervention focusing on smoking[594] utilised seven different BCTs, and the intervention looking at sleeping position utilised only two BCTs[271,593] (See Table 8.3).

The multi-target interventions used a total of seventeen different BCTs. “Instructions on how to perform the behaviour”, “Information about health consequences” and “Adding objects to the environment” were the most used BCTs in multi-component interventions. The maximum number of BCTs used was 10[594], and the minimum number of BCTs used was 8[589].

The BCT "Information about health consequences" was operationalised in different ways depending on the intervention such as providing information to women who smoked and providing complementary educational resources. “Adding objects to the

environment" was operationalised as providing videotapes, leaflets, brochures, charts, or a positioning device (Supplementary file 8.5).

Table 8. 3: BCTs identified in the interventions.

Behavioural target of intervention	Multi-target interventions				Smoking	Maternal monitoring of fetal movements			Sleep position
Intervention	1.The safer baby bundle	2. Intervention to Reduce Behavioural and Psychosocial Risks	3. Keeping baby SAFE in pregnancy	4. Saving babies' lives project	5. Financial incentives for smoking cessation	6. My Baby's Movements	7. Introduction of fetal movement information and guidelines	8. A controlled study of daily fetal movement counts	9. Modifying Maternal Sleep Position
1.1. Goal setting (behaviour)	x				x				
1.2 Problem solving		x							
1.4. Action planning		x	x				x	x	
2.1. Monitoring of behaviour	x			x					
2.2 Feedback on behaviour				x					
2.3. Self-monitoring of behaviour		x				x			
2.4. Self-monitoring of outcomes of behaviour						x	x		
2.5. Monitoring of outcome(s) of behaviour without feedback	x								
2.6. Biofeedback				x					
3.1. Social support (unspecified)		x		x	x	x	x		
3.2 Social support (practical)	x								
3.3. Social support (emotional)			x						
4.1. Instruction on how to perform the behaviour	x	x	x				x	x	
5.1. Information about health consequences	x	x	x	x	x	x	x		

5.2. Salience of consequences			x						
6.2. Social comparison			x						
7.1. Prompts cues						x			x
7.5. Remove aversive stimulus		x							
10.1. Material incentive (behaviour)					x				
10.2. Material reward (behaviour)					x				
10.4. Social reward		x							
10.5. Social incentive									
10.10. Reward (outcome)					x				
11.1. Pharmacological support	x				x				
12.2. Restructuring the social environment		x							
12.5. Adding objects to the environment	x	x	x	x		x	x	x	x
16.3. Vicarious consequences			x						

Intervention effects

The interventions effects of the interventions included in this review have been reported narratively in this section. The efficacy findings are reported as per the original authors.

The interventions focused on maternal monitoring of fetal movements show different results. Intervention number 7 “Reduction of late stillbirth with the introduction of fetal movement information and guidelines”[596] reported a reduction in the rates of stillbirth amongst women presenting with decreased fetal movements from 4.2% to 2.4% (OR 0.51 95% CI 0.32–0.81, p = 0.004) during the intervention in primiparous women compared to women who presented with decreased fetal movements in the period before the intervention. Furthermore, the authors compare their stillbirth rates at baseline and during the intervention period with data from the Medical Birth Registry in Norway, and conclude that the rates of stillbirth at baseline were comparable, but that they were significantly lower during the intervention period (OR 0.64; 95% CI 0.47–0.87, p = 0.005).. Stillbirth rates among women in the study cohort were reduced as well from 3.0 per 1000 to 2.0 per 1000 (OR 0.67; 95% CI 0.48–0.93, p = 0.02); also compared with the general national stillbirth rate the cohort stillbirth rate was also significantly lower during the study period (OR 0.64; 95% CI 0.47–0.87, p = 0.005). This intervention was associated with better detection of fetal growth restriction as well (from 2.4% to 4.4%, p = 0.020 in term pregnancies (≥ 37 weeks) and remained unchanged in the preterm (4.5% vs. 4.0%, p = 0.604). However, in Intervention number 8 "A controlled study of daily fetal movement counts in the prevention of stillbirths”[599] the authors report no changes in pregnancy outcomes or the rates of stillbirth after the intervention. Intervention number 6 “My Baby’s Movements”[599] did not show statistically significant differences in stillbirth rates at ≥28 weeks of gestation (odds ratio adjusted for calendar time and hospital effects, aOR 1.18, 95% CI 0.93– 1.50; P = 0.18). Further, when looking specifically at the “My Baby’s Movements” app users, there were no statistically significant lower rates of stillbirth at ≥28 weeks gestation either (aOR 0.79; 95% CI 0.51– 1.23; P = 0.30). App users had higher rates of induction of labour (aOR 1.10; CI

1.06– 1.14; P < 0.01) and caesarean section, although not statistically significant in the last case (aOR 1.02; CI 0.98– 1.06; P = 0.33).

One of the included interventions focused on smoking cessation. Intervention number 5 “Financial incentives for smoking cessation in pregnancy: randomised controlled trial"[590] reported that more intervention participants stopped smoking compared to the control group (22.5% vs. 8.5%). However, no differences were observed when looking at perinatal outcomes such as stillbirth, miscarriage or premature birth.

Regarding the intervention focusing on sleeping position, Intervention number 9 “Modifying Maternal Sleep Position in Late Pregnancy Through Positional Therapy: A Feasibility Study[593]” concluded that the use of the PrenaBelt device reduced the time that women spent in the supine position (3.5% vs. 16.4%, p = 0.03), however, it also reduced the length of their sleep. No stillbirths occurred in the study sample.

Intervention number 7 “Reduction of late stillbirth with the introduction of fetal movement information and guidelines”[594,598] reported a decrease in the psychosocial risk factors (smoking, environmental smoking exposure, depression and intimate partner violence)[603], however, there were no differences regarding perinatal outcomes (miscarriage, perinatal death, pre-term and very pre-term birth, low and very low birth weight, small for gestational age, NICU admission, hospitalisation)[594].

Intervention number 3 “Keeping Baby Safe”[589] reported increases in the participant’s knowledge regarding stillbirth and risk factors for stillbirth in the post-intervention measurements.

Intervention number 4, “The Saving Babies’ Lives care bundle”[237,238,589,600] has shown results in the UK. This care bundle has four main elements: reducing smoking, prevention and surveillance of pregnancies at risk of growth restriction, raising awareness about reduced fetal movements, increasing effectiveness of fetal monitoring during labour and reduction of preterm birth. The authors reported reduced stillbirth rates in the 20 maternity participating units from 4.2 to 3.4 per 1,000 births, which is a greater reduction than the occurred in the rest of England. However, the authors are not able to attribute this decrease in stillbirth rates to the

implementation of the care bundle, however, as they report, it is highly plausible that the implementation of the Saving Babies' Lives care bundle contributed to this improvement.

Intervention number 1 “The Safer baby bundle” has the objective of reducing stillbirth by tackling smoking, increasing surveillance of fetal growth restriction, increasing awareness and monitoring of fetal movements, awareness of sleep position, and improving decision-making around the time of birth. The intervention is still in the implementation phase in most regions of Australia. However, the care bundle has already been evaluated in the Victoria Jurisdiction, showing a decrease in the stillbirth rates of 21% (from 0.24% to 0.19%), and an increase in the number of days without any stillbirths by 131%, from an average of 3.5 days to 8.1 days. The authors also reported an increase in smoking cessation rates from an average of 11% to 33%[243].

8.8 Discussion

This is the first review to systematically examine BCTs in behaviour change interventions that were designed to prevent stillbirth. Nine interventions were identified. Of them, 4 were interventions targeting multiple behaviours (multi-target interventions), one focused on smoking, three on monitoring fetal movements, and one on sleep position.

A total of 27 BCTs were identified across all interventions. Overall, the number of BCTs used in the nine interventions was small, accounting for 29% of the total BCTs available in the BCT(v1) Taxonomy. The BCT that was most used across all interventions were "Information about health consequences" and "Adding objects to the environment" which usually were additional informative materials, followed by the BCTs associated with social support combined. All of these BCTs have the potential to produce behaviour change by improving women’s psychological capabilities and social opportunities. This is understandable given pregnant women’s desire for education and concerns on how to improve or maintain their health status during their pregnancy [604]. However, we know from previous research that the

information provided in general antenatal care regarding health habits and risk factors for stillbirth is very scarce, or non-existent[605].

We have not been able to link the different BCTs to intervention effects due to the heterogeneity of the interventions. Our results show that, by looking at the individual findings of each intervention, two interventions "Safer Baby Bundle" and the "Saving Babies Lives" care bundle, which both have a multi-component approach, might have had a positive influence in reducing stillbirth rates. This is in concordance with previously developed behaviour change interventions targeting different outcomes, where authors have concluded that multi-layered interventions targeting individual, societal and environmental-level determinants are more effective [606–608].

Behaviour change during pregnancy is influenced by several different factors including psychological and cognitive factors, social factors, structure/service level factors, social factors and environmental factors[410,556,609]. Alcántara et al. (2020) conducted a systematic review of behaviour change interventions to determine which types of social determinants were tested as moderators of behaviour change interventions. The authors concluded that, even though almost half of the interventions tested social determinants of health moderators, most studies were atheoretical, lacked diversity, were methodologically poor, and were too focused on the individual level[610]. The fact that multi-component interventions tackle different social determinants or behaviours at once might be the reason why they have had success tackling the risk factors for stillbirth or producing behaviour change. These types of interventions are also the ones that were composed of the biggest number of different BCTs. However, as Alcántara et al. discussed in their review, there is an increased need to design better theoretically informed and diverse behaviour change interventions to determine which social determinants of health moderators are effective in health behaviour change interventions and which BCTs are the most appropriate to tackle them[610].

Our review findings show that most of the interventions have a strong focus on providing information either by directly communicating with women or by providing women with additional materials such as books, leaflets, websites or videos. Providing women with information on the importance of engaging in certain

behaviours and disengaging from others is very relevant and might act as a first step towards behaviour change[363] but it also involves some challenges. Previous research has demonstrated that women might show different attitudes towards receiving information about stillbirth or behavioural risk factors for stillbirths, and whereas some women would receive this information positively and understand it as a tool to support prevention efforts, for others it might cause anxiety[605]. Hence, the importance of language and communication style is essential. Communicating about stillbirth also poses some challenges for healthcare professionals. Previous research has shown that talking about sensitive issues such as weight management or substance use can be uncomfortable or stressful for healthcare professionals[611–613]. Intervention designers need to consider these complications when assessing the feasibility and acceptability of their interventions. Further, it is possible that adding concrete elements such as action or goal planning, instructions on how to perform the behaviour, or practical social support contributes to addressing the potential uncertainties that the information provided might arise.

We know from previous research that lack of social support can act as a barrier to behaviour change during pregnancy[299,556,557]. Social contexts and norms are important and some women may have few positive role models in their social milieu, adding pressure on women to avoid social exclusion, or providing inaccurate information[556,557]. Further, the lack of social support to help women share their daily responsibilities (e.g, with other children) can also act as a barrier to engaging in healthy behaviours [556,557]. However, social support can also act as a facilitator of behaviour change when it is present. Having supportive partners that do not engage in unhealthy behaviours themselves or encourage family and friends has been reported as facilitators[299,556,557], as well as access to empathetic and understanding healthcare professionals[556,557]. This might explain the high level of social support BCTs (either practical, emotional, or unspecified) in the interventions included in this review. These BCTs are present in seven of the nine interventions included in this review; however, their operationalisation is in general quite vague. Hence, further research is necessary to determine how to establish consistent social support that facilitates engagement with behaviour-change interventions during pregnancy.

After examining the interventions included in this systematic review in detail, it became evident that none of the interventions included reported explicit links to behaviour change theory. This fact raises concerns about the effectiveness of these interventions, as behaviour change theory is considered to be essential during intervention design, as it helps to understand and predict how individuals might respond to an intervention[614]. Without a theoretical underpinning, it is even more difficult to determine which are the mechanisms of action that are leading individuals to behaviour change, and it limits the potential for further refinement of the intervention[614].

Additionally, most interventions included in this review had a very poor description of their intervention design process. This hinders the possibility of assessing the rigor of the intervention design process[615]. Without enough information, it is not possible to determine if the intervention was designed in a systematic way, based on evidence and theory, and with sufficient information about their target population or context[616]. Therefore, it is important for future research to not only explicitly link behaviour change interventions to theory but also provide a clear and detailed description of the intervention design process in order to enhance the transparency and reproducibility of behaviour change research.

8.8.1 Strengths and limitations

Firstly, this is the first review to systematically examine behaviour change interventions to reduce the risk of stillbirth and to identify the BCTs included in such interventions. There are several reviews published in the literature examining behaviour change interventions targeting risk factors for stillbirth (such as smoking or high BMI), and also reviews examining all available interventions to reduce the risk of stillbirth. However, we ensured this review only included interventions that were designed with the explicit purpose of reducing stillbirth and that included only behaviour change interventions, as opposed to, for example, interventions affecting care management. The use of the BCT Taxonomy V1[302] for the identification and coding of BCTs is a strength of this review. Coding the interventions using the BCTTv1 Taxonomy [302] provides a more detailed examination of the different components of an intervention, which might result in the identification of potentially effective

components [579]. However, there is a need for further research to develop an approach that facilitates linking BCTs to effectiveness across heterogeneous interventions and outcomes.

To date, establishing the link between individual BCTs and intervention effectiveness is not possible given the heterogeneity of the different interventions tackling stillbirth. However, some approaches are being currently developed to tackle this issue. Johnson et al., (2022) proposed in their protocol a method to provide preliminary information about the effectiveness of commonly used intervention components (used in five or more interventions) in their review of early childhood obesity prevention interventions [581]. Although the authors acknowledge that their approach will not be able to determine the independent effects of each component, it might be a step closer to linking individual BCTs to intervention effectiveness, which would inform intervention design as to which "active ingredients" are necessary for a behaviour change intervention to be effective. Taxonomies and ontologies can be used to deconstruct intervention components. These new approaches might facilitate linking individual BCTs to intervention effectiveness. Further research in this regard is also necessary to develop methodologies that allow intervention designers to determine the independent effects of each component over an outcome of interest. However, identifying which BCTs have been used to date in successful interventions is important to inform the development of new interventions.

We have focused our analysis on the behavioural aspects of the interventions included. However, it is important to highlight that some interventions such as the Saving Babies' Lives Care Bundle [237,238,597,600], which has reported reductions in stillbirth rates, also included elements associated with the behaviour of and care provided by healthcare professionals. This highlights the fact that strong support by the institutions and professionals providing care is possibly necessary to reduce the rates of stillbirth.

8.9 Conclusion

Our findings show that multi-target interventions (interventions targeting multiple behaviours) seem to have the best potential for success. Further research on the factors affecting behaviour change during pregnancy needs to be conducted to design evidence-based behaviour change interventions that go beyond information

provision and that include more concrete action to produce behaviour change, utilising BCTs such as “Instructions on how to perform a behaviour”, “Goal settings” and “Action Planning”. Using structured methodologies like the Behaviour Change Wheel [259] for intervention design can help to identify and address all the factors influencing behaviour change by selecting appropriate intervention functions, and guides to select useful BCTs as well as potential operationalisation of BCTs[259].

Finally, further research is essential to establish the efficacy of the different BCTs to inform the development of future stillbirth prevention interventions.

8.10 Inter-chapter synopsis

Chapter 8 aimed to identify which behaviour change interventions have been designed to date in the context of stillbirth prevention, and what BCTs were composing them. The systematic review identified nine interventions with 27 different BCTs used. The most commonly used BCT was "Information about health consequences," followed by "Adding objects to the environment.". This study highlights the need for evidence-based behaviour change interventions that address the behavioural risk factors for stillbirth, including social and environmental barriers. The gap in behaviour change intervention design identified in this study can be addressed by utilising the data obtained in all the previous chapters composing this Thesis to inform the development of a behaviour change intervention. In Chapter 9, the findings of all of the previous studies are brought together and their implications for policy, practice and reserach are discussed.

Chapter 9 – Discussion

Chapter 9 – Discussion

9.1 Chapter overview

This final chapter summarises and integrates the main findings presented within the different studies in this thesis. The findings are considered in the context of the existing evidence and presented highlighting the relevance of the multi-layered influences over the behaviours that have been associated with an increased risk of stillbirth during pregnancy. Further, the implications of the research in this thesis for policy development in education and public health awareness, and maternity care practice are outlined. The strengths and limitations of this research and its implications for future research, including the next steps in intervention development are discussed.

9.2 Summary of research findings

In order to be able to change maternal behaviours related to stillbirth it is essential to identify and understand the underlying factors that influence these behaviours. The overall aim of the research within this Thesis was to explore and generate evidence regarding the influences affecting women's behaviour change during pregnancy when tackling the modifiable behavioural risk factors for stillbirth. This research enhances the understanding of the modifiable behavioural risk factors for stillbirth and can be used to inform the development of stillbirth prevention and intervention strategies. The studies included in this thesis make a valuable contribution to existing knowledge and they focus on a neglected area with the potential to produce change in stillbirth rates.

The findings from the different studies will be discussed here based on the following overarching topics, which move from more person-centered specific aspects to broader societal aspects:

1. Health literacy and sources of information (Chapters 2 to 8).
2. Relationship with healthcare providers (Chapters 4 to 7).
3. Healthcare system structural barriers (Chapters 3 to 8).

4. Interpersonal and social factors: silence around stillbirth and societal stigma (Chapters 2 – 8)

A summary of the research findings of each study can be found in Table 9.1.

Table 9. 1: Summary of research findings presented in this Thesis based on overarching topics.

Chapters	Main findings	Overarching topics			
		Health literacy and sources of information	**Relationship with HCPs[1] and communication issues**	**Healthcare service structural barriers**	**Interpersonal and social factors: silence around stillbirth and societal stigma**
Chapter 2	Four main modifiable risk factors with a behavioural component were found to have the strongest evidence: ▪ Substance use (smoking, alcohol, illicit drugs) ▪ Maternal weight ▪ Attendance & compliance with antenatal care. ▪ Sleep position.	▪ The evidence regarding some of the behavioural modifiable risk factors for stillbirth is conflicting (e.g., small amounts of alcohol, some prescription drugs, herbal and dietary supplements, gestational age at first booking, sleeping less than 6hrs per night).	NA	NA	NA
Chapter 3	▪ <50% of websites contained information about stillbirth ▪ <30% of websites contained information about risk factors for stillbirth ▪ Only one website contained all the information sought about stillbirth (e.g. definition, prevalence, etc.) & risk factors.	▪ Online resources have been underused to provide information about stillbirth and risk factors.	NA	▪ Professional body health services websites are not being used to provide information about stillbirth and risk factors. ▪ Professional body websites and health body websites rely heavily on providing links to clinical guidelines, which are not user friendly.	▪ The lack of information about stillbirth and behavioural risk factors for stillbirth in general websites directed at the general pregnant population might be a consequence of the stigma and taboo associated with these topics.

[1] Healthcare professionals

Chapter 4 – 6	Identified areas of concern: ▪ Health literacy, awareness of risks & benefits ▪ Insufficient & overwhelming sources of information ▪ Lack of opportunities & HCPs' attitudes interfering with communication & discussion ▪ Social influence of the environment ▪ Social judgement, stigmatisation of women. *A search to identify facilitators and barriers influencing sleep position was also conducted at two different points in time during this PhD, however, no qualitative research was identified.	▪ Lack of reproductive knowledge acted as a barrier for care seeking behaviour as it delayed women's recognition of their pregnancies. ▪ The lack of reliable sources of information led some women to rely on anecdotal evidence and creating misconceptions about the risks of their behaviour. ▪ A lack of satisfaction with education and advice received during antenatal care was identified. ▪ Advice during antenatal care was too general and not individualised to specific needs. ▪ Women's levels of health literacy have an influence on their perception of risk and incentives, which will influence their engagement in behaviour change.	▪ HCPs attitudes (stigmatisation, racism, lack of empathy, insensitivity) had an influence on women's willingness to engage with antenatal care. ▪ Non-judgemental, supporting, and empathetic HCPs were highly valued. ▪ Communication problems with healthcare professional were identified (contradictive messages, having concerns dismissed, feeling uninformed, being confused due to jargon) ▪ Authoritarian or coercive communication styles. ▪ Active listening from HCPs, taking time to address concerns, and empowerment of patients were perceived as positive communication.	▪ Negative clinical experienced affected women's behaviour negatively. ▪ These might be rushed appointments, administrative delays, problems with referral, lack of flexibility and of continuity of carer, lack of individualised care, complexity of the application process for care. ▪ Cultural inappropriateness and language barriers were identified as barriers to behaviour change. ▪ Limited financial resources of healthcare systems resulting in shortage of staff, closure of clinics and limitations associated with government health programmes and cost of care.	▪ Social position and lifestyle factors such as homelessness, living in deprived areas, low economic resources were identified as barriers for behaviour change. ▪ Social discrimination of certain collectives (ethnic minorities, socioeconomic status) ▪ Lack of social support influenced women's behaviour. ▪ Social pressure and stigma generated feelings of shame and guilt, that acted as a stressor for women and prevented engagement in behaviour change. ▪ Environmental barriers such as lack of access to sports facilities, or distance to maternity services or clinics.
Chapter 7	▪ Behaviour change during pregnancy perceived as easy and natural. ▪ Women had high level of awareness regarding health	▪ Women were aware of what habits are recommended and encouraged during pregnancy for better health. ▪ Awareness about stillbirth and the link of the behavioural risk factors with stillbirth was very limited.	▪ The importance of communication with HCPs was highlighted, encouraging the use of sensitive and non-blaming language.	▪ Women expressed a preference for interventions delivered in group settings, rather than targeted at one-on-one level.	▪ Stillbirth was perceived as a taboo work for some women, and it was described as a difficult topic to talk about.

	advice, but very limited regarding stillbirth. ▪ There is a lack of discussion with HCPs about stillbirth & risks, so women rely on their own information-seeking behaviours. ▪ Women had a general positive attitude towards receiving information about stillbirth; knowledge perceived as key.	▪ Women reported lack of discussion regarding stillbirth and risk factors during their antenatal care. ▪ Lack of advice regarding health behaviours and behavioural risk factors during antenatal care. ▪ Women were not informed about the reasons why those behaviours were relevant. ▪ Women relied on their own information seeking behaviours to raise their own awareness about health habits during pregnancy. ▪ Most women perceived receiving information about stillbirth as helpful to support prevention efforts.			
Chapter 8	▪ 9 interventions were included in analysis. ▪ The most common BCT used was "Information about health consequences", followed by "Adding objects to the environment". ▪ The maximum number of BCTs was 11 and the minimum was 2.	▪ The behaviour change interventions conducted to date in the context of stillbirth prevention have a high focus on information provision.	NA	▪ The interventions do not take into consideration potential structural barriers, and the ones that have shown effects on stillbirth rates require great investment at system level.	▪ The very limited number of behaviour change interventions highlights how this is an issue that has been neglected in the research agenda, potentially in part due to the stigma associated with the topic.

9.2.1 Health literacy and sources of information

Issues related to knowledge, awareness, education and availability and quality of sources of information regarding stillbirth and modifiable behavioural risk factors are common amongst most of the studies comprising this Thesis. In the three meta-synthesis of qualitative research (Chapters 3-6), a lack of awareness related to the different behaviours linked with potential adverse pregnancy outcomes, and the effects of such lack of awareness on risk perception and behavioural choices, as well as issues related to sources of information and conflicting evidence therein, were identified.

Women in the studies reviewed for the meta-synthesis of qualitative research work held misconceptions about the risks of substance use and engaging in weight management behaviours, and they also held misconceptions that altered for them, the perceived value of attending antenatal care (Chapters 4-6). All of these misconceptions, mostly consequences of a lack of information, or of using anecdotal evidence, or their social context, as sources of information, had an effect on women's behaviour by altering their perceptions of benefits of action versus risks of consequences. The qualitative study with postpartum women (Chapter 7) and the survey study (Supplementary file 1.1) conducted with HCPs show that information provision during antenatal care about stillbirth, health behaviours and modifiable behavioural risk factors for stillbirth is very scarce. Findings from the qualitative study (Chapter 7) highlighted that women were able to use external sources of information (e.g., websites, apps, books, videos) for their own pregnancy related education, however this is not a good or sufficient solution for all women. Hence, we can conclude from the findings of these studies that opportunities for education and information provision are being missed during antenatal care to address health behaviours and behaviour change for stillbirth prevention. In Chapter 2, we were also able to identify conflicting evidence in the literature regarding some behavioural risk factors for stillbirth (e.g., small amounts of alcohol reported as safe, contradictory information about some prescription drugs and herbal supplements, gestational age at first booking considered not relevant for stillbirth risk) and this lack of clear

evidence might potentially be influencing both information provision and awareness of HCPs and the public negatively.

Further, in Chapter 3, the findings indicated that online resources in Ireland and the UK are not a good source of information when it comes to stillbirth and modifiable behavioural risk factors. Most websites provided very scarce general information about stillbirth (e.g., prevalence, causes, etc.), and the information regarding modifiable behavioural risk factors was mostly focused on smoking. These findings might partially explain the findings of a previous study conducted by Nuzum et al. (2018) showing that the awareness of stillbirth and its risk factors amongst the general Irish population was very limited[258]. The role of the internet for health related purposes has increased in the last couple of decades[617], however, the availability of online sources and ease of accessibility to varied information raises concerns about the quality and accuracy of the information accessed. This study showed that official websites such as those provided by professional bodies and national health services relied heavily on external links to guidelines to provide information, and did not use their own websites as an information tool. These external links are useful for people who are interested in expanding their learning and exploring the main evidence sources, however, they are not user friendly, hence, not appropriate for the general public. Therefore, the internet is still a resource that can be positively exploited for information provision regarding stillbirth and modifiable behavioural risk factors, as well as serving to provide advice to support health and behaviour change during pregnancy.

The role of information is also critical in risk perception[618]. The creation of accurate or inaccurate risk perception is relevant for health and health behaviour change[618], with unrealistic optimism being a common bias amongst people[618]. Unrealistic optimism refers to the belief that our own personal outcomes will be more favourable than what might be expected from existing evidence[619]. Previous researchers have theorised that unrealistic optimism can be a consequence of people's motivation to anticipate favourable future events, people's overreliance on personal information when making judgements, and people's processing of information in ways that lead them to be optimistic[619]. However, the literature

exploring unrealistic optimism in relationship with health outcomes and health behaviour is contradictory. Unrealistic optimism has been at times linked to increased hope, positive affect and greater goal-directed persistent leading people to engage in health-promoting behaviours[619]. However, most studies exploring realistic optimism in relation to health behaviours correlate unrealistic optimism with unnecessary risks, misplaced hope, disappointment, regret and other negative emotions, and paying less attention to relevant information[619]. Hence, it is necessary to ensure that women have realistic perceptions of risks during their pregnancies in order to make informed decisions.

9.2.2 Relationship with HCPs

The influence of HCPs attitudes and behaviours over women's experiences of care and behaviour change is also a common topic that has been identified in the studies comprising this Thesis. In the three meta-synthesis of qualitative research (Chapters 3-6), women who encountered supportive, non-judgemental, and empathetic HCPs were more willing to engage in their care and behaviour change than women who encountered HCPs that were judgemental and/or lacked communication skills. Having a negative clinical experience influenced by HCPs' negative attitudes acted as an obstacle to behaviour change for pregnant women, regardless of the behaviour explored. Previous research in the field of oncology has established that the type of relationship and communication between patients and HCPs will influence patients' ability to cope with their condition and their overall satisfaction with care[620]. This finding is supported by the results of a systematic review conducted by Mannava et al. (2015) exploring the types of impact that maternity health care providers (MHCPs) attitudes and behaviours have towards women[621]. The authors identified different types of positive attitudes and behaviours adopted by MHCPs such as being caring, encouraging, supportive, friendly, sympathetic, polite, informative, helpful and attentive. Some of the factors the authors associated with MHCPs adopting these behaviours were related to the MHCPs familiarity with the cultural practices and community of the patients[621]. This explanation is also supported by the findings of the meta-synthesis exploring attendance at antenatal care (Chapter 5), where

women from different ethnicities or sociodemographic backgrounds encountered additional barriers to accessing and engaging with their care.

Mannava et al. also found that MHCPs attitudes and behaviours are affected by factors at the individual, organisational, and societal level[621]. At the individual level, issues related to the MHCPs beliefs and characteristics (stigmatisation, stress, fatigue, level of tolerance, self-esteem, motivation and training), the provider-patient relationship (hierarchy, familiarity, expectations), and patient attitudes and behaviours (fear, shyness, attitudes towards service) were observed[621]. Work conditions (workload, work hours), work environmental factors (supportive supervision, relationship with co-workers, availability of equipment) and being part of the public or private sectors were factors identified at an organisational level[621]. When it comes to societal-level factors, the authors identified cultural beliefs and contextual factors as influences over MHCPs behaviours[621]. These findings were partially supported by the survey study with HCPs (Supplementary file 1.1) where some of the HCPs identified lack of time and training as barriers to enhancing their communication regarding health habits with women under their care. Similarly, in the meta-synthesis of qualitative research work, women were found to experience rushed/medically focused appointments (Chapters 4-6), and in the qualitative study, women spoke about receiving very limited education and information during their antenatal care (Chapter 7). This limitation in time and staffing levels is not restricted to Ireland; the "crisis in human resources" in the healthcare sector is a global health issue[622]. Therefore, when considering HCPs' attitudes and behaviours and potential consequences over the relationship they create with patients, issues such as burnout, stress, work overload, and personal burden need to be considered. The studies in the Thesis have reflected how some of the topics associated with stillbirth communication and behavioural risk factors are perceived as difficult topics to discuss (Chapters 4-7). Women in the qualitative study (Chapter 7) highlighted the importance of using sensible language when providing information about stillbirth. However, the women participating in the reviewed studies (Chapters 4-6) reported different difficulties when discussing issues such as substance use and weight management with HCPs, given the social disapproval and stigma associated with

topics such as substance use and having overweight during pregnancy which will be discussed in section 9.2.4. Therefore, HCPs' attitudes, behaviours and cultural appropriateness might have an even stronger influence on women's experiences when it comes to sensitive topics such as stillbirth or its risk factors (Chapters 4-7).

Sharing the responsibility of improving reproductive health and pregnancy health of women with education (e.g., schools, secondary schools, universities) and/or community supports (e.g., public health nurse antenatal classes in local community centres) could be a way to unburden HCPs when caring for women. HCPs attitudes and behaviours and women's consequent relationship with healthcare providers are affected at multiple layers (personal, institutional, structural, societal), as observed in the findings of these studies (Chapters 4-7). All of these layers need to be considered when addressing the prevention of stillbirth via behavioural risk factors.

9.2.3 Healthcare system structural issues

Issues related to the healthcare system were also identified in six of the studies that make up this Thesis (Chapters 3-8). Firstly, the website quantitative content analysis (Chapter 2) identified that healthcare services and professional bodies were not utilising their online resources effectively as a source of information for the public, including pregnant women. Providing women and partners with up-to-date, reliable sources of information from credible sources could help unburden the maternity health system. This could be achieved by giving patients the necessary education to better able to identify risk factors and first signs of disease, and patients that are better informed about their treatment options, etc.

The meta-synthesis of qualitative research (Chapter 3-6) identified barriers related to the healthcare system that affected all behavioural risk factors explored such as administrative delays, issues with referral services, lack of flexibility, lack of continuity of care/carer, lack of individualised care, insurance status-related issues, cultural inappropriateness, and language barriers. Some of these issues were also highlighted by women in the qualitative study (Chapter 7) who felt that the appointments with their HCPs were fast, they understood HCPs need to prioritise medical procedures rather than education around health during pregnancy.

These structural barriers have an impact on women's perceived clinical experience (Chapters 4-6). Findings showed that women in the reviewed articles were willing to seek care, but the barriers they encountered affected their motivation levels (Chapter 4). The influences of structural barriers over an individual's behaviour can be understood through the COM-B model, which takes into consideration these aspects in its "Opportunity" element[277]. Encountering "physical opportunity" barriers such as administrative delays and lack of access to clinics or healthy food shops within a short distance (Chapters 4-6), as well as barriers associated with "social opportunity", such as overworked HCPs who aren't available for women (Chapters 4-6, Supplementary file 1.1) can directly influence women's individual motivation and subsequently, behaviour.

Previous research has suggested that one way of tackling structural barriers to accessing antenatal care is by making changes in the format of antenatal care and policies around maternity services[623]. Some of the facilitators related to the structural barriers that were identified in the meta-synthesis of qualitative research, and could be tackled through policy development, include: providing flexible walk-in clinics; providing specialised services for women from different socioeconomic backgrounds (e.g., clinics for immigrant women, translation services); facilitating flexible appointments and support to engage with the health system; and creation of policies to support women in disadvantaged situations to prioritise their care during pregnancy (e.g., access to childcare, access to social housing, access to free healthcare). However, to identify which are the most appropriate measures for a specific health service, further research is necessary to identify the specific needs and preferences of different groups, such as women from different socioeconomic backgrounds.

9.2.4 Interpersonal, social and structural factors: Silence around stillbirth and societal stigma.

The findings of the studies in this Thesis (Chapters 2 – 8) have highlighted that there is a silence surrounding stillbirth. In Chapter 3, we identified that websites avoid dedicating space to stillbirth, as the number of websites containing this information is small. Stillbirth has been described in the literature before as a silenced topic by society[570]. The social stigma associated with stillbirth might contribute to preventing website content developers to include more information about stillbirth and related behavioural risk factors in their websites. Online resources need to have the ability to attract and maintain users in order to be sustainable[624]; therefore, focusing on topics that might cause controversy amongst users due to the social stigma associated with them might work against this purpose. Findings from the meta-synthesis of qualitative research work (Chapters 4-6), the qualitative study (Chapter 7) and the survey with HCPs (Supplementary file 1.1) indicated that stillbirth and its associated behavioural risk factors are difficult topics to discuss and highlighted the importance of using the appropriate language when talking to women. The qualitative study (Chapter 7) with postpartum women also exposed some issues related to stillbirth silence. Women reported that that "the word [stillbirth] was never mentioned" to them and that never received any information. Some of the women confused the term stillbirth with miscarriage or sudden infant death syndrome. Women also acknowledged that stillbirth is a topic that they would not choose to speak about during their care and suggested that the conversations about behavioural risks factors should be conducted without using the word stillbirth. In the survey study a lack of HCPs knowledge about the definition and prevalence of stillbirth was identified (Supplementary file 1.1). The majority of HCPs (89%) reported having learnt about stillbirth and behavioural risk factors for stillbirth during postgraduate training and through professional guidelines, even though they started caring for women experiencing stillbirth very early in their careers. This shows again the lack of discussion of this topic during HCPs' early training. Moreover, findings of Chapter 8 showed that the number of behaviour change interventions identified in the literature designed to prevent stillbirth was only nine, once again highlighting the

lack of attention that has been given to this topic from a behaviour change intervention design perspective.

The topic of silence, shame and stigma was not only associated with stillbirth in the findings of this Rhesis. Shame and guilt were factors that were also identified as influencing with behaviour change during pregnancy, especially in the case of women who engage in substance use behaviours or that have increased BMI (Chapters 4 and 6). Women in the different studies (Chapters 4-6) expressed feelings of shame and guilt for not seeking antenatal care (Chapter 4), for engaging in substance use behaviour (Chapter 5), or for having an increased BMI (Chapter 6). In some instances, this shame led women to avoid speaking with HCPs or avoid engaging in healthy behaviours (such as physical activity). For other women shame and guilt also acted as an additional stressor that led them to engage in their unhealthy behaviour even further (e.g., continuing or increasing their substance use) (Chapter 5).

As shown in the results (Chapters 4-6) and previous research[625–627], stigma can also impact women at organisational and societal levels through institutional stigma[625]. Institutional stigma is described as the process in which the negative attitudes, beliefs and subsequent policies of an organisation will negatively impact how a condition or a group of people is treated in a specific institution[625]. In the case of smoking, alcohol consumption and illicit drug use, previous research has highlighted that stigma is amplified during pregnancy, with pregnant women reporting increased judgement and shame from individuals who were previously tolerant or participating in their behaviours before pregnancy (e.g., smoking together)[3,625,626]. As found in Chapter 5, women with a substance use disorder might delay seeking care and might avoid disclosure of their use, especially when they are afraid of potential legal consequences. Findings (Chapter 5) and previous research have also described how this stigma leads women with substance use disorders to social isolation and concealment of the pregnancy and avoid medical care[628].

Similarly, smoking during pregnancy has been reported in the literature as being "demonised" and being perceived as a moral judgement on the woman, which has consequences on the women´s relationships with their carers[3]. Alcohol consumption is also associated with discrimination and stigmatisation, with research reporting

that women who did not abstain from alcohol use during their pregnancies experienced fear of judgement from their HCPs, again leading to concealment of the alcohol use[3]. These findings are also supported by the findings of the qualitative study (Chapter 5), where women expressed negative, strong attitudes towards substance use during pregnancy. Furthermore, pregnancy-related weight stigma is also a phenomenon that has been widely highlighted in the literature[626]. In the findings (Chapter 6), the stigma associated with weight led some women to delay care and to feel less prepared to engage in physical activity due to embarrassment. Previous research has associated pregnancy-related weight stigmatisation with depressive symptoms, psychological stress, maladaptive dieting behaviour, emotional eating behaviour and postpartum weight retention[627]. Stigmatisation related to ethnicity or socioeconomic background was also highlighted as a barrier to attending and engaging with antenatal care (Chapter 4). For example, women with a substance use disorder, without private health insurance or from different ethnicities, were subjected to further discrimination. For women from disadvantaged backgrounds, institutional discrimination and racism were huge barriers to engaging in behaviour change.

These findings show that the silence and stigma around stillbirth and its associated behavioural risk factors are observed in clinical practice (Chapter 7 and Supplementary file 1.1), in intervention development research (Chapter 8) and in broader settings such as online resources (Chapter 3). The silence around stillbirth hinders prevention efforts by interfering with information provision and prioritisation in research and intervention development but most importantly, it dismisses bereaved parents' grief[629]. Previous research has described how bereaved parents have felt silenced when trying to discuss their experience or talk about their stillborn baby[27]. This silence is stigmatising[570,571] and has been reported to be one of the biggest impediments to reducing stillbirth rates and supporting bereaved parents[630,631]. Moreover, a more socio-ecological approach addressing all of the different influences over women's behaviour needs to be taken into place when tackling women´s behaviours during pregnancy to avoid victim blaming[3]. Enhancing HCPs' understanding of all of the influences on women's behaviour (e.g., health

literacy, psychological factors, sociodemographic factors, the influence of stigmatisation and shame, and societal influences) that go beyond their control, might serve to reduce HCPs' judgement and relationship with patients, thus enhancing self-disclosure.

Findings showed that external factors will also alter women's perceived value of engaging in a behaviour, women's knowledge influencing their perceived risk, their clinical experience and their capacity to seek support for behaviour change (Chapters 4-6). All the topics discussed (health literacy, relationship with HCPs, health system structural barriers, and silence around stillbirth) are influenced by the sociocultural context (Chapters 4-6). Hence, the influence of the social environment has also been highlighted as a relevant factor in most studies (Chapters 4-6, 8). In the meta-synthesis of qualitative research (Chapters 4-6) the findings showed the importance for women of availing of social support (e.g., from a partner, family, or friends) during pregnancy, especially when it comes to trying to make a change over their own pregnancy health. Previous research has described social support as the main influence over the quality of coping with stress for pregnant women[632], as well as influencing women's depressive symptoms and quality of life[3]. The people in the close social network of a woman also affect her behaviour by acting as role models, positively and negatively. For example, women in the studies reviewed in Chapter 5 speak about the normalisation of substance use within their family and friends, and how not engaging in such behaviour made them feel isolated. On the contrary, women in the studies reviewed in Chapter 6 reported how having someone in their close context that encouraged them and accompanied them when engaging in physical activity was positive. Women who chose to act differently than the rest of their family, friends or partner (e.g., ceasing substance use, changing dietary habits, engaging in physical activity) felt pressured and isolated, questioned, and judged (Chapters 4 – 6).

The evidence of the influence of a woman's close social network over her own behaviour might explain why the findings of Chapter 8 show that intervention designers have included social support as an element to incorporate in their interventions to tackle modifiable behavioural risk factors for stillbirth. The relevance

of the woman's partner, family and friends over behaviour change is well known. Social support as a BCT has been used in behaviour change interventions to support behaviour change in multiple contexts other than pregnancy showing potential to increase the intervention effectiveness[633–636]. The BCTs associated with social support that can be used together or in isolation are “Social support (unspecified)”, which is defined as “Advice on, arrange or provide social support (e.g. from friends, relatives, colleagues,' buddies' or staff) or non-contingent praise or reward for the performance of the behaviour. It includes encouragement and counselling, but only when it is directed at the behaviour"[637]; Social support (practical) which is defined as "Advice on, arrange or provide practical help (e.g. from friends, relatives, colleagues, 'buddies' or staff) for the performance of the behaviour"[637]; and Social Support (emotional) which is defined as “Advice on, arrange, or provide emotional social support (e.g. from friends, relatives, colleagues, 'buddies' or staff) for the performance of the behaviour "[637]. Chapter 8 showed that the BCTs related to Social Support were present in seven out of the nine interventions included, highlighting the importance that researchers have given to social support as an element of their interventions. Furthermore, given the evidence on the influence of a person’s close social network on their behaviour, the potential impact of dyadic interventions to promote behaviour change (e.g., physical activity) has been described in the literature[3]. The evidence regarding the effectiveness of dyad-based behaviour change interventions in aspects such as physical activity is sparse and conflicting, however, and thus, more research is needed, particularly in the context of stillbirth related behaviours[3,638].

The sociodemographic characteristics of a woman can also act as a barrier or a facilitator for her to engage in behaviour change. The results show that women from disadvantaged backgrounds without support from their social context had to prioritise other more pressing demands over their own care (e.g., taking care of older children, working extra hours, etc.) (Chapters 4-6). These findings are also supported by the literature, where sociodemographic factors such as older age, higher level of maternal education, higher level of income, employment status, and private health insurance were predictors of a higher likelihood of smoking cessation during

pregnancy[639]. Similarly, Brown et al. (2019) found in their review that maternal age was associated with alcohol, smoking and cannabis use; whereas lone-parent households had increased odds of all substance use[640].

9.3 Contribution of this research and implications for policy and practice

This research has developed the evidence base necessary to comprehensively understand the modifiable behavioural risk factors associated with an increased risk of stillbirth. This evidence base informs the potential future development of a behaviour change intervention to prevent stillbirth. The implications of the overall Thesis findings for policy and practice are discussed in this section, and they complement the implications raised in the discussion sections of the individual study chapters.

9.3.1 Implications for policy

Improving education and information sources

All chapters in this thesis highlighted issues related to information provision and educational sources. The importance of information and education over risk perception and behaviour change has been discussed in this Thesis (Chapters 2 – 8). However, the lack of adequate official informational resources and lack of discussion during antenatal care regarding modifiable behavioural risk factors for stillbirth and health habits were also identified as problematic (Chapters 2 – 8). These findings highlight the importance of creating consistent, reliable, and evidence-based information sources for women and HCPs around behavioural stillbirth risk factors and their prevention. Since the publication of the website content analysis study reported in Chapter 3, the Health Service Executive (HSE) launched an independent website providing information about a range of pregnancy and child-related aspects entitled “mychild.ie”[641]. This website (mychild.ie) was not included in the websites analysed in Chapter 3 because it was launched after finalising data collection for such study. After the development of the "mychild.ie" website, members of the Pregnancy Loss Research Group (PLRG) linked with the HSE to collaborate on enhancing the information provided concerning pregnancy loss. During the process of reviewing the website to provide feedback to the HSE working group managing it, some limitations

related to modifiable behavioural risk factors were identified. For instance, the information on modifiable behavioural risk factors for stillbirth was "unlinked" from the potential pregnancy outcomes including pregnancy loss. This lack of a link between behavioural risk factors and potential adverse outcomes such as stillbirth was also found in the women participating in the qualitative study (Chapter 7). This work is still in progress although no changes have been made to date, however, it is an example of how improvements can be made and provide a good example of how research findings such as those presented in this Thesis can inform policy.

Pregnancy-specific supports

Currently, the HSE already avails of structures in place to address some of the mentioned modifiable behavioural risk factors for stillbirth. For example, to support the cessation of smoking during pregnancy, the HSE provides women with a series of smoking cessation services called QUIT[642]. However, most of those services are not pregnancy-specific. Regarding alcohol consumption, the HSE website provides advice as to how to reduce it and a telephone helpline for support[643]. Some maternity hospitals avail of Drug Liaison Midwives specialised in supporting women with drug addictions throughout pregnancy[644]. In terms of weight management, some MHCPs offer women with overweight or obesity an appointment with a hospital-based dietician. Availing of these services in the Irish maternity health system for women is very positive, however, the findings highlighted that there are many barriers and facilitators affecting women's access to these types of services during pregnancy. Some of the main reasons identified in the studies that interfere with women's access to services were fear of the consequences or stigma, holding misconceptions altering their risk perception, and lack of awareness of the available services due to the lack of discussion about risk factors during antenatal care (Chapters 4-6). Further, the findings from the survey study also revealed that HCPs tended to consider supporting women with behaviour change during pregnancy as part of their role, which might reflect a lack of awareness of the potential support services. The development of services like the ones mentioned above needs to be informed by research with patient involvement to ensure that these barriers are addressed and

that services meet the specific needs of the different women attending antenatal care.

Pre-conceptual education

Health literacy issues and awareness about risks and recommendations around the behavioural risk factors for stillbirth are problems that were identified in 4 of the studies in this thesis (Chapters 4-7). One way to address awareness around health habits during pregnancy, as well as reproductive and health literacy, is by enhancing pre-conceptual education from an early age. As shown in the findings of the survey study (Supplementary file 1.1) and the qualitative study (Chapter 7), some topics are perceived by HCPs as more difficult to discuss with women, such as weight-related risks. These findings were also reinforced by some of the comments of the women in the qualitative study reflecting how some topics like stillbirth are societal taboos. These attitudes towards certain topics show how sociocultural aspects have an influence on education and information provision and consequently, on health and reproductive health literacy.

Pre-conceptual health is especially relevant to promote healthy maternal and pregnancy outcomes[645]. However, pre-conceptual interventions usually require that individuals who are not yet thinking about becoming pregnant engage with them; and these individuals are unlikely to engage with the maternal health services[646]. The pregnancy period is short and potentially full of challenges that can interfere with behaviour change as shown in the findings such as fatigue, nausea, tiredness and stress (Chapters 4-7). Hence, supporting women and men to arrive at that stage of their lives in their best health status possible is crucial to minimise the need to make changes during that period and reduce risks of adverse pregnancy outcomes[646]. Previous research has found that women with higher levels of education and who planned their pregnancies were more likely to engage in behaviour change by improving their diet and reporting less smoking and alcohol use in the pre-conceptual period (one year before conception)[646]. Some studies have shown that pregnancy planning is common, however preconception health and care is low, which means that an important opportunity for intervention has been missed[646]. Most of the factors related to stillbirth risk discussed in this thesis (e.g., substance use, lack of

engagement with healthcare and weight-related risks) are not only risk factors for adverse pregnancy outcomes such as stillbirth but for many other adverse health outcomes[647,648]. Hence, programmes focusing on enhancing health literacy and health promotion in general from early stages in educational settings would not only contribute to better pregnancy outcomes, but to the better health status of society[648].

Community services

Utilising community resources to support behaviour change could be another approach that might help unburden the health system. There are several public health services provided by the community in Ireland, including public health nurses who are based in HSE local health centres. Public health nurses visit newborn infants and their mothers at home within the first 6 weeks postpartum and also provide services to expectant mothers[649]. For women who deliver their babies in the hospital, the role of public health nurses is mostly focused on the postpartum period[649]. Availing of this service in the community for antenatal care as well has been utilised in other contexts to address some concerns related to hospital-based antenatal care such as lack of continuity of caregiver, prolonged waiting times or inaccessibility[650]. Community-based antenatal and postpartum care for low-risk women has proved to have benefits in terms of accessibility and convenience[650] and might be also a solution to unburden the health services when addressing behavioural change during pregnancy. However, staff shortages are also affecting the development of maternity services in Ireland[651]. In the latest months after the slow re-opening of society after the COVID-19 pandemic-related lockdowns, the HSE decided to modify the public health nurse (PNH) system to a "prioritisation" system, meaning that only babies with proven medical risk receive care by the PHN, and instead, parents are receiving written information about child development[652]. This means that many babies in Ireland are missing their key milestones checks during their first year of life, which is concerning as home visits by PHN facilitate the identification of problems that require early detection and act as support for parents' wellbeing[652]. These limitations within the public health nurse services mean that an opportunity to develop community services and to support women during pregnancy to engage in

behaviour change, when necessary, is missed, interfering with possible stillbirth prevention efforts.

Development of a care bundle to reduce the risk of stillbirth

As discussed in Chapter 1, while some initiatives have taken place in Ireland to tackle the rates of stillbirth, much more can be done. The results of this thesis provide a comprehensive evidence base that can be used to inform policy decisions and the development of public health approaches and intervention designs to raise awareness about stillbirth and the modifiable behavioural risk factors for stillbirth. The evidence built from the studies in this Thesis, together with the learning obtained from the initiatives taken internationally can inform the future development of a care bundle to reduce the risk of stillbirth in Ireland. Section 9.6 discusses the proposed next steps to continuing the development of this project.

9.3.2 Implications for practice

Clinical guidelines to support HCPs in providing care

The findings from this Thesis have highlighted multiple factors that influence women's behaviour during pregnancy, and how these are observed at different levels from the woman individually to the social context. Hence, it is especially relevant that antenatal clinical guidelines provide guidance and encourage HCPs to assess and explore women's experiences during antenatal care at different socioecological levels. Further, this assessment should be continuous throughout women's care, as women's situations can change over time.

The HSE has produced guidelines for practice concerning the modifiable behavioural risk factors for stillbirth, although these guidelines do not have the explicit purpose of reducing stillbirth. The Obesity and Pregnancy guideline (published 2011)[653] recommends HCPs conduct care and communication sensitively and respectfully, advise women to lose weight before becoming pregnant, and calculate women's BMI at booking[653]. The guideline also recommends that women with overweight or obesity are referred to a dietician and that physical activity is encouraged[653]. However, the guidance provided in terms of behaviour change is general and does not provide HCPs with tools to support women[653]. The guideline also does not

explore the possibility of assessing the women's individual situation at deeper levels to be able to identify her specific barriers to, in this case, managing their weight during pregnancy or in the pre-conceptual period. Comprehensive guidelines that provide specific advice to HCPs to support women, as well as encourage and provide tools for HCPS to explore women's situation at every level of influence over her behaviour are necessary and should be developed.

The HSE has also produced a guideline on "Alcohol consumption in pregnancy" (2018)[654], this guideline recommends that women should be advised that heavy drinking during pregnancy "may be harmful to the baby"[654], the guideline also states that there is no evidence of harm regarding low levels of alcohol consumption. This guideline discusses the evidence regarding alcohol drinking and some potential outcomes (e.g., miscarriage, stillbirth, Fetal Alcohol Syndrome), but does not provide specific advice as to how to support women who consume alcohol during pregnancy[654]. The Department of Health National Clinical Guideline No. 28 "Stop Smoking" (2022) highlights pregnancy as one of their prioritised groups[655]. This guideline recommends that all HCPS ask pregnant women for and document smoking behaviour of pregnant women, which should be updated regularly throughout the pregnancy[655]. The guideline also recommends that all women should be provided with information about the risks of smoking and second-hand smoking, and to support women to make an informed decision regarding their stop-smoking plan. The guideline also acknowledges the socioeconomic inequalities associated with smoking behaviour during pregnancy[655].

Currently, a HSE guideline focused on the risk factors, diagnosis, investigations, management, classification, audit and review, follow-up care, and future research priorities for stillbirth is being developed and will be published in 2023[257]. This guideline will also include recommendations related to the modifiable behavioural risk factors for stillbirth that were exposed in section 1.7.5.

Prioritisation of health promotion during antenatal care

The findings of the meta-synthesis of qualitative research (Chapters 4-6) work and the qualitative study (Chapter 7) demonstrate that women perceive antenatal care to be focused on prioritising medical needs. In a context where resources and staff

are limited, it is crucial to prioritise factors that might have a direct influence on the pregnancy outcome and provide therapeutic measures as soon as possible. However, dedicating time to discussing health habits is also relevant to enhance pregnancy outcomes for women[656]. Having discussions around health habits might not only have an impact on stillbirth rates by increasing health literacy and adjusting risk perception (Chapters 4-6) but also on many other adverse health and pregnancy outcomes associated with the modifiable behavioural risk factors that have been discussed in this thesis (i.e., substance use, lack of attendance at antenatal care, weight management risks)[657]. Hence, dedicating additional time during antenatal care to explore women's health habits and all their influencing factors, not only during the booking visit but throughout pregnancy, would be beneficial. However, it is well documented that HCPs already experience time constraints during their regular working days[658] and can experience burnout syndrome[659]. The development of training programmes to enhance education about potential adverse outcomes during pregnancy, health habits, behaviour change and communication style might provide HCPs with the resources they need to provide the best care for women. Currently, the HSE offers HCPs a training programme named "Making Every Contact Count", which includes training on behaviour change (including behaviour change theory and techniques), six topic modules focused on smoking, alcohol and drugs, healthy eating, active living, overweight and obesity and mental health and wellbeing, and a module to practice the gained skills on how to carry a brief intervention with real-life scenarios[660]. The survey study in Supplementary file 1.1 provided evidence that HCPs consider it their responsibility to support women with behaviour change and to create and maintain healthy habits regardless of their specific discipline (midwives, doctors, nurses). However, time and training were identified as barriers to supporting women around these behaviours. Therefore, even though training HPCS is essential, utilising community services to support women with their health habits during pregnancy and exploiting settings where people attend pre-conceptually (General practitioners (GPs), schools, and universities) might be a better solution to provide behaviour change counselling.

Vallis et al. (2018) proposed a framework for behaviour change counselling in primary healthcare in the context of chronic disease management that HCPs can use[661]. The framework is based on four principles: (1) establishing a change-based relationship, which requires building an empowering and non-judgemental bond with the patient[661]; (2) Getting to behaviour, clarifying and isolating specific behaviours, assessing readiness for change and addressing underlying health beliefs including the pros and cons of behaviour change[661]; (3) Behaviour modification, which includes setting behaviour change goals and shaping them when appropriate, identifying and encouraging reinforcement and controlling environmental stimulus that is shaping the behaviour[661]; and (4) addressing the psychosocial determinants of behaviour, which encourages maintaining a holistic view[661]. This framework appears compatible with most of the barriers and facilitators for behaviour change during pregnancy that has been identified in the studies in this Thesis, since it takes into account the importance of interpersonal factors, relationships and social context. The framework considers aspects identified in Chapters 4-6 such as misconceptions about risks and value of care, social stigmatisation and judgemental attitudes from HCPs, lack of awareness of the behaviour and social influences of the close social network and the wider social context (Chapters 4-6). Some of the limitations observed in this framework however are concerning the high number of resources needed to implement it. The authors report that for the successful implementation of a programme based on the framework, aspects such as managerial support, resources, a team culture that embraces behaviour change, behaviour change peer leaders, supervision, mentorship, monitoring and supporting of ongoing learning are necessary[661]. The development of a future framework such as the one outlined above to aid HCPs in supporting women with behaviour change would be beneficial, as long as they are designed taking into consideration that maternity services are under time and workforce constraints.

9.4 Strengths and limitations of this research

One of the main strengths of this work is that it provides a comprehensive exploration of the issues related to modifiable behavioural risk factors for stillbirth. The studies that make up this Thesis form a much-needed evidence base tackling a research area that has been neglected so far, namely behavioural factors related to stillbirth risk. This evidence-based can also inform the development of future preventive approaches to reducing stillbirth rates in Ireland. Further, the research was conducted as informed by the BCW[259], in particular the understanding of the behaviour stage, which will allow for further development in future work.

To build the evidence base that enhances the understanding of the modifiable behavioural risk factors associated with stillbirth, different sources of information were used. This is a strength of this thesis as it allowed the incorporation of findings from different contexts and populations, as well as exploring more in-depth the specific context in which the research was being conducted. Building this evidence base with such a variety of information sources helped us to address two research gaps: enhancing the understanding of the modifiable behavioural risk factors for stillbirth, and informing the future development of behaviour change interventions to address them.

Different methodologies were utilised to conduct the studies included in this thesis including quantitative and qualitative methods, as well as the use of primary and secondary data. These decisions were made depending on the different research questions and the objectives of each study and allowed exploration of all the different aspects of the topic at different levels. Firstly, the literature review was helpful to identify which were the modifiable behavioural risk factors that had more evidence in relation to the risk of stillbirth, in order to focus the aim of the project. Conducting a systematic quantitative content analysis was useful to be able to quickly and effectively identify a problem, which was the lack of information about stillbirth and modifiable behavioural risk factors on Irish and UK websites. meta-synthesis of qualitative research are useful to enhance interpretations of an individual phenomenon by bringing together the findings from primary qualitative research[662]. The meta-synthesis of qualitative research process allows the researcher

to go beyond the individual study findings to produce greater interpretations[662]. This research method was appropriate to identify facilitators and barriers to behaviour change during pregnancy as it allowed the production of findings that were more generalisable to different contexts and populations (Chapters 4 - 6). Qualitative research allows researchers to learn from people's direct experiences, identifying relevant contextual and community factors that might be overlooked in quantitative designs but hold relevance for intervention development. Conducting a qualitative study (Chapter 7) with postpartum women gave information on the more specific nuances relevant to the hospital care women received in the hospital, which might inform the more context-specific aspects of the future intervention development. Lastly, conducting a systematic review of the literature (Chapter 8) to identify interventions ensured that relevant interventions were not missed and that learnings from those interventions available in the extant literature were obtained. Involving a multidisciplinary team to design and conduct the studies is a strength as it incorporates different types of expertise (e.g., medicine, behavioural science, epidemiology, and health psychology) and provides multiple learning opportunities. Further, being part of a research group with the involvement of patient representatives also provided an opportunity to incorporate informal stakeholder input when necessary.

However, this research also has some limitations. One of the main aspects is the limited research conducted with HCPs. Originally, it was planned to conduct a qualitative study to explore HCPs' experiences of supporting women with behaviour change. This plan had to change due to the COVID-19 pandemic. During this period of time, HCPs worked under restrictions and experienced an increased workload with additional healthcare management burdens, which meant that their time to participate in research was extremely limited. This is the reason why this study was re-designed to become an online survey study that was administered at a later date and led by a collaborator. However, the survey (Supplementary file 1.1) was designed with input from the meta-synthesis of qualitative research (Chapters 4-6) and the qualitative study with women (Chapter 7) which ensured that its contents were

relevant to the topic under study. Therefore, further research is necessary to incorporate HCPs input further and build on the findings of this research.

Similarly, I did not incorporate women with lived experiences of pregnancy loss in these studies. It was decided to conduct a qualitative study with postpartum women who had uncomplicated pregnancies and deliveries because the study aimed to capture the normal antenatal "day-to-day" practices of information provided regarding modifiable behavioural risk factors and stillbirths. However, it is well established in the literature that women who experience stillbirth have a higher risk of experiencing a subsequent stillbirth in their following pregnancy[92]. Hence, incorporating the voices of women with lived experiences of pregnancy loss into the development of prevention strategies and public health campaigns is crucial to assess their specific needs and anticipate potential impacts on their well-being. This is also the case for women from different minority groups. All women who participated in the qualitative study were white, heterosexual, working, well-educated, and partnered women. It is known from epidemiological data that women from different disadvantaged sociodemographic and minority ethnic groups have different rates of stillbirth[239,663], which means that, potentially, the people who could benefit the most from an intervention to reduce the risk of stillbirth are not reflected in the findings of our qualitative study (Chapter 7). However, some of these limitations might be mitigated by the diversity of women participating in the reviewed studies and the findings obtained from the meta-synthesis of qualitative research. Therefore, there is scope for further research to explore if these findings are generalisable to other populations and to assess the potential specific needs of these populations. Even though this is a limitation to the research in this thesis, recruiting diverse groups for pregnancy-related research is a challenge across groups and study foci[664,665]. Evidence of rationale for why some populations might be harder to recruit for research includes individuals not wanting to be identified (e.g., people with substance use disorders), people that belong to self-identifying groups (e.g., gender-diverse people), or being small numbers (e.g., indigenous populations)[665]. Once again, the stigma identified in Chapters 4-7 surrounding stillbirth and its behavioural modifiable risk factors might also be associated with increased difficulties to recruit

women. To overcome these challenges, three strategies were proposed by Bonevski et al. (2014) to recruit "hard-to-reach" pregnant women at high psychosocial risks[665]. These strategies include 1) Practitioner-led referral, by a health or social care practitioner, for example; 2) researcher-led referral, which involves the researchers attending clinics or community settings and directly explaining the study to potential participants; and 3) Self-referral, which might be precipitated by advertising material such a social media advertisement or poster placement in communities[665].

Given the high number of studies identified as relevant for the meta-synthesis of qualitative research, a criterion of quality was used to include studies in the synthesis. This means that only studies rated as high-quality were included in the analysis. It has been argued in the literature that adding low-quality studies into a research synthesis does not have a strong influence on the findings of such synthesis[383]. Deciding to reduce the number of studies included in the synthesis allowed more effective use of time and capacity while ensuring that the meta-synthesis of qualitative research was based on the highest quality evidence which would reflect in the quality of future intervention development. However, it is possible that by not including some studies based on a quality criterion, important insights might have been missed[666], so it must be acknowledged as a limitation. On the other hand, conducting the CER-Qual assessments on the individual review findings (See supplementary files 4.3, 5.6 and 6.3) ensures confidence in the findings[667].

The assessment of sources of information available in the different studies (Chapter 2 and Chapter 7) is also limited. In Chapter 2 Irish and UK websites were assessed to determine whether they contained information about stillbirth and behavioural risk factors or not, and in chapter 7 information sources women utilised during their pregnancies to educate themselves about health habits were explored. However, the quality of such information was not explored. This is very relevant as well to ensure that women are not receiving misleading or problematic information.

Lastly, another limitation of this research is that it focuses on high-income English-speaking settings only. This decision to focus the Thesis on these settings was made because the intention is that future strategies developed from this work will be

utilised in high-income settings. The inclusion of low- and middle-income countries might have influenced the applicability of the thesis findings, but middle- and low-income countries' challenges to reducing stillbirth rates are different[5]. However, as seen in Chapter 1, some of the most common risk factors associated with stillbirth are similar in low-, middle- income countries and high-income countries[2,45,46,48]. For example, issues related to maternal age and sociodemographic disadvantages, and lack of access to care are risk factors associated with low-, middle- and high-income countries. Thus, by making this decision, relevant learning points available in the literature might have been missed by not exploring those settings further. Despite the limitations of this Thesis described in this section, the decisions made throughout the process were made to ensure that resources were used most effectively and efficiently while maintaining their relevance with the research objectives of each study in this Thesis.

9.5 Implications for future research

Findings from the studies included in this Thesis and some of the limitations discussed in section 9.4 have highlighted some areas of interest that can be explored further through future research.

9.5.1 PPI involvement

To begin with, more research should be conducted involving women with a history of pregnancy loss, and women from different socioeconomic backgrounds. Given the sensitivity of the topic, including women with experiences of stillbirth in the design of future research in this area is crucial to make sure that no involuntary harm is produced at any stage to this already vulnerable population and that the needs and preferences of this group of women are addressed in research. The experiences of women who engage in risky behaviour have been explored in this Thesis using meta-synthesis. Involving stakeholders in research from the study design phase is relevant to ensure that the "relevance gap" is solved and that the research has an impact by making interventions better informed by the stakeholders' preferences and needs[668]. Further, involving HCPs and other relevant professional stakeholders in the development phases of a behaviour change intervention and its implementation

strategy might facilitate its future implementation by incorporating input related to the feasibility of such intervention.

In this way, public and patient involvement (PPI) is understood as the active involvement of the public, patients, health professionals and other decision-makers throughout a research process or an intervention destined, can be used as a strategy to enhance the impact of such research or interventions by making them more relevant to users needs[669]. Further, it can also increase the likelihood of the intervention being implemented more effectively[669]. The process of involving PPI representatives in research or intervention development requires careful pre-planning, training for PPI representatives and researchers, and working on building a trusting working relationships[670]. Additionally, it has been reported in the literature that adequate funding and time, planning and procedures put in place, clear definition of roles, positive attitude from researchers towards PPI, and trust and respect have a positive impact on both PPI and researchers[670]. Thus, interventions designed in the context of stillbirth prevention to tackle the modifiable behavioural risk factors for stillbirth should incorporate PPI representatives and stakeholder involvement to ensure they are sensible and relevant for women.

Even though stakeholder involvement in intervention design has benefits and its importance has been highlighted in the literature[671], there is no clear evidence as to how best to involve PPI representatives in research or intervention design[672]. The Department of Health in Ireland published a Framework for Public Involvement in Clinical Effectiveness Processes in 2018 that might have the potential to be utilised for intervention development as well[673]. This framework proposes five stages to consider for public involvement that vary depending on the level of involvement of the public[673] and might be of use to incorporate PPIs into the future development stages of this project.

Some authors have described their process of involving PPI in pregnancy-related research. For example, Hennessy et al. (2022) described in their paper an approach to involve PPI representatives in the development of guideline-based Key Performance Indicators (KPIs) for recurrent miscarriage[674]. This process involved a two-round Delphi process, four consensus meetings, a final survey and a final review

meeting[674]. Some of the benefits of this process described by the authors are related to the richness found in the diversity of their group, and some aspects that facilitated accessibility according to the PPI representatives were having skilled facilitators, good communication with the research team, having virtual access to the meetings, and making the process user/friendly (by providing lay explanations, a reference guide with definitions of medical terms and time for discussions)[674]. In sensitive areas such as stillbirth, it is particularly important to pay attention to the well-being of PPI representatives when participating in research. Siasskos et al. (2015) described how involving a PPI representative, which in this case was a bereaved parent with experience in supporting other parents through their loss, in their stillbirth research ensured that their project met all the relevant ethical requirements for approval, including considerations about the sensitivity of the recruitment materials and the best timing and contact method to approach potential participants[674].

These examples in the literature as to how to best incorporate PPI representatives into pregnancy loss research, could be beneficial to inform the future steps of the development of this project, which will be discussed in section 9.5.4.

9.5.2 Quality and availability of sources of information

Another area of future research is related to the additional exploration of sources of information for women about stillbirth and modifiable behavioural risk factors. In this thesis, I established that the websites available at the time of the study in Ireland and the UK did not provide sufficient information about stillbirth and risk factors. This leads to proposals that more online resources could be created; however, it is very important to also continue to monitor and explore the quality, acceptability and reach of such resources. Additionally, other sources of information online such as social media platforms or forums could also be explored in terms of quantity and quality of information as a potential new resource that could be utilised to educate women during their pregnancies. A participatory design methodology could be beneficial in the creation of new online resources, where users (in this case, pregnant women or women with experience of pregnancy loss) would be involved in the design of the online resources. This approach has been utilised before to test the

usability of online resources, as well as to culturally adapt and translate content, as well as to collect feedback on how to improve online resources[675].

In terms of the future assessment of online resources, different tools are proposed to assess the quality of online resources. Beaunoyer et al. (2017) conducted a systematic review of popular tools used in the assessment of readability, emotional content, understandability and usability of online health information[675]. According to the authors' findings, these parameters are assessed by different tools. The authors discuss eighteen different tools that address these different aspects, however, these tools have some limitations[675.] Firstly, the tools have been tested with online materials covering a limited amount of health issues, limiting their generalisation[675]. Secondly, there are a limited number of studies analysing the comparability of the tools, and the few available have concluded that these tools are not comparable[675]. Thirdly, some elements such as using active voice or visual cues might determine whether a website gets a high score in some of these tools, however, it might not be perceived as beneficial for a real user (e.g., patient or HCPs) [675]. Thus, the authors suggest that a mixed method approach using analytical scales that generate normative scores complemented by data obtained using qualitative research tools might be the best approach to assess online resources' quality[675]. This approach including analytical scales complemented by a qualitative approach (e.g., focus groups with bereaved parents) could be utilised to develop appropriate online resources to provide information about stillbirth and its modifiable behavioural risk factors in a sensitive way, while also ensuring good accessibility and readability of the contents.

Furthermore, it is not only relevant to have accessible sources of information, the messaging on those sources also needs to be considered with regard to the literature in risk messaging to ensure that it can work as effectively as possible. Personalisation is a widely used strategy in key messaging that involves tailoring the message to the individual's characteristics or behaviour[676]. Previous studies have demonstrated that personalised risk messages are more effective than non-personalised risk messages. McDaniel et al. 2020 found in their systematic review that personalisation in health communication can improve several outcomes relevant for behaviour change such

as knowledge, attitudes, beliefs, intentions and behaviours[677], and other studies have also found that personalisation increases engagement with the risk messaging[678].

Framing is another important strategy in risk messaging; this involves emphasising either the positives or negatives aspects of the risk message ("gain" vs. "loss" frames). Previous research has shown that the effectiveness of framing depends on the type of behaviour promoted (e.g., gain-framed messages work better for proactive behaviours which are taken to prevent negative outcomes such as getting a flu vaccine, and loss-framed messages work better for reactive behaviours which are actions taken in response to existing problems such as taking medication to treat a condition)[679].

Other relevant aspects that might influence risk messaging effectiveness are the perceived trust in the source of the message, as it can impact the credibility of the message[680]; emotional appeals which can be used to increase the perceived severity of the risk[681]; and the importance of cultural and linguistic considerations, as previous research has shown that messages that are culturally and linguistically appropriate are more likely to be understood and adopted by their target audience[682]. Behavioural risk messaging in regards to stillbirth prevention can be very sensitive, and hence, it is important that future strategies take all of these potential strategies into consideration when creating new information sources or public health messages. Risk messaging regarding stillbirth needs to be tailored, culturally appropriate and timely.

9.5.3 Qualitative research in the area of sleep

When conducting searches for the meta-synthesis of qualitative research work, a search focused on sleep position research was also conducted at two different times. Those searches did not reveal any relevant qualitative research. Thus, one further area of research is related to conducting qualitative research on the facilitators and barriers to modifying women's sleep position during pregnancy, to identify potential elements that could complement the findings of the other three meta-synthesis of qualitative research related to antenatal care attendance, substance use and weight management behaviours.

9.5.4 Next steps in intervention development

The evidence base developed from this research has the potential to inform the future development of a behaviour change intervention to prevent stillbirth. In this section, we will describe how this process could be conducted.

During the future process of intervention design, we hope to include PPI representatives and diverse stakeholder groups throughout. With the creation of a stakeholder group, it is hoped to address some of the limitations of this research such as the limited involvement of women with lived experiences of pregnancy loss and HCPs. The PPI representatives and stakeholder groups should then involve HCPs, women with lived experience of loss, and women from different sociodemographic backgrounds, as well as women who have used the currently available support for behaviour change.

The future development of behaviour change interventions to tackle the modifiable behavioural risk factors for stillbirth could utilise the BCW. The BWC is a methodology that allows researchers to systematically design behaviour change interventions underpinned by the COM-B model of behaviour change[259] (See Introduction section 1.9.3). To systematically develop this intervention in the future, the different Stages and Steps of the BCW[259] will be followed (See Table 1.5.).

Table 1.5: Stages and steps composing the Behaviour Change Wheel systematic process for intervention design.

Stage 1: understanding the target behaviour	Step 1: define the problem in behavioural terms.
	Step 2: Select the target behaviour.
	Step 3: Specify the target behaviour.
	Step 4: identify what needs to change.
Stage 2: Identify intervention options	Step 5: Identify intervention functions.
	Step 6: Identify policy categories.
Stage 3: Identify content and implementation options	Step 7: Identify behaviour change techniques (BCTs)
	Step 8: Identify the mode of delivery.

In this sub-section, the contribution of the different studies in informing the different steps of the BCW[259] will be discussed (See table 9.2). This is a proposed approach that can be taken to progress this research and will be described in relation to the different stages and steps of the BCW.

Stage 1 of the BCW will be informed by all chapters (Chapters 2 – 8). Chapter 2, the literature review of behavioural risk factors for stillbirth, allowed us to define the problem of stillbirth prevention in behavioural terms (Stage 1, Step 1), and it was also helpful to select the target behaviours (Stage 1, Step 2) by focusing on those behaviours with the strongest evidence concerning the risk of stillbirth. In this case, the relevant behaviours are substance use, attendance and engaging with antenatal care, weight management behaviours (diet, physical activity) and sleep position. It is important that any proposed approach simultaneously takes into consideration all of the different behaviours relevant to stillbirth, as interventions for these individual behaviours already exist. Chapter 3, the website quantitative analysis, feeds into stage 1 of the BCW as it allowed identification of one of the aspects that need to change (Step 4), which in this case is the use of the internet as a potential source of information. Further, it also feeds into Stage 3 step 8, as it informed how online resources have the potential to be utilised as a mode of delivery for a future intervention. Chapters 4-8, the meta-synthesis of qualitative research on facilitators and barriers for attendance at antenatal care, substance use and weight management behaviours, and the qualitative study with post-partum women also feed into stage 1 of the BCW[259]. The findings of these studies are useful to define the problem in behavioural terms and select the specific target behaviours, as part of Stage 1 of the BCW. As the behaviours we want to address are broad, utilising the specific aspects identified in the meta-synthesis of qualitative research for the different behaviours such as diet, physical activity, smoking, alcohol consumption, etc. will facilitate defining the target behaviours, particularly with the consideration of their relationship to stillbirth as informed by Chapter 2. Similarly, the findings from Chapter 8 allowed the identification of behaviours targeted to date in the context of stillbirth prevention. Most importantly, the meta-synthesis of qualitative research will/can inform step 4 of stage 1 to "Identify what needs to change". The facilitators

and barriers identified in the studies can be coded using the components of the COM-B framework to conduct a thorough behavioural analysis and prioritise aspects that need to be addressed in an intervention.

Following the identification of the most relevant elements of the COM-B model to be addressed, it is proposed to use the BCW[259] framework to identify and select intervention functions (Stage 2). Intervention functions are defined as “broad intervention components which describe how an intervention aims to change behaviour”[259]. The different intervention functions are mapped into the elements of the COM-B model which means they are designed to influence the different elements of the COM-B model and consequently produce behaviour change[259] (See supplementary file 1.2 for definitions). Hence, the findings of Stage 1 will inform Stage 2 by enabling the mapping of the identified behavioural components of the COM-B model into intervention functions. If multiple intervention functions are identified as relevant, the APEASE criteria will be used to prioritise the selection of the most affordable, practical, effective, acceptable, safe and equitable (APEASE).

Stage 3 of the BCW involves identifying BCTs (Step 7) and modes of delivery (Step 8). BCTs are defined as the "active, observable and replicable components" designed to produce behaviour change[302]. The findings from chapters 4-7 contain information about facilitators for behaviour change, and preference for intervention delivery that can be used to inform the steps of identifying BCTs and mode of delivery of the BCW process[259]. Chapter 8 also informs the steps related to the identification of BCTs and delivery methods, as it provides evidence on which BCTs have been used to date in the context of stillbirth prevention and multiple examples as to how to operationalise those BCTs that can be used to inform this process. To identify BCTs, the BCTTv1[637] will be used. Then, the relevant BCTs will be operationalised by translating them into a concrete application. The APEASE criteria and stakeholder input will also be utilised in this stage to prioritise the selection of BCTs. Then, the selected BCTs will be operationalised working with the PPI representatives and stakeholders and a cohesive intervention will be outlined based on this process.

Table 9. 2: Studies conducted presented in this thesis and relevant steps of the BCW.

Studies, sources of information	**Behaviour Change Wheel phase(s) and step(s) informed**
Chapter 2: Literature review of modifiable risk factors for stillbirth.	Stage 1: Steps 1, 2
Chapter 3: Websites content analysis	Stage 1: Step 4 Stage 3: Step 8
Chapters 4-6: Meta-synthesis of qualitative research of facilitators and barriers (substance use, weight management, attendance at antenatal care)	Stage 1: Steps 1, 2, 3, 4 Stage 2: Steps 5, 6 Stage 3: Steps 7, 8
Chapter 7: Qualitative interview study with postpartum women.	Stage 1: Steps 1, 2, 3, 4 Stage 2: Steps 5, 6 Stage 3: Steps 7, 8
Chapter 8: Systematic review of behaviour change interventions to prevent stillbirth.	Phase 1: Steps 3, 4 Phase 2: Steps 5, 6 Phase 3: Steps 7, 8

For an intervention to be effective, it must be implemented appropriately, and future research is needed to examine appropriate and optimal implementation approaches. The following steps will then involve the development of an implementation strategy for the designed intervention.

Implementation strategies are defined as "methods or techniques used to enhance the adoption, implementation, and sustainability of a clinical program or practice"[683], and typically involve "package approaches"[684]. The process of designing the intervention will determine who would be the best suited to administer the intervention (e.g., HCPs from maternity services, community workers, educational settings, or online resources). Hence, stakeholder input will continue to be crucial

during this process to ensure the intervention and its implementation strategy are acceptable.

In terms of evaluating the intervention and implementation approaches, one potential framework that could be utilised to assess the acceptability of the designed intervention is the Theoretical Framework of Acceptability[685]. Within this framework, Acceptability is defined as "a multi-faceted construct that reflects the extent to which people delivering or receiving a healthcare intervention consider it to be appropriate, based on anticipated or experiential cognitive and emotional responses to the intervention". The multi-faceted constructs theorised by the authors affecting acceptability are affective attitude (feelings about the intervention), burden (perceived amount of effort), ethicality (fit with individual's value system), intervention coherence (understanding of the intervention and how it works), opportunity costs (benefits, costs or profits), perceived effectiveness (likelihood to achieve its purpose), self-efficacy (confidence in performing the behaviour(s) required). Future, longer-term impacts could also look at the fidelity of intervention delivery, intervention receipt, and intervention effectiveness.

In summary, following this process could enable the definition of the behaviours that need to be addressed to reduce the rates of stillbirth, and the identification of intervention functions and BCTs relevant to this purpose. This process could also facilitate the identification of options to translate BCTs into actual intervention content. Involving PPIs and stakeholders and utilising the APEASE criteria throughout the process could enhance the chances of the intervention being acceptable and effective for its purpose of changing specific behaviours to prevent stillbirth, as well as identifying the best implementation strategies to ensure the intervention's acceptability and feasibility.

9.6 Conclusion

The overall aim of the research in this Thesis was to enhance understanding of the modifiable behavioural risk factors associated with stillbirth and to produce an evidence-base to inform the development of future stillbirth prevention strategies. Stillbirth is a devastating pregnancy outcome that affects families, healthcare professionals, health systems and the whole of the social context. In Ireland, further efforts are necessary to reduce the rates of stillbirth, which have been increasing in the last years[663].

Four overarching topics were identified from the findings of the different studies composing this Thesis. The findings show how issues related to health literacy and sources of information affect women's behaviour. Increasing health literacy through the creation of reliable, high-quality and user-friendly sources of information, improving reproductive and health education from school age, and encouraging discussion about health habits and behavioural risk factors for stillbirth are some of the proposed strategies to address this issue. However, limitations in resources such as time, training, and availability of HCPs need to be considered when creating policies to address these issues.

Limitations in resources are translated into issues affecting HCPs behaviour and consequent relationships with their patients by overburdening HCPs with too many tasks and forcing them to prioritise emergencies. Furthermore, limitations in resources also increase the effect of the health system's structural barriers such as lack of availability or flexibility in the services they provide. To address these limitations, it is necessary to produce specific, acceptable, and efficient tools and guidelines that HCPs could use to explore a woman’s situation considering all aspects affecting her behaviour, and quickly identifying which potential additional supports she might need (e.g., further education, referral to a smoking cessation service, referral to a dietician, personalised physical activity plan, etc.).

The influence of the social environment over stillbirth prevention efforts is also highlighted in the findings of this Thesis. The stigma associated with stillbirth, and with the behavioural risk factors for stillbirth, hinders prevention efforts by interfering in the communication between patients and HCPs. This might be either

because HCPs and patients avoid discussing “difficult topics” or because women choose to conceal their situation (e.g., substance use) or avoid seeking care (e.g., concealment of pregnancy, women with increased BMI). Often, the purpose of this concealment is to avoid stigmatising situations in healthcare settings or when trying to engage in behaviour change (e.g., joining a physical activity group). Raising awareness about stillbirth and the modifiable behavioural risk factors generally in society might help to break the silence around stillbirth and reduce its associated stigma, facilitating prevention efforts.

To conclude, this Thesis makes a significant contribution to the knowledge base on the modifiable behavioural risk factors for stillbirth and identified key areas for future work in practice, policy and research. Importantly, the findings provide the groundwork for the future development and/or adaptation of interventions to promote behaviour change during pregnancy to reduce women’s individual risk of stillbirth and help national efforts at stillbirth rate reduction.

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Reflexivity statement

One of my favourite things about qualitative research is that it acknowledges that the researcher's own background, feelings, opinions, perceptions, and attitudes will have an influence on their analysis and interpretation on the data, and that then, the findings of their qualitative research are a product of incorporating all those elements with the participants accounts.

Conducting this research has resulted to be a very rich learning experiences that has posed me various challenges along the way. Coming from a different country and never having had contact with the maternity services in Ireland, learning about is culture and functioning was one of the first things I tried to do and I continue to do so every day.

In my native languages, we do not even have a word to refer to stillbirth. The same word is used to refer to stillbirth, miscarriage and termination of pregnancy either for medical reasons or by choice. When I started working on this project, I had no knowledge about pregnancy beyond the basics from secondary school education, I had no knowledge at all about pregnancy loss, and I had no knowledge about the Irish maternity healthcare system. Working on this project alongside the PLRG group has helped me understand the different challenges associated with pregnancy and pregnancy loss, and it has taught me to grow a massive respect for all women who go through those experiences and for the healthcare professionals that care for them, as well as for the researchers that dedicate their lives to improve outcomes for women and babies. This growing respect and admiration, especially for women, has made me feel like this research has a purpose and has motivated me to perform at the best of my capacities, which I hope is reflected in this Thesis. Also, it makes me feel excited about the next steps to take in the future of this project.

My background is in Health Psychology. I think this has been really helpful to try and understand women's experiences of pregnancy through a holistic point of view, trying to keep into consideration all psychosocial aspects interfering in a woman's behaviour. Further, the different backgrounds of my supervisory team (obstetrics

and gynaecology, public health, health psychology, sociology and epidemiology) has made this experience much richer, contributing even more to my learning process.

Even though I feel like I have learnt so much about pregnancy and pregnancy loss, they are not experiences that I have lived myself, and as a researcher, I think it is very important to keep that in mind when conducting qualitative studies. Thus, when conducting the interview study on this Thesis, I tried my best to maintain to actively listen to the women a non-judgemental attitude. I hope to continue to learn and grow from working alongside women with lived experiences and healthcare professionals. I think that process would not only enrich me personally as a psychologist and researcher, but also personally.

Supplementary materials

Supplementary file 1.1 – Exploring healthcare professionals' experiences when communicating with pregnant women about stillbirth (Survey study)

Survey study protocol

Study title

Exploring healthcare professionals' experiences when communicating with pregnant women about stillbirth

Lead investigators / Core team

a) Professor Keelin O' Donoghue, Professor and Pregnancy Loss Research Group Lead, University College Cork (UCC), and Consultant Obstetrician and Gynaecologist, Cork University Maternity Hospital (CUMH) – Principal Investigator

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c) Ms. Tamara Escañuela Sánchez, PhD candidate, Pregnancy Loss Research Group, INFANT Centre, Department of Obstetrics and Gynaecology, Cork University Maternity Hospital

Background

Stillbirth is one of the worst outcomes that pregnant women can face. They represent a significant clinical issue, with 2.6 million stillbirths recorded each year all over the world[1]. Although rates of stillbirth for high-income countries are lower than in middle or low-income countries, 1 in 200 babies will be stillborn and this rate has remained steady since 2000[2]. According to the last report published by the National Perinatal Epidemiology Centre in 2020, the rate of stillbirth at >24 weeks gestation or a birthweight of >500 grammes was 3.9 per 1,000 births in Ireland[3].

The stillbirth series published in the Lancet in 2016 highlighted the importance of focusing future stillbirth prevention efforts in targeting specific causes, risk factors, and vulnerable groups in high-income countries[4]. In these countries, most of the stillbirths occur prior to labour and are associated with placental pathology[5]. However, previous research has shown that stillbirth is associated with a wide range

of risk factors, such as behavioural factors, maternal medical factors, factors associated with women's obstetric history, and pregnancy related disorders. By behavioural risk factors we understand all of those that consist on different habits, lifestyle or choices that women make and that have a modifiable nature. In the case of stillbirth, these risk factors include ethnic and socioeconomic status, maternal overweight and obesity, maternal age, inadequate antenatal care, sleep position, smoking, maternal alcohol intake, and illicit drug use[6–9]. In this work, a focus on modifiable maternal behaviours that have been associated with a higher risk of stillbirth is proposed. Briefly, these comprise smoking[10,11], alcohol use[12], drug use[13,14], overweight or obesity[15,16], level of antenatal care attendance[17] and sleeping position[18,19].

Although there is considerable evidence supporting the association between these risk factors and stillbirth, this information is not widely available to the public. Moreover, research has shown that stillbirth remains a taboo subject for society [20,21], thus parents need increased awareness and recognition. Identifying barriers and facilitators towards behaviour change and communication in pregnancy from the maternity services healthcare professionals might be beneficial to improve awareness of stillbirth and its modifiable risk factors. Therefore, the aim of this study is to explore how healthcare professionals tackle the issue of stillbirth and associated risk factors, and to gain understanding regarding perceived barriers towards addressing these issues with their patients.

Research aims and objective

The aim of the study is to gain an overall understanding of maternity healthcare professionals views, attitudes and opinions regarding stillbirth and stillbirth prevention.

Specific objectives:

1. Explore maternity healthcare professionals experience and knowledge regarding stillibirth and modifiable risk factors for stillbirth.
2. Explore healthcare professionals' common practices regarding information provision about stillbirth, risk factors and health advice.

3. Explore common barriers to information provision about stillbirth, risk factors and health advice.

An online survey (Qualtrix) and a physical survey will be used to fulfill these objectives. This data collection instrument has been built on previous studies within the research group to inform the development of a survey which will be administered through online platforms and physically. Within this survey we will explore the following themes:

- Healthcare provider knowledge about stillbirth and associated risk factors.
- Attitudes towards behaviour change during pregnancy, including HCP's perceived facilitators and barriers to providing support, advice and information about stillbirth and risk factors.

The survey comprises mainly multiple-choice questions and Likert scale type questions. Comment boxes will be used throughout the survey to facilitate participants to add any further information if necessary.

The online survey will be created using the Qualtrix platform and administered via email, support networks and direct private contact with potential participants. The paper survey (which will be exactly the same as the online survey) will be administered in the potential participants places of work in CUMH.

Eligibility criteria

Clinical Nurses and midwives, specialist nurses/midwives, SHOs, Registrars/Specialist Registrars and Consultant Obstetricians/Gynecologists from the HSE and private clinics will be invited to participate.

Participants can refuse to participate or withdraw from the study at any time without any repercussion. No identifiable factors that would link the participant will be present in the data.

The participants will be selected based on their competency to consent to the research and their understanding of the survey process. Competency to consent refers to current physical and mental health status and sufficient level of education to understand the consent form.

Data analysis

The online survey responses will be extracted into an SPSS database and the physical survey responses will be manually entered into the same SPSS database. A combination of descriptive and inferential statistics will be utilised to analyse the data using SPSS.

In the case of receiving responses in the open-ended comment boxes, these will be imported into qualitative data management system NVivo and analysed thematically.

Protocol Duration

This evaluation will take approximately 12 months from initiation to conclusion.

Location of research

The core research team is normally based in Cork University Maternity Hopistal. All data will be stored and secured on UCC servers which are accessible remotely through a two-step verification process when necessary. The survey will be administered online to maternity healthcare professionals across the Republic of Ireland, and physically in CUMH.

Potential risks and benefits

The potential benefits which are anticipated for the participant relate to the opportunity of expressing their opinions as healthcare professionals on topics relating to behavior change, stillbirth, and barriers to communication. Each individual will benefit from expressing their needs in order to improve the quality of interactions with their patients. Partaking in this study will also provide the participants with an opportunity to influence the development of new communication strategies and interventions related to stillbirth prevention.

There are no direct benefits to taking part in this survey. Participants will not receive any material compensation or reward for their participation.

There are no anticipated risks associated with the study. All details to participants will be kept confidential, and the findings from this study will be reported in a way

that ensures that participants can not be identified. Data will be stored securely on the UCC server in files accessible only to the core members of the research team only.

Before completing the online survey, potential participants will be provided with an information sheet to read, which outlines the study's purpose, what it involves, and risk and benefits to taking part. They will then be presented with a consent statement and will be invited to actively tick a box to opt in (or out) of the online survey.

In the case of the paper survey, potential participants will be provided with the same information sheet to read, they will then be asked to physically tick a box to opt in (or out) in order to continue filling up the survey.

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Manuscript in preparation

Exploring healthcare professionals' experiences when communicating with pregnant women about stillbirth

Emily Rutherford, Tamara Escañuela Sánchez, Keelin O'Donoghue (2022).

Background

Substance use, lack of attendance at antenatal care, maternal weight and sleeping position are modifiable risk factors for stillbirth. The aim of this study was to gain understanding of maternity healthcare professionals (HCP) views, attitudes and opinions regarding stillbirth and stillbirth prevention, with a view to informing the development of risk reduction strategies.

Methods

An online survey (Qualtrix) was administered to HCPs in a large tertiary hospital in Cork, Ireland, exploring HCP knowledge and attitudes regarding stillbirth and associated risk factors.

Results

While 87% (79/91) of respondents had cared for women after a stillbirth, only 50% of respondents correctly identified the Irish definition for Stillbirth. Modifiable risk factors perceived as most important to discuss with pregnant women were attendance at antenatal care (48%), followed by smoking (34.2%). Maternal weight was the risk factor that HCPs found hardest to discuss with women (70%), and was also perceived as the risk factor that pregnant women are most reluctant to discuss with HCPs, according to 59% of respondents. Time constraints were identified as a major barrier to both providing education and supporting behaviour change in pregnancy (37.5%). Overall 65.2% of HCPs considered informing women regarding health behaviours and stillbirth risks to be part of their role; however, only 56.4% felt confident and trained to do so.

Conclusion

Our findings show that HCPs face multiple challenges in discussing important risk factors for stillbirth. Prioritisation of HCP education and protected time to discuss

modifiable risk factors during antenatal care are key to enhancing stillbirth preventive efforts

Introduction

Stillbirth is one of the most devastating outcomes that families can experience. Worldwide, there are 2.6 million stillbirths every year[1]. In high-income countries, the rates of stillbirth are lower than in middle- or low-income countries, but they still represent 1 in every 200 births, a rate that has remained steady since the year 2000[2]. In Ireland, the rate of stillbirth in 2020 was 4.20 per 1000 births and accounted for 67.2% of all perinatal deaths[3]. According to the National Perinatal Epidemiology Centre perinatal mortality clinical audit data, since 2018, there has been an increase in perinatal mortality in Ireland (rate ratio, RR=1.18, 95%CI=1.01-1.37, p-value=0.032)[3].

Previous research has highlighted the importance of focusing prevention efforts on tackling the risk factors for stillbirth[4]. These risk factors include maternal and fetal medical factors, obstetric related factors, sociodemographic factors, and behavioural factors. Some of these risk factors are modifiable and can be tackled through behaviour change interventions. A focus of work to prevent stillbirth, therefore, is on the behavioural risk factors for stillbirth such as substance use (smoking, alcohol, illicit drug use)[5–9], antenatal care attendance[10], sleep position[11,12] and maternal weight-related risks[13,14].

Although there is considerable evidence supporting the association between these risk factors and stillbirth, this information is not easily available to women[15]. There is also a paucity of online resources to provide information about stillbirth and risk factors[16]. Stillbirth remains a taboo topic in society[17–19], which hinders preventive efforts and leaves bereaved parents feeling isolated[17–19]. Previous research has established that healthcare professionals encounter barriers to discuss topics that are perceived as sensitive with their patients, such as weight related risks or substance use[20–22]. However, misinformation and lack of education have been established multiple times as obstacles to behaviour change in pregnant women[23–25].

Given the clinical need to address the ongoing issue of stillbirth, evidence-based behaviour change interventions to tackle behavioural risk factors for stillbirth need to be designed. To ensure that these interventions are feasible and acceptable, healthcare professionals' input is essential. Identifying barriers to communicating with women about stillbirth and healthcare professionals is a first step towards that purpose.

Therefore, the aim of this study was to explore how healthcare professionals understand the issue of stillbirth and associated risk factors, and to gain understanding regarding perceived barriers towards addressing these issues with pregnant women.

Methods

Setting and Participant recruitment

Cork University Maternity Hospital (CUMH) is one of 19 maternity units in the Republic of Ireland and is situated within the south/south-west hospital group. The hospital delivered 7040 babies in 2020[26].

After piloting, the survey was built into the platform Qualtrics and was distributed through CUMH staff via e-mail and QR codes. Participants were recruited opportunistically in the course of clinical work and educational sessions.

All HCP currently working at CUMH were targeted to complete this online survey. HCP who chose to participate in the survey provided consent before accessing its content in the Qualtrics platform. Multiple reminders were sent via e-mail to the HCP official e-mail addresses. No incentives were offered for participating in this study. Partially completed surveys were submitted to the system and added to the analysis as we believed that they provided valuable data. Between February and May 2022, 92 HCP engaged with the survey, however, not all participants fully completed all questions of the survey.

Ethics approval to conduct this study was obtained from the Clinical Research Ethics Committee of the Cork Teaching Hospitals University College (Reference number ECM 4 (i) 6/7/2021 & ECM 3 (gg) 10/08/2021).

Data collection

An online survey built in Qualtrics was used as a data collection tool for this study. The survey was designed to explore HCP general knowledge about stillbirth. It also explored attitudes towards providing information to women about stillbirth and modifiable risk factors for stillbirth, as well as barriers to do so (See supplementary file 1.1).

The survey was composed of 3 sections:

- **Section 1. About your experience:** role, place of work, years of experience, involvement in the care of stillborn babies.
- **Section 2. Stillbirth and risk factors:** knowledge about definition, prevalence, risk factors.
- **Section 3. Behaviour change and communication with patients:** habits regarding discussing risk factors with patients, attitude towards discussing risk factors, perceived barriers.

Data analysis

Responses were exported from Qualtrics and imported into SPSS v25 for analysis. Open ended questions were coded according to a pre-established criteria previously discussed amongst the multidisciplinary team. Descriptive statistics and cross-sectional tabs were used to analyse the data.

Results

Characteristics of the sample

Overall, 92 healthcare professionals engaged with the survey, however, only 72 completed the survey fully. Regardless of the level of completion of the survey, we included all responses for analysis.

The characteristics of the sample are described in Table 1. Our sample was composed of a wide range of maternity healthcare professionals, with midwifery representing more than half of the sample. Over sixty percent of the sample had more than 10 years of experience working in maternity services. Eighty-five per cent of the sample had previously cared for a woman after stillbirth at some point, and fifty percent of the sample had done so when they had less than one year of experience working in the maternity services. More than 50% of the sample had experience diagnosing a stillbirth and had been present at the delivery of a stillborn baby, and 40% of the sample reported caring for parents after stillbirth several times a year.

Table 1. Characteristics and experience of the sample.

		n	**Valid %**
Professional role (n = 92)	Senior House Officer (SHO)	6	6.5
	Specialist Registrar (SpR)	14	15.2
	Consultant	15	16.3
	Midwife	44	47.8
	Specialist Midwife	7	7.6
	Midwifery Management	5	5.4
	Nurse	1	1.1
Place of work* (n = 92)	Outpatient clinic	41	44.6
	Antenatal wards	46	50
	Postnatal wards	52	56.5
	Labour ward	47	51.1
	Early pregnancy Unit	29	31.5
	Fetal assessment unit/ultrasound department	29	31.5
	Emergency room	36	39.1
	Outreach clinics (outside of main hospital)	16	17.4
Years working in maternity services (n = 92)	Less than a year	5	5.4
	1-3 years	18	19.6
	4-6 years	21	22.8
	10-15 years	18	19.6
	16-20 years	6	6.5
	>20 years	24	26.1

Professional experience related to stillbirth			
Involvement in caring for a woman after stillbirth (n = 91)	Yes	79	85.9
	No	12	13
Years of experience at first care of woman after stillbirth (n = 76)	Less than a year	38	50
	1-3 years	24	31.6
	4-6 years	6	7.9
	7-10 years	2	2.6
	10-15 years	3	3.9
	>20 years	3	3.9
Experience diagnosing a stillbirth (N= 90)	Yes	50	55.6
	No	40	44.4
Presence at delivery of stillborn baby (N= 90)	Yes	58	64.4
	No	32	35.6
Frequency of caring for parents after stillbirth (n = 91)	Never	13	14.3
	Around once a year	28	30.8
	Several times a year	36	39.6
	Around once a month	10	11
	Around once a week	4	4.4

*Categories are not exclusive

Knowledge of stillbirth definition and prevalence

Healthcare professionals were asked the definition of stillbirth. In this question, HCP had to choose between 4 different birthweights, 4 different gestational weights, and the connector between the two. As a reminder, stillbirth is currently defined in Ireland as "a child born weighing **500 grammes** or more **<u>or</u>** having a gestational age of **24 weeks** or more who shows no sign of life" [27].

The survey results showed that 50% (n = 38 of 76) HCP were able to identify all elements of the definition correctly. 13.2% of HCP (n = 10 of 76) were able to identify both the birthweight and the gestational age correctly but picked "and" as a connector instead of "or", and 9.2% (n = 7 of 76) picked no connector at all. These two scenarios could lead to exclusion of some cases when making a clinical decision. 14.5% (n = 11 of 76) were able to identify only the birth weight element correctly, and 9.2% (n = 7 of 76) were able to identify only the gestational age element correctly. 3.9% (n = 3 of 72) were not able to identify any element of the definition

correctly (See Table 2). Midwives represented 36.8% (n = 14 of 38) of the people that identified the definition correctly, followed by consultants (23.7%, n = 9 of 38).

Table 2. Knowledge of the definition of stillbirth

	N	Valid %
All elements identified incorrectly (birthweight, connector and gestational age)	3	3.9
All elements identified correctly	38	50
Birthweight only identified correctly	11	14.5
Gestational week only identified correctly	7	9.2
Birthweight and gestational age identified correctly but use of connector “AND”	10	13.2
Birthweight and gestational age identified correctly but no connector selected	10	9.2
Total	76	100

HCP were also asked how prevalent they estimate stillbirth to be in Ireland as a figure per 1000 births. Responses ranged from 0.4 to 300. We considered a response as valid if it ranged between 3.5 and 4.5. Of the 73 HCPs that responded to this question, only 15.1% (n = 11) gave a value between those two parameters.

Knowledge of risk factors for stillbirth

Healthcare professionals were asked to state which are the most common risk factors associated with the risk of stillbirth. The most common risk factors cited were hypertensive disorders (60%, n = 45), diabetes (54.7%, n = 41), bacterial and viral infections (44%, n = 33), placental abruption (43.4%, n = 33), fetal growth restriction (40.8%, n = 31), maternal age (39.5% , n = 30), and placental insufficiency (35.5%, n = 27) (See Figure 1).

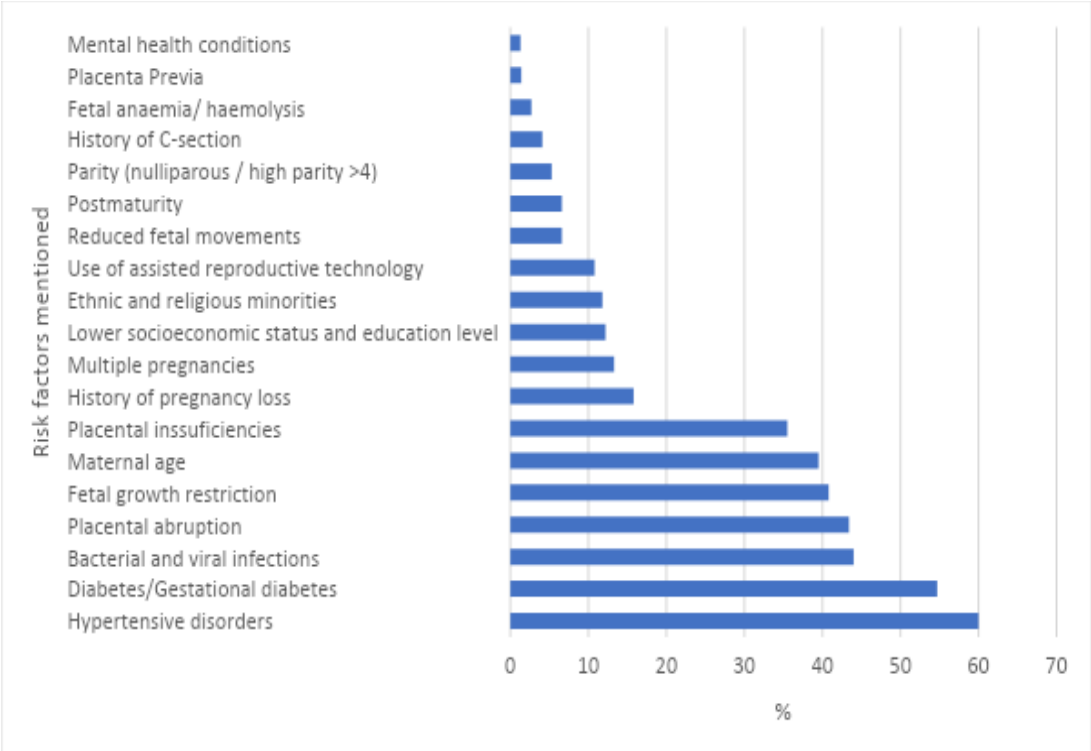


Figure 1. Risk factors mentioned by HCPs.

HCPs were also asked regarding their perception of the most common modifiable risk factors associated with the risk of stillbirth. The most commonly modifiable risk factors mentioned were maternal weight (52.2%, n = 48) and smoking (50%, n = 46) (See Figure 2).

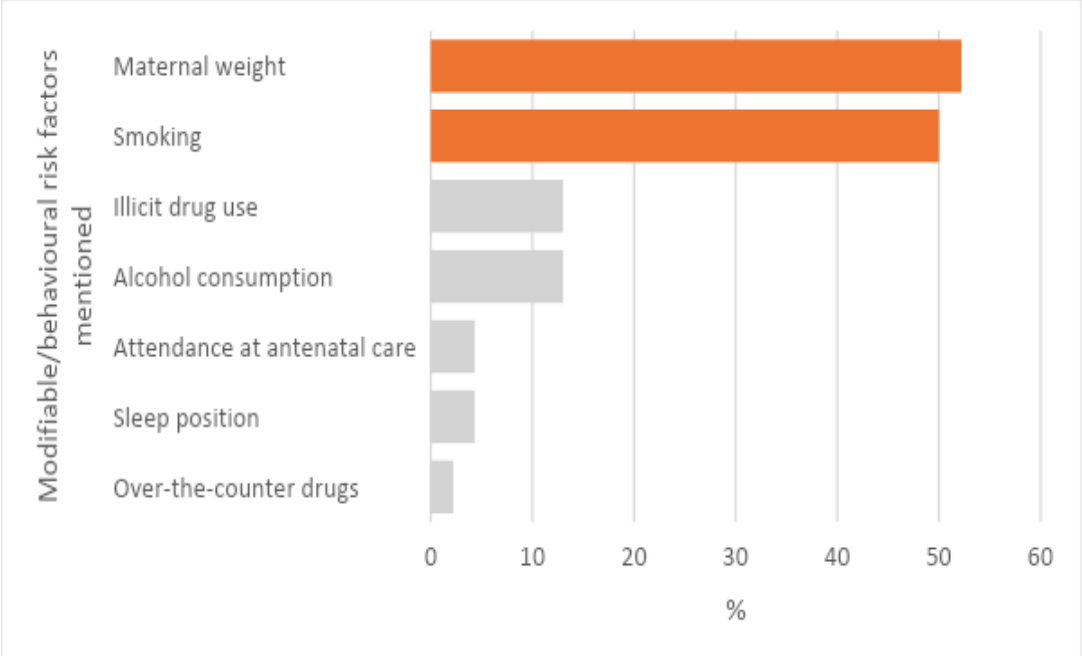


Figure 2. Most commonly mentioned modifiable/behavioural risk factors.

Training

Our survey explored the sources of information and training that HCPs had used to learn about stillbirth and risk factors for stillbirth. Over 89% of the sample reported having learned about stillbirth and associated risk factors in postgraduate training and through professional guidelines. 61% of the sample reported learning about stillbirth in undergraduate training (See Table 3).

Our results show that HCPs are more likely to attend educational events locally. 48% of our sample reported having attended a hospital-based study day on stillbirth, whereas only 14% had ever attended an international conference (See Table 3).

Table 3. Training and resources

		N	Valid %
Sources of knowledge about stillbirth	Undergraduate teaching	52	61.2
	Hospital based education	60	65.2
	Postgraduate training	33	89.2
	Professional guidelines	33	97.1
	Journal articles	24	26.1
Attendance at educational events on stillbirth	Hospital based study day	45	48.9
	National conference	22	23.9
	International conference	13	14.1

Communication with patients

The survey also explored healthcare professionals giving advice about risks factors and health habits to women. Statements were designed to differentiate HCPs' communication behaviour with all women and communication behaviour with women with risk factors.

90.3% of HCP (n = 65 of 72) reported that they provide tailored advice to all women in their care always or often. Regarding substance use, less than half of the sample (40.3%, n = 29 of 72) reported discussing smoking risks with all women under their care often or always, regardless of their smoking status. Only 29.1% of HCP (n = 21 of 72) reported discussing alcohol risks with all women in their care, regardless of their drinking status. Only 15.2% of HCP (n = 11 of 72) reported discussing illicit drug use with all women in their care often or always, regardless of their drug use status. 56% (n = 40 of 71) reported discussing substance use risks always or often with women who disclose substance use. 21% (n = 15 of 71) never or rarely discuss substance use with women who disclose substance use behaviours.

Regarding weight, only 23% of HCP (n = 17 of 72) reported discussing weight related risks with all their patients always or often. 9.8% of HCP (n = 9 of 72) reported

providing weight management advice to all women always or often. Almost half of the sample (47%, n = 34 of 72) stated that they focus weight management advice to women who are overweight or obese always or often.

84.7% of the HCP (n = 61 of 72) reported advising women in their care to attend and engage with all their antenatal care visits often or always, and 83% (n = 60 of 72) reported recommending all women to seek unscheduled care in case of concerns often or always. 97% of HCP (n = 70 of 72) reported providing advice regarding monitoring fetal movements to all women in their care.

Regarding sleeping position, only 50% (n = 36) of HCP reported providing advice to women about the safest sleep position during pregnancy always or often.

Healthcare professionals were asked to rank the main behavioural risk factors (smoking, alcohol, illicit drug use, weight related risks, attendance at antenatal care and sleep position) on a scale of 1 to 6 where 1 was the topic perceived as most important to discuss and 6 is the topic perceived as least important to discuss. Attendance at antenatal care (47.9%, n = 35 of 73) was the risk factor that HCPs considered most important to discuss, followed by smoking risks (34.2%, n = 25 of 73). Sleep position was the risk factor considered least important to discuss according to 47.9% of (n = 34 of 71) HCPs (see Table 4).

Table 4. Risk factors prioritised from most important to discuss (1) to least important to discuss (6) as perceived by HCPs.

	Smoking (n = 73)	Alcohol (n = 72)	Illicit Drugs (n = 72)	Weight related risks (n = 72)	Attendance at antenatal care (n = 73)	Sleeping position (n = 71)
	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)
1	34.2 (25)	0	11.1 (8)	2.8 (2)	47.9 (35)	4.2 (3)
2	32.9 (24)	13.9 (10)	13.9 (10)	22.2 (16)	12.3 (9)	5.6 (4)
3	20.5 (15)	33.3 (24)	15.3 (11)	9.7 (7)	13.7 (10)	8.5 (6)
4	8.2 (6)	29.2 (21)	16.7 (12)	25.0 (18)	8.2 (6)	12.7 (9)
5	1.4 (1)	18.1 (13)	20.8 (15)	27.8 (20)	11.0 (8)	21.1 (15)
6	2.7 (2)	5.6 (6)	22.2 (16)	12.5 (9)	6.8 (5)	47.9 (34)

Further, HCPs were asked to rank the main behavioural risk factors (smoking, alcohol, illicit drug use, weight related risks, attendance at antenatal care and sleep position) on a scale of 1 to 6 (where 1 was the topic perceived as most difficult to discuss and 6 is the topic perceived as least difficult to discuss). HCPs were asked to rank these factors in order of what they personally perceived as difficult for them to discuss, but also what they perceived as most and least difficult to discuss for their patients. The majority of HCPs (70.5%, n = 43 of 61) consider weight related risks as the most difficult topic to discuss, followed by illicit drug use (24.2%, n = 15 of 62). Similarly, 59.7% (n = 40 of 67) of HCPs considered that women under their care find weight related risks, followed by drug use (22.7%, n = 15 of 66) most difficult or are most reluctant to discuss. The topic perceived as easiest to discuss by HCPs was attendance at antenatal care (50. 9%, n = 29), and for women (as perceived by HCPs) the easiest topic to discuss was sleeping position (73.7%, 42 of 57).

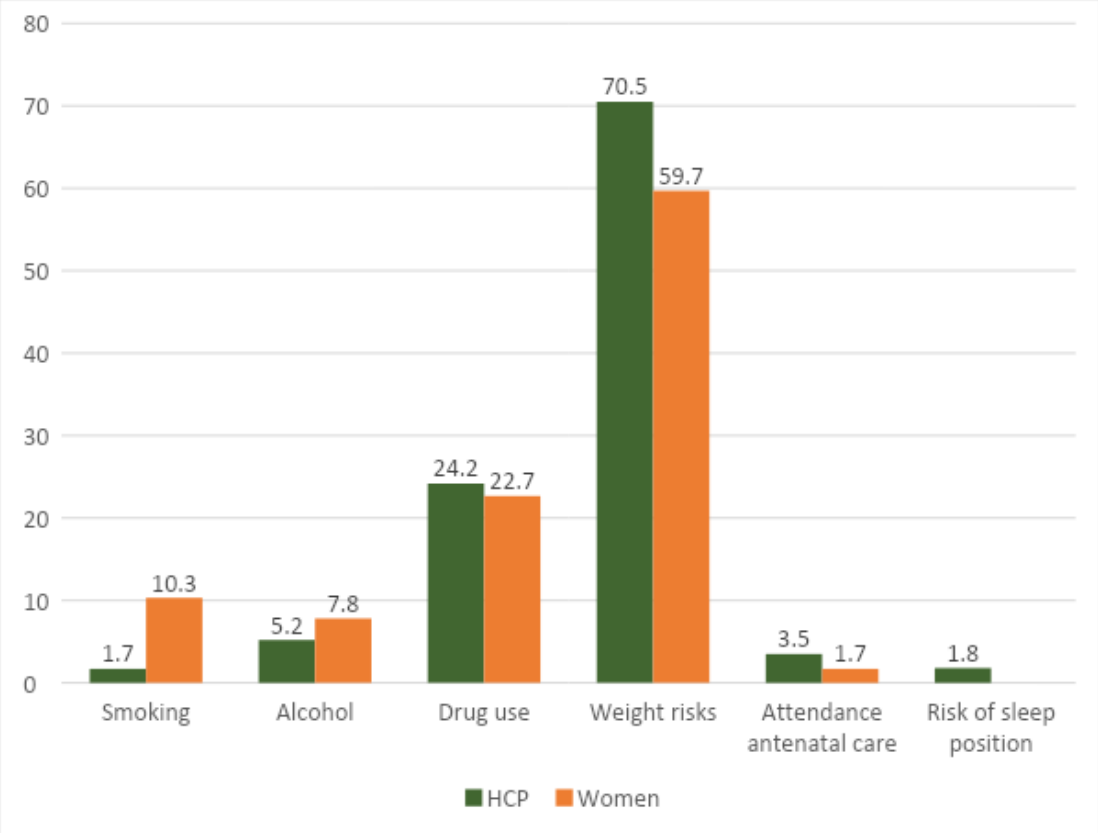


Figure 3. Comparison of topic perceived by HCP as most difficult/reluctant to discuss for themselves and for women.

Table 4. Risk factors prioritised as most difficult/reluctant to discuss (1) to least difficult/reluctant to discuss (6) as perceived by HCPs for themselves and for the women.

	Smoking		Alcohol		Illicit Drugs		Weight related risks		Attendance at antenatal care		Sleeping position	
	HCP (n = 58)	Woman (n = 68)	HCP (n = 58)	Woman (n = 64)	HCP (n = 62)	Woman (n = 66)	HCP (n = 61)	Woman (n = 67)	HCP (n = 57)	Woman (n = 58)	HCP (n = 56)	Woman (n = 57)
	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)
1	1.7 (1)	10.3 (7)	5.2 (3)	7.8 (5)	24.2 (15)	22.7 (15)	70.5 (43)	59.7 (40)	3.5 (2)	1.7 (1)	1.8 (1)	0
2	25.9 (15)	44.1 (30)	19.0 (11)	21.9 (14)	35. 5 (22)	24.2 (16)	9.8 (6)	10.4 (7)	0	0	8.9 (5)	1.8 (1)
3	17.2 (10)	19.1 (13)	55.2 (32)	46.9 (30)	11.3 (7)	24.2 (16)	3.3 (2)	9.0 (6)	8.8 (5)	1.7 (1)	3.6 (2)	0
4	32.8 (19)	25.0 (17)	13.8 (8)	18.8 (12)	27.4 (17)	27.3 (18)	9.8 (6)	16.4 (11)	7.0 (4)	8.6 (5)	5.4 (3)	0
5	12.1 (7)	0	5.2 (3)	4.7 (3)	0	1.5 (1)	4.9 (3)	4.5 (3)	29. 8 (17)	63.8 (37)	48.2 (27)	24.6 (14)
6	10. 3 (6)	1.5 (1)	1.7 (1)	0	1.6 (1)	0	1.6 (1)	0	50. 9 (29)	24.1 (14)	32.1 (6)	73.7 (42)

Attitudes and barriers towards communication and supporting behaviour change

Communication habits and opinions were explored within the survey to elicit if the responding HCP felt that they were responsible for conducting certain tasks with the women, and if they were realistically able to conduct this activity within the constraints of their clinical roles (See Table 5).

Explore women's habits at booking: 75% (n = 54 of 72) of the HCP in the sample reported that this activity fell under their role. Of the total of HCPs that considered this task to be part of their role, 51.9% (n = 28 of 54) were midwives, and 14.8% were consultants (n = 8 of 54). 100% (n = 5/5) of SHOs and 90% of SPRs (n = 9/10) reported that exploring women's habits at booking fell within their role. 62.5% (n = 45 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work, whereas 25% (n = 18 of 72) identified time as a barrier to conducting this task.

Explore the pregnant woman's previous clinical history: 83.1% (n = 59 of 71) of HCPs considered that this activity fell within their role. Of the HCPs that considered this task to be part of their role, 47.5% (n = 28) were midwives, and 18.6% (n = 11) were consultants. 100% of SpRs (n = 10) considered that exploring the pregnant women's previous clinical history is part of their role. 82.4% of midwives and 84.6% of consultants considered that exploring the woman's previous clinical history is part of their role. 94.4% (n = 68 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work.

Conduct examinations to monitor the woman's health: 79.5% (n = 58 of 73) of HCPs considered this task to be part of their role. Of the HCPs that considered this task to fall within their role, 51.9% (n = 28 of 58) were midwives. 100% of the SHOs and the SpRs considered this task to be part of their role, 77.8% of midwifes and 66.7% of consultants considered this task to be part of their role. 90.3% (n = 65 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work.

Conduct examinations to monitor the fetus' health: 78.9% (n = 56 of 72) considered that this task felt within their role. Of the HCPs that considered this task to fall within their role, 48.2% (n = 27 of 56) were midwifes. 100% of SpRs (n = 10), 83.3% of consultants (n = 10) 75% of midwifes (n = 27) and 75% of SHOs (n = 3) considered

that this task felt within their role. 90.3% (n = 65 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work.

Providing advice and information regarding health habits to all pregnant women: 77.8% (n = 56 of 72) reported that this task falls within their role. Of the HCPs that considered this task to fall within their role, 51.8% were midwives followed by SpRs (17.9%). 83.3% of consultants reported that this task felt within their role. 58.3% (n = 42 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work, whereas 37.5% (n = 27 of 72) identified time as a barriers to conduct this task.

Supporting pregnant women to make informed choices regarding their antenatal care and birth options: 81.7% of HCPs considered that this task fell within their role, of them, 53.4% were midwives, 15.5% were consultants and 15.5% were SpRs. 86.1% of midwives and 75% of consultants considered that this task fell within their role. 79.2% (n = 57 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work, whereas 12.5% (n = 9 of 72) identified time as a barriers to conduct this task.

Supporting the pregnant woman with behaviour change when necessary: 76.7% (n = 56 of 73) considered this task to fall within their role. Of the HCPs that considered this task to fall within their role, 53.6% were midwives, 16.1% were SpRs and 12.5% were consultants. 83.3% of midwives and 90% of SpRs agreed that this task fell within their role. 56.9% (n = 41 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work, whereas 37.5% (n = 27 of 72) identified time as a barrier to conducting this task. Furthermore, 4.2% (n = 3 of 72) of HCPs identified lack of adequate knowledge as an additional barrier.

Monitoring the pregnant woman's behaviour change when necessary: 77.5% of HCPs considered this task to fall within their role. Of the HCPs that considered this task to fall within their role, 54.5% were midwives and 16.4% were SpRs. 90% of SpRs and 83.3% of midwifes considered this task to fall within their role. 54.2% (n = 39 of 72) of HCPs agreed that they are able to conduct this task as part of their normal

work, whereas 38.9% (n = 28 of 72) identified time as a barrier to conducting this task.

Referring the pregnant woman to the appropriate services to support their behaviour change when necessary: 83.6% (n = 61/73) agreed that this task falls within their role. Of the HCPs that considered this task to fall within their role, 50.8% were midwives. 100% of SpRs, 80% of SHOs, 86.1% of midwives and 69.2% of consultants consider this task to be part of their role. 91.7% (n = 66 of 72) of HCPs agreed that they are able to conduct this task as part of their normal work, whereas 5.6% (n = 4 of 72) identified lack of knowledge as a barrier.

Table 5. Percentage of participants in the different roles considering that each task falls within their role.

	Total	**SHO**	**SpR**	**Consultant**	**Midwife**	**Specialist Midwife**	**Midwifery management**
	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)	% (n)
Explore the pregnant woman's habits at booking.	75 (54/72)	100 (5/5)	90 (9/10)	66.7 (8/12)	77.8 (28/36)	0	100 (4/4)
Explore the pregnant woman's previous clinical history	83.1 (59/71)	80 (4/5)	100 (10/10)	84.6 (11/13)	82.4 (28/34)	40 (2/5)	100 (4/4)
Conduct examinations to monitor the pregnant woman's health	79.5 (58/73)	100 (5/5)	100 (10/10)	84.6 (11/13)	75 (27/36)	20 (1/4)	100 (4/4)
Conduct examinations to monitor the fetus' health	78.9 (56/71)	75 (3/4)	100 (10/10)	83.3 (10/12)	75 (27/36)	40 (2/5)	100 (4/4)
Provide advice and information regarding health habits to all pregnant women	77.8 (56/72)	80 (4/5)	100 (10/10)	53.8 (7/13)	80.6 (29/36)	50 (2/4)	100 (4/4)
Support pregnant women to make informed choices regarding their antenatal care and birth options	81.7 (58/71)	80 (4/5)	90 (9/10)	75 (9/12)	86.1 (31/36)	25 (1/4)	100 (4/4)

Support the pregnant woman's with behaviour change when necessary	76.7 (56/73)	60 (1/3)	90 (9/10)	53.8 (7/13)	83.3 (30/36)	60 (3/5)	100 (4/4)
Monitor the pregnant woman's behaviour change when necessary	77.5 (55/71)	60 (1/3)	90 (9/10)	58.3 (7/12)	83.3 (30/36)	75 (3/4)	75 (3/4)
Refer the pregnant woman to the appropriate services to support their behaviour change when necessary	83.6 (61/73)	80 (4/5)	100 (10/10)	69.2 (9/13)	86.1 (31/36)	60 (3/5)	100 (4/4)

The survey also explored HCP-specific attitudes regarding providing information about stillbirth to women. 50.7% (n = 36 of 71) of the sample disagreed or strongly disagreed that they had enough time to discuss stillbirth, risk factors and prevention with their patients. 56.3% (n = 40 of 71) of the HCPs agreed or strongly agreed that they felt confident and trained to educate and support women in their behaviour change during pregnancy. Of the people who strongly agreed with this statement, 40% were midwives and 50% were consultants; 100% of them had been working in maternity services for over 10 years.

61% of HCPs agreed or strongly agreed that they could avail of sufficient resources to educate and support women to make health changes in their behaviour. 57.7% agreed or strongly agreed that they felt confident and trained to speak about stillbirth with women under their care. 100% of respondents that had been working in maternity services for less than a year, and 46.2% of respondents that had worked in maternity services between 1-3 years disagreed or strongly disagreed that they felt confident and trained to speak with women about stillbirth.

71.8% (n = 51 of 71) of respondents agreed or strongly agreed that they felt confident in advising patients regarding risk factors and behavioural modification in the context of stillbirth prevention. 60% of SHOs disagreed or strongly disagreed with this statement (n = 4 of 5). 80.3% (n = 57 of 71) of HCPs agreed or strongly agreed that is necessary to mention the risks of stillbirth to women to influence their behaviour change.

Discussion

As outlined above, the survey was broadly divided into three sections. The first dealt with participant experience and involvement in the care of stillborn babies. There was a spectrum of different roles reflected in the responses, including midwives, specialist midwives, and doctors across the training gradient, as well as consultants with several decades of experience. The vast majority of participants had cared for women and stillborn babies in some capacity during their career, including diagnosing intrauterine deaths, attending the delivery of stillborn babies, and caring

for women in the peripartum period. Interestingly, half of respondents had been in their role for less than a year when they were first involved in caring for families experiencing a stillbirth. Given the sensitive nature of this patient contact, this underlines the need for comprehensive training in pregnancy loss at undergraduate level. A significant body of work describes the lived experience of bereaved parents[28,29], with whom poorly managed engagement has the potential to cause further injury in an already difficult situation[30]. Patients report this to be the case even in well-intentioned attempts to comfort them[31], suggesting that informal "on the job" learning in how to communicate with bereaved families may be inadequate. Work with research collaborators in our group and others suggests that simulated training may represent one pathway for improving and practicing communication skills in this specific context for both medical[32] and nursing staff[33].

Moreover, the experience of clinicians in communicating with, and caring for, bereaved families is documented to be emotionally challenging, with previous research suggesting that for some clinicians it can be a factor in leaving the field[34] . Previous work in our group highlights the profound personal and professional impact of caring for families experiencing a stillbirth[35]. This previous work also raised specific concerns regarding junior clinical staff who received no formal debrief or acknowledgement of their human distress. The emotional impact on HCPs was not addressed in our study; however, we confirmed that caring for families after stillbirth is common at early career stages, which suggests a need for formal educational and emotional supports. It is proposed that these interventions would have a positive effect on both clinicians and bereaved families.

The second section considered HCP knowledge regarding stillbirth prevalence and risk factors. The Irish definition of stillbirth is "a child born weighing 500 grammes or more or having a gestational age of 24 weeks or more who shows no sign of life"; half of respondents could correctly identify this definition. The use of the correct definition is important as an incorrect assessment may lead to exclusion of some cases (or inappropriate inclusion) when making clinical decisions, as well as in collating accurate statistics and following correct coronial protocols. Moreover, incorrect categorisation can have significant logistical implications for bereaved

parents at an already difficult time. A pertinent example of this is maternity leave, which in Ireland is extended to women who have had a stillbirth as defined by the national guidelines[36]. However, it should be noted that there are variations in the definition in different jurisdictions, with the CDC in the United States using a cut-off of 20 weeks[37]; conversely, the WHO suggests a global cut-off of 28 weeks, which may reflect realistic viability in resource-deprived settings[38]. Given that many of our cohort of HCPs may have trained and/or worked in other countries, this may reflect some of the variation in responses. Moreover, more recent work in our group suggests that this definition could be revised to ≥22 weeks' gestation and ≥400 g to comply with medical developments improving survival at extreme prematurity; thus underlining the need for continuing professional education and maintaining an up-to-date knowledge of current guidelines[39]. HCP knowledge of risk factors associated with stillbirth was generally good. Our results show that most participants had learned about stillbirth and associated risk factors during postgraduate training. Given the findings from our cohort regarding their experiences with stillbirth early in their careers, it is suggested that stillbirth education at undergraduate level before entering the workforce represents a priority, from both a patient and HCP point of view.

In terms of prevalence of stillbirth, the most recent Irish figures from NPEC suggest a stillbirth rate of 4.20 per 1000 births[3]. We considered a response as valid if it ranged between 3.5 and 4.5. 15% of respondents gave a value between those two parameters, with a wide range outside of these on either side. Accurate knowledge of stillbirth prevalence is crucial so that HCPs can factually educate women, allowing them to make informed choices regarding their behaviours during pregnancy. However, a large body of research suggests that the patient response to "risk" has both rational and emotional components[40]. In relation to health risks in general, the assumption that patient perception of risk is entirely rational is incorrect, and may be detrimental to the efficacy of interventions[41]. As such, if an intervention is to be effective, both *what* HCPs convey and *how* they do so are crucial components of informing patients. This is particularly relevant in areas of sensitive areas of healthcare such as stillbirth, which both patients and healthcare providers report as

being difficult to discuss (15). In discussions of potential negative outcomes in healthcare in general, clinicians report a fear of triggering anxiety in patients[42]. Stillbirth in particular carries its own specific stigma[19]; as do many of the risk factors associated with it, as discussed below. Extensive work done in other fields, such as oncology, around the communication of risks and benefits can inform future strategies. For example, some authors have recommended the use of summary tables and comparisons of outcomes of specific risk factors with baseline population risk[43]. Moreover, expansion of established work around use of visual aids, consistent denominators, absolute numbers and a standardised "risk vocabulary" tailored to pregnancy loss represents an area of future interest[44].

The third section of this work dealt with behaviour change and communication with patients. Several topics were explored in this section. A notable finding was the commitment of respondents to engaging in risk reducing measures. The majority of respondents- over 90%- reported that they provide tailored advice to all women in their care regarding stillbirth risk reduction always or often; over 80% felt it necessary to mention the risks of stillbirth to women to influence their behaviour change. This is consistent with a growing movement for patient autonomy and involvement in healthcare, which in the specific context of stillbirth posits that omitting to discuss the small but real risk of stillbirth represents a negligence of its own. A recent article in the Iowa Law Review postulates that "that a woman has a right to know of the risk of stillbirth before it happens to her child, a right enforceable through an informed consent medical malpractice tort claim for a doctor's failure to disclose. This right also includes the right to information on the preventative measures the woman can perform to help prevent her child's stillbirth"[45]. Bereaved parents also report feelings of stigma around stillbirth being exacerbated by a silence around the topic during their antenatal care[30].

Another topic that was explored in this section was obstacles to care. Several barriers to discussion of risk factors and behavioural change were reported, including inadequate time, lack of confidence and training. Our data suggested that more experienced HCPs felt confident and trained to speak about stillbirth with women under their care and to support behavioural change as compared to less experienced

HCPs. This suggests that a combination of "real world" experience and formal training improves HCP confidence in both education and patient support. As above, it is suggested that training- such as simulation- may provide a safe way for HCPs to learn to "get it right on the first go"[33,46–49]. This has potential to improve the patient experience but also to support HCPs with a structured approach to difficult conversations. Moreover, it is posited that focused communications training will have applications to other of clinical practice to improve patient safety more generall[50].

Similarly, barriers on the part of both HCPs and patients were observed in the discussion of specific risk factors. Regarding substance use, a minority of respondents reported discussing smoking risks, alcohol risks, or illicit drug use with all women under their care often or always, regardless of their substance use status. This universality is important, as a large body of research suggests that women may be reluctant to disclose tobacco use[51] or other substance use Smoking during pregnancy, stigma and secrets: Visual methods exploration in the UK in pregnancy[52,53]. Conversely, over half of HCPs reported discussing substance use risks always or often with women who disclose substance use, though as mentioned previously, this may miss a vulnerable population who do not feel able to disclose. HCPs must acknowledge the manner in which social stigma may influence their discourse around behavioural change, with some qualitative work suggesting "directive" or "critical" public health campaigns have the opposite to intended effect[54,55]. Stigma often represents a significant barrier to engaging in risk reducing measures[56], with vulnerable women often having multiple overlapping exposures to risk[57]. HCPs must also be aware of their own contexts and biases, with some authors noting different reactions in the opinions of non-smoker respondents to smoking in pregnancy[58]. Likewise, weight management in particular continues to be a sensitive issue. In our study, HCPs described their own reluctance to discuss weight management, and also perceived patients as being reluctant to engage with this. The negative impact of weight-associated stigma in both healthcare settings and more generally is well described[59,60], with overweight women more likely to report negative antenatal and peripartum care experiences[61]. Previous research describes a

perceived over-emphasis on weight management in pregnancy by overweight women ("Everything is revolved around me being heavy"[62]. In that study, active engagement with risk reduction rather than merely describing risk was perceived as more useful, underlining the need for practical advice and support. Indeed, women with a higher BMI report an a dehumanizing effect of over-emphasis of weight risks which "eclipsed the ability of maternity carers to view them as a whole person"[63]. By the same token, long term outcomes besides stillbirth must be considered. For example, the"WOMBS" (Weight gain, Obesity, Maternal-child Biobehavioral pathways, and Stigma) Framework "highlights pregnant and postpartum women as a theoretically unique at-risk population for whom this social stigma engages maternal physiology and transfers obesity risk to the child via social and physiological mechanisms"[64], underlining the need for particular sensitivity in this population. Thus, HCP and patient reluctance to engage in difficult weight-related discussions may be understood in the context of the challenge of balancing these risks and benefits. In the context of stillbirth prevention, further research is warranted in how best to compassionately address this challenge.

In terms of planning for future pregnancies after loss, poor obstetric history was noted to be a risk factor for pregnancy loss by many respondents. Previous research in the UK[65,66] suggest that significant variations in antenatal care exist for women who have previously suffered a pregnancy loss, and specifically highlighted the need for emotional and psychological support in this special cohort. Work in our group suggests that men and women have different needs and priorities following a stillbirth, but that in general additional scheduled care and reassurance was considered important[67]. Moreover, a past history of stillbirth represents an independent risk factor for another stillbirth[68], which must be recognised in planning future care. Addressing specific causes, such as placental vascular disorders, represents a key component of reducing the risk of another stillbirth[69].

Adherence to guidelines and best practice, while important, is not the only component of providing good care to families; a large systematic review describes parental distress in relation to a perception of HCPS "hiding behind 'doing' and ritualising guidelines"[70]. More generally, women report valuing continuity of care and

person-centred care[71]. Other authors have explored the concept of "complementarity" in combining "evidence-based practice and patient-centered practice"[72], with a reminder that each case will be different depending on a myriad of patient, socio-cultural and contextual factors[73]. International data[74–76] suggests that being part of an ethnic minority is a risk factor for stillbirth (though the mechanism underlying this is unclear[74–76], and likely related to socioeconomic disadvantage[75] which underlines the need for sensitive and culturally appropriate outreach, interventions, and supports, rather than a "one size fits all" approach[77]. Policy design should ideally be intersectional, recognising the interactions between a myriad of different factors in stillbirth risk (52). Acknowledgement of the global health context is also of crucial importance in order to maximise benefits of any interventions[78]. Further work regarding the integration of tailored peripartum care is warranted, given the prevalence and impact of stillbirth in our patient population. Any such work should address knowledge, practical, educational and logistical elements to improve care and reduce the risk of stillbirth.

Conclusion

Stillbirth is a devastating pregnancy outcome for parents, families and the healthcare professionals involved. HCPs describe significant issues in discussing risk factors and behavioural changes with patients under their care. Lack of time and patient reluctance are reported as major barriers. Given the devastating outcome of stillbirth on women and their families, prioritisation of protected time to discuss risk factors and behaviour modification are of crucial importance. In addition, important gaps in HCP knowledge are evident, strengthening the case for additional resourcing of education at undergraduate and postgraduate levels. This may include tailored intervention training and simulation; more research is warranted.

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Survey

Survey draft V3_08/06/2021

RELEVANT study

REducing stiLlbirth through bEhaViour chAnge iNtervenTions.

Section 1: About your experience

Q1. Professional role of respondent:

- ❑ SHO
- ❑ Registrar
- ❑ SPR
- ❑ Consultant
- ❑ Midwife
- ❑ Specialist Midwife
- ❑ Midwifery Management
- ❑ Nurse
- ❑ GP trainee

Q2. Place of work

- ❑ Outpatient clinics
- ❑ Antenatal wards
- ❑ Postnatal wards
- ❑ Labour ward
- ❑ Early pregnancy unit
- ❑ Fetal assessment unit/ ultrasound department
- ❑ Emergency room
- ❑ Outreach clinics (outside of the main hospital)
- ❑ All of the above

Q3. Gender

- ❑ Male
- ❑ Female
- ❑ Prefer not to say
- ❑ Prefer to self-identify (use space below)

Q4. Years working in maternity services:

- ❑ Less than a year
- ❑ 1-3 years
- ❑ 4-6 years
- ❑ 10-15 years
- ❑ 16-20 years
- ❑ >20 years

Q.5 Have you previously been involved in caring for women after stillbirth in any capacity?

- ❑ Yes
- ❑ No

Q.5.1 If yes, how long approximately had you been working in maternity services when this happened?

- ❑ Less than a year
- ❑ 1-3 years
- ❑ 4-6 years
- ❑ 7-10 years
- ❑ 10-15 years
- ❑ 16-20 years
- ❑ >20 years

Q6. Have you ever diagnosed an intrauterine death? (by ultrasound, CTG, hand-held Doppler etc)

- ❑ Yes
- ❑ No

Q7. Have you been present at the delivery of a stillborn baby?

- ❑ Yes
- ❑ No

Q8. How often do you care for parents that have experienced a stillbirth?

- ❑ Never
- ❑ Around once a year
- ❑ Several times a year
- ❑ Around once a month
- ❑ Around once a week

Section 2: Stillbirth and risk factors

Q9. How would you define a stillbirth in Ireland (gestation and/or birthweight)?

A baby born with no signs of life

- ❑ At a gestational age of 22 weeks or more
- ❑ At a gestational age of 24 weeks or more
- ❑ At a gestational age of 26 weeks or more
- ❑ At a gestational age of 28 weeks or more

❑ AND
❑ OR

- ❑ A birthweight of 400g
- ❑ A birthweight of 500g
- ❑ A birthweight of 600g
- ❑ A birthweight of 1000g

Q10. How prevalent do you estimate stillbirth in Ireland to be, as a figure per 1000 births?

Q11. What are the most common risk factors associated with the risk of stillbirth, to the best of your knowledge? Please list as many as you can.

Q12. Which of these would you estimate to be the most important preventable cause of stillbirth?

Q13. What are the most common modifiable risk factors associated with the risk of stillbirth, to the best of your knowledge? Please list as many as you can.

Q14. Where did you learn about these risk factors?

- ❑ Undergraduate/ university teaching
- ❑ Hospital based education
- ❑ Postgraduate training (e.g: webinars, conferences, formal study modules)
- ❑ Professional guidelines
- ❑ Journal articles
- ❑ Other (please state)

Q15. Have you ever attended any of the following educational events (specifically relating to the topic of stillbirth)?

- ❑ Hospital based study day
- ❑ National conference
- ❑ International conference

Q16. What resources do you use to keep your knowledge up to date?

- ❑ Professional guidelines
- ❑ Journal articles
- ❑ Study days/ CPD within hospital group
- ❑ Study days/ CPD outside of the hospital group (IOG, RCPI etc)
- ❑ Other (please state)

Section 3: Behaviour change and communication with patients

… Q17. Please rate your level of agreement with the following statements:

	Never	Rarely	Sometimes	Often	Always
I provide tailored advice to all women in my care					
I discuss smoking risks with all women in my care, regardless of their smoking status.					
I discuss alcohol use risks during pregnancy with all women in my care, regardless of their drinking status.					
I discuss drug use during pregnancy with all women in my care, regardless of their drug use status					
I only discuss substance use (smoking, alcohol or other illicit drugs) with women in my care when they disclose using such substances.					
I discuss body-weight risks with all women in my care, regardless of their weight at booking.					
I give weight management advice to all women in my care, regardless of their weight at booking.					
I focus weight management advice to women in my care who are overweight or obese at booking.					
I advise women in my care to attend and engage with all their antenatal appointments, regardless of their booking time.					
I only stress the importance of attending appointments with women who book late in pregnancy, or who have a tendency to reschedule appointments.					
I provide advice about safest sleeping positions to all					

women in my care					
I provide advice regarding monitoring baby's movements to all women in my care					
I provide advice regarding the importance of attending scheduled antenatal care to all women in my care					
I provide advice regarding the importance of seeking unscheduled/emergency antenatal care if they have concerns to all women in my care.					

Q18. Which of these, if any, do you think is most **important** to discuss? Please enumerate from 1 "most important" to 6 "Least important".

... Smoking risks
... Alcohol use risks
... Illicit drug use risks
... Weight risks/weight management
... Importance of attending/engaging with antenatal care
... Risks of sleep position

Q19. Which of these, if any, do you find **difficult**/are most reluctant to discuss? Please enumerate from 1 "most difficult" to 6 "Least difficult".

... Smoking risks
... Alcohol use risks
... Illicit drug use risks
... Weight risks/weight management
... Importance of attending/engaging with antenatal care
... Risks of sleep position

20. Which of these, if any, do you perceive that the **pregnant women** finds most difficult/ are most reluctant to discuss? Please enumerate from 1 "most difficult" to 6 "Least difficult".

... Smoking risks
... Alcohol use risks
... Illicit drug use risks
... Weight risks/weight management
... Importance of attending/engaging with antenatal care
... Risks of sleep position

Q21. Do you use additional resources to provide information to the pregnant women in your care about risk factors for stillbirth?

- ❑ Yes
- ❑ No

If so, what resources do you use?

Q22. Which of the following tasks do you think are part of your role? If you consider this task to be part of your role, please choose the option "This falls within my role", if you consider this task to not be part of your role, please select a different option accordingly.

	This falls within my role	Role of the doctor	Role of the midwife	Role of specialist midwife	Role of the dietician	Role of the social worker	Other
Explore the pregnant woman's habits at booking.							
Explore the pregnant woman's previous clinical history							
Conduct examinations to monitor the pregnant woman's health							
Conduct examinations to monitor the fetus' health							
Provide advice and information regarding health habits to all pregnant women							
Support pregnant women to make informed choices regarding their antenatal care and birth options							
Support the pregnant woman's with behaviour change when necessary							
Monitor the pregnant woman's behaviour change when necessary							

Refer the pregnant woman to the appropriate services to support their behaviour change when necessary							

Q23. Please consider the following tasks and whether you are realistically able to conduct them within your job considering limitations?

	I am able to conduct this within my normal work	I am not able to conduct this within my normal work due to time constraints	I am not able to conduct this within my normal work as I do not feel I have adequate knowledge	I am not able to conduct this within my normal work as I do not feel I have had the required training	I am afraid to conduct this within my normal work	Other
Explore the pregnant woman's habits at booking.						
Explore the pregnant woman's previous clinical history						
Conduct examinations to monitor the pregnant woman's health						
Conduct examinations to monitor the fetus' health						
Provide advice and information regarding health habits to all pregnant women						
Support pregnant women to make informed choices regarding their antenatal care and birth options						
Support the pregnant woman's with behaviour change when necessary						
Monitor the pregnant woman's behaviour change when necessary						

Q24. Please rate your agreement with the following statements

	Strongly disagree	Disagree	Neither agree nor disagree	Agree	Strongly agree
Informing women about health behaviours and risks is part of my role.					
Supporting women to make healthy changes (e.g: stop using substances, support healthy eating and active lifestyle, promote engagement with antenatal care, inform about safe sleeping positions, etc.) in their behaviour is part of my role.					
Informing women about health behaviours and risks is part of my role.					
I feel like I have enough time to discuss stillbirth, risk factors, and prevention with my patients					
I feel confident and trained to educate and support women in their behaviour change within my job					
I feel like I can avail of enough resources (sources of information, specialist services, etc.) to educate and support women to make health changes in their behaviour.					
I feel confident and trained to speak about stillbirth to women under my care.					
I feel confident in advising my patients regarding risk factors and behaviour modification in the context of stillbirth prevention.					
I think that it is necessary to mention the risks of stillbirth to influence patient behaviours during pregnancy.					

Thank you very much for completing this survey.

Supplementary file 1.2 – Intervention functions and policy categories

Intervention function, policy categories and definitions obtained from Michie et al. (2014)[259]

Intervention functions	
Education	"Increasing knowledge and understanding"
Persuasion	"Using communication to induce positive or negative feelings or stimulate action"
Incentivisation	"Creating expectations of reward"
Coercion	"Creating expectations of punishment or cost"
Training	"Imparting skills"
Restriction	"Using rules to reduce the opportunity to engage in the target behaviour (or to increase the target behaviour by reducing the opportunity to engage in competing behaviours)"
Environmental restructuring	"Changing the physical or social context"
Modelling	"Providing an example for people to aspire to or imitate."
Enablement	"Increasing means/reducing barriers to increase capability or opportunity"
Policy categories	
Communication/Marketing	"Using print, electronic, telephonic or broadcast media"
Guidelines	"Creating documents that recommend or mandate practice. This includes all changes to service provision."
Fiscal	"Using the tax system to reduce or increase the financial cost."
Regulation	"Establishing rules or principles of behaviour or practice."
Legislation	"Making or changing laws."
Environmental/Social Planning	"Designing and/or controlling the physical or social environment."
Service provision	"Delivering a service."

Supplementary file 2.1 – Definitions of stillbirth as per different agencies

Agency	Definition
Irish Stillbirth Registration Act	Infant born weighing **500grammes or more** or at a gestational age of at least **24 weeks** who shows no signs of life
MBRRACE-UK Perinatal Mortality Surveillance Report	Baby delivered at or after **24+0 weeks** gestational age showing no signs of life
Wold health organization (for international comparison)	Baby born with no signs of life at or after **28 weeks'** gestation or weighing at least **1000g** or being 35cm length
International classification of diseases 10th edition (WHO)	Fetus that has suffered intrauterine or intrapartum death after **24 weeks** gestation
International classification of diseases 11th edition (WHO)	Early stillbirth is defined as an early fetal death after **22 weeks** gestation and a birthweight of **500gm** or more
European Medicine Agency	Death of a fetus after **22 weeks** of gestation
Centers for Disease control and prevention (USA)	Loss of a baby after **20 weeks** of pregnancy or weighting 350gr.
Australian Institute of Health and Welfare	The birth of a baby who is **20 or more completed weeks** of gestation or of 400 grams or more birthweight who is expelled or extracted from his or her mother and shows no signs of life such as a heartbeat, voluntary muscle movement or pulsation of the umbilical cord

Supplementary file 3.1 – List of websites divided by provider

Type of provider	Definition of provider	Websites included		Date/s accessed	Topic
Professional bodies	Websites from official professional organizations	British Association of Perinatal Medicine	http://www.bapm.org/	21/05/2019	General
		British Maternal & Fetal Medicine Society	https://www.bmfms.org.uk/	21/05/2019	General
		Institute of Obstetricians and Gynaecologists	https://www.rcpi.ie/faculties/obstetricians-and-gynaecologists/	17/05/2019	General
		National Institute for Health and Clinical Excellence	http://www.nice.org.uk	22/05/2019 23/05/2019	General
		National Patient Safety Office	https://health.gov.ie/national-patient-safety-office/ncec/	21/05/2019	General
		Nursing and midwifery board of Ireland	https://www.nmbi.ie/Home	21/05/2019	General
		Royal College of Obstetricians and Gynaecologists	https://www.rcog.org.uk/	17/05/2019	General
		Royal College of Physicians of Ireland	https://www.rcpi.ie/	17/05/2019	General

Charities and voluntary organizations	Containing the websites from public organizations that are registered as charities	A little lifetime foundation	https://alittlelifetime.ie/	10/06/2019	Specialized
		AIMS Ireland	http://aimsireland.ie/pregnancy-loss/	06/06/2019	General
		Anam Cara	https://anamcara.ie/	10/06/2019	Specialized
		Antenatal Results Choices	https://www.arc-uk.org/	27/05/2019	General
		Baby lifeline	http://babylifeline.org.uk/home/	28/05/2019	General
		Bernardos	https://www.barnardos.ie/	10/06/2019	Others
		Child Bereavement UK	https://childbereavementuk.org/	28/05/2019	Specialized
		Every life counts	https://www.everylifecounts.ie/	11/06/2019	Specialized
		Feileacain	https://feileacain.ie/	10/06/2019	Specialized
		International Stillbirth Alliance (ISA)	http://www.stillbirthalliance.org	10/06/2019	Specialized
		Irish childhood bereavement network	https://www.childhoodbereavement.ie/	10/06/2019	Others

	Irish hospice foundation	https://hospicefoundation.ie/	10/06/2019	Others
	Irish Neonatal Health Alliance	http://www.inha.ie/bereavement/	22/05/2019	Specialized
	Kicks count	https://www.kickscount.org.uk/	28/05/2019	Specialized
	Little things and Co	http://www.ltandco.org/	29/05/2019	Specialized
	Mama academy	https://www.mamaacademy.org.uk/	29/05/2019	Specialized
	Miscarriage Association	https://www.miscarriageassociation.org.uk/	01/07/2019	Specialized
	National infertility and support and information group	http://nisig.com/	10/06/2019	General
	National Maternity Support Foundation	https://www.jakescharity.org/	06/06/2019	Specialized
	NCT	https://www.nct.org.uk/	10/06/2019	General
	One day more	https://onedaymore.ie/	10/06/2019	Specialized
	Safer Pregnancy	https://saferpregnancy.org.uk/	23/05/2019	General

		SANDS stillbirth and neonatal death charity	https://www.sands.org.uk/	22/05/2019	Specialized
		Saying Goodbye	https://www.sayinggoodbye.org/	06/06/2019	Specialized
		Termination for medical reasons Ireland	http://tfmrireland.com/	10/06/2019	Specialized
		The Lullaby Trust	http://www.lullabytrust.org.uk/LThome	22/05/2019	Specialized
		The miscarriage association of Ireland	http://www.miscarriage.ie/	01/07/2019	Specialized
		Tommy's	https://www.tommys.org/	27/05/2019	General
Support groups	Websites created by groups of parents or affected people not registered as charities	4Louis	https://4louis.co.uk/	11/06/2019	Specialized
		Abbie's fund	http://www.abigailsfootsteps.co.uk/	11/06/2019	Specialized
		Aching Arms	http://www.achingarms.co.uk/	11/06/2019	Specialized
		Aidan's Elephant	http://www.aidanselephants.co.uk/	11/06/2019	Specialized
		Bethany Bereavement Support	https://www.bethany.ie/	11/06/2019	Others

		Cariad Angel Gowns	http://www.cariadangelgowns.org.uk/	11/06/2019	Specialized
		Forever stars	http://www.foreverstars.org/	11/06/2019	Specialized
		Jude Brady Foundation	http://www.judebradyfoundation.co.uk/	11/06/2019	Specialized
		Leanbh Mo Chroi	https://lmcsupport.ie/	11/06/2019	Specialized
		Life after loss	http://www.lifeafterloss.org.uk/site/	11/06/2019	Specialized
		Sophia - Pregnancy Loss Support	https://www.sophiapregnancylosssupport.com/	11/06/2019	Specialized
		The Lily Mae Foundation	https://www.lilymaefoundation.org/	11/06/2019	Specialized
Commercial sites	Websites provided by private companies	Baby Center	https://www.babycentre.co.uk/	12/06/2019	General
		Emma's diary	https://www.emmasdiary.co.uk/	24/06/2019	General
		Everymum	https://www.everymum.ie/	12/06/2019	General
		Irish Life health Pregnancy	https://www.irishlifehealth.ie/the-parenting-path/getting-pregnant	24/06/2019	General
		Laya healthcare	https://www.layahealthcare.ie/formembers/mumbaby/	24/06/2019	General

Mummy pages	https://www.mummypages.ie/	13/06/2019	General
Mumset	https://www.mumsnet.com/	12/06/2019	General
Pampers	https://www.pampers.co.uk/pregnancy	13/06/2019	General
Pregnancy and baby	https://www.pregnancyandbaby.ie/	24/06/2019	General
Pregnancy counselling and care	https://www.counsellingandcare.co.uk/	12/06/2019	General
Pregnancy UK	http://pregnancy.uk/	13/06/2019	General
Reflect, pregnancy support	https://reflectyork.co.uk/	12/06/2019	General
Rollercoaster	https://rollercoaster.ie/	24/06/2019	General
VHI pregnancy articles	https://www.vhiblog.ie/tag/pregnancy/	24/06/2019	General
Whatsupmum	https://whatsupmum.ie/	24/06/2019	General

Health services websites	Websites from national health providers	Coombe women and infant university hospital	http://www.coombe.ie/	01/07/2019	General
		CUMH	http://www.cuh.hse.ie/Cork-University-Maternity-Hospital/	01/07/2019	General
		Health Service Executive	https://www.hse.ie/eng/	26/06/2019	General
		Miscarriage and pregnancy loss CUH	http://www.cuh.hse.ie/Cork-University-Maternity-Hospital/Maternity-Care/Miscarriage-Pregnancy-Loss/	26/06/2019	Specialized
		National Health Service	https://www.nhs.uk/	03/07/2019	General
		Perinatal institute for maternal and child health	https://www.perinatal.org.uk/	26/06/2019	General
		Pregnancy and infant loss	https://pregnancyandinfantloss.ie/	02/07/2019	Specialized
		Private midwives	https://privatemidwives.com/	01/07/2019	General
		Rotunda hospital	https://rotunda.ie/	02/07/2019	General
		Rotunda private	https://rotundaprivate.ie/	02/07/2019	General

		Saolta University Health Care Group	https://saolta.ie/	02/07/2019	General
		The National Maternity Hospital	http://www.nmh.ie/	01/07/2019	General
Health sites	Private websites with the purpose to offer a service or provide information	Beacon care fertility clinic	https://www.beaconcarefertility.ie/	03/07/2019	General
		Bon Secours health system	https://www.bonsecours.ie/	03/07/2019	General
		Cork obstetrics and gynaecology associates	http://www.coga.ie/	03/07/2019	General
		Cork Womens Clinic	https://corkwomensclinic.com/	03/07/2019	General
		Dr. Dan McKenna. Consultant Obstetrician and Gynaecologist	https://www.drdanmckenna.ie/	03/07/2019	General
		Dr. Moya McMenamin	https://www.drmoyamcmenamin.ie/gynaecologist-cork/home	03/07/2019	General
		EVIE preparation for life	https://evie.ie/	03/07/2019	General
		Galway Fertility clinic	https://www.fertilityclinic.ie/	03/07/2019	General

	Irish health	http://www.irishhealth.com/	26/06/2019	General
	Mater private Cork	https://www.materprivate.ie/cork/	03/07/2019	General
	Merrion fetal health	https://www.merrionfetalhealth.ie/	03/07/2019	General
	Repromed fertility clinic	https://repromed.ie/	03/07/2019	General
	SIMS fertility clinic	https://www.sims.ie/	03/07/2019	General
	The Scotia Clinic	https://scotiaclinic.ie/	03/07/2019	General
	UPMC Whitfield	http://www.whitfieldclinic.ie/	03/07/2019	General
	Waterstone clinic	https://waterstoneclinic.ie/	03/07/2019	General
	Women's health	https://www.womenshealth.ie/	03/07/2019	General

Supplementary file 3.2 – Codebook

Name of website		
Link		
Date accessed		
Main topic of website	General	1
	Specialized	2
Website provider	Professional body	1
	Charity	2
	Support group / patient organization	3
	Commercial sites	4
	Health Service	5
	Health private sites	6
Addressed to	Professionals	1
	General Public	2
Country	Ireland	1
	UK	2
Presence of advertisement	Yes / No	
Accreditation from official organization	Yes / No	

BASIC INFORMATION ABOUT STILLBIRTH		Yes	No
1. Is there any information about stillbirth in the body of website?			
2.1 Is there information about stillbirth's prevalence?			
2.2 Is there information about causes of stillbirth?			
2.3 Is there information about hospital procedures (post-mortem, independent investigations, role of the coroner, etc.)?			
2.3 Is there information about medical/psychological consequences of stillbirth?			
2.4 Is there information about support for parents or families who have experienced a stillbirth?			
2. If not, are there any links to external resources (other websites, guidelines, etc.) that contain this information?			
2.1 If yes, specify:	Clinical guideline		1
	Patient information leaflet		2
	Website		3
	Others		4

INFORMATION ABOUT RISK FACTORS	Yes	No
1. Is there information about risk factor for stillbirth in the body of the website?		
2. Is there any information about behavioural risk factors for stillbirth in body of the website?		
2.1 Is there information stating that **smoking** is a risk factor for stillbirth in the body of the website?		
2.2 Is there information stating that **Illicit drug use** is a risk factor for stillbirth in the body of the website?		
2.3 Is there information stating that **medical drug use** is a risk factor for stillbirth in the body of the website?		
2.4 Is there information stating that **sleeping position** is a risk factor for stillbirth in the body of the website?		
2.5 Is there information stating that **Attendance to antenatal care** is a risk factor for stillbirth in the body of the website?		
2.6 Is there information stating that **Alcohol use** is a risk factor for stillbirth in the body of the website?		
2.7 Is there information stating that **Obesity and overweight** are risks factors for stillbirth in the body of the website?		
3. If not, are there any links to external resources (other websites, guidelines, etc.) that contain this information?		
3.1 If yes, specify: (Clinical guideline 1 / Patient information leaflet 2 / website 3 / others 4)		
4. Other risk factors mentioned on the body of the website:		

Supplementary file 4.1 – Example of search

Database: Web of Science	
	Search Terms
Concept 1 Facilitators and Barriers	
1.	TO Facilitator*
2.	TO Barrier*
3.	TO Promoter*
4.	TO Benefit*
5.	TO Attitude*
6.	TO Opportunit*
7.	TO Determinant*
8.	TO Promotion
9.	TO Intention
10.	TO Education
11.	TO Initiative
12.	TO Prevention
13.	TO Pregnancy
14.	**S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 AND S13**
Concept 2. Attendance at antenatal care	
15.	TO Attendance
16.	TO “Antenatal care”
17.	TO “Prenatal Care”
18.	TO “Antenatal care booking”
19.	**S20 AND S21 OR S22 OR S23**
20.	**S14 AND S24**
Total	

Criteria for selection of studies

Inclusion criteria:

- Use of qualitative methods
- Written in English
- Participants were pregnant women or women up to 12 months postpartum.
- Conducted in high-income countries
- Included extractable data about facilitators and barriers to seeking, attending or complying with antenatal care.

Exclusion criteria

- Not qualitative data or no use of primary data.
- Studies conducted in low- or middle-income countries.
- If a study included other types of participants besides pregnant or postpartum women up to 12 months, they were discarded unless the data were reported in a manner where the data could differentiated by eligibility characteristics.

Supplementary file 4.2 – CASPs Scores

Reference	Clear aims	Qualitative methodology appropiate	Research design appropiate to meet aims	Recruitment strategy appropiate	Data collected in a way that addressed the research issue	Relationship between researcher and participants considered	Ethical issues	Data analysis rigorous	clear statement of findings	How valuable is the research	Total Score
Callaghan, (2011)	2	2	1	2	2	0	2	2	2	2	17
Daniels, P., 2006)	2	2	2	2	2	0	1	1	1	1	14
Docherty, A., (2012)	2	2	2	2	2	0	1	1	2	2	16
Haddrill, R.(2014).	2	2	2	2	2	0	1	1	2	2	16
Meyer, E.,(2016)	2	2	1	1	2	0	1	2	1	1	13
Mehta, (2017)	2	2	2	1	2	1	1	2	2	1	16
Roman, . (2017)	2	2	1	1	2	0	1	2	2	2	15

Ross, E. (2012).	2	2	1	1	2	0	1	2	2	2	15
Hatherall, B., (2016).	2	2	1	1	2	0	1	2	1	2	14
Larsson, Å., (2017).	2	2	2	1	2	1	2	1	2	1	16
Tighe, S. M. (2010).	2	2	2	1	2	0	1	1	2	1	14
Salm Ward TC, (2013)	2	2	2	2	2	1	1	2	2	2	18
Johnson JL,	1	2	1	1	1	0	1	1	1	1	10
Funge et al. (2020)	2	2	0	2	1	0	2	1	1	1	12
Sami et al. (2019)	2	1	1	1	1	1	1	1	1	1	11

Supplementary file 4.3 – GRADE CerQual Analysis

Attitudes towards pregnancy and antenatal care summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 1: Some women had ambivalent feelings towards their pregnancies, or felt overwhelmed with the idea of parenthood, and these feelings were intensified when the pregnancy was not planned. This led some women to delay care in order to avoid disclosure or to spend time considering termination.	390–398,402	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 5/10 had a poor justification of their choice of research design • 7/10 studies recruitment strategy poorly described. • 8/10 no reflexivity statement, 2/10 unclear reflexivity. 11/18 studies poorly reported ethical implications. • 4/10 data analysis method poorly discussed or justified. • 3/10 described their findings poorly/ • 0/10 studies borderline score of 10 **Coherence:** No or very minor concerns.	High confidence.

		Adequacy: No or very minor concerns. **Relevance:** No or very minor concerns.	
Findings 2: Women who had planned their pregnancies and had positive feelings towards their pregnancy had a tendency to access antenatal care in a timely manner.	390,392,394–396	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/5 had a poor justification of their choice of research design • 3/5 studies recruitment strategy poorly described. • 3/5 no reflexivity statement, 2/5 unclear reflexivity. 3/5 studies poorly reported ethical implications. • 3/5 data analysis method poorly discussed or justified. • 1/5 described their findings poorly • 0/5 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	High confidence.

Finding 3: Women perceived the importance of antenatal care differently. Some women considered pregnancy as a normal life event that doesn't need medical attention unless feeling unwell, and relied on their own skills and knowledge, engaging in a process of self-care.	390–396,398,401,402	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 2/6 had a poor justification of their choice of research design • 3/6 studies recruitment strategy poorly described. • 4/6 no reflexivity statement, 2/6 unclear reflexivity. 3/5 studies poorly reported ethical implications. • 3/6 data analysis method poorly discussed or justified. • 1/6 described their findings poorly • 0/6 studies borderline score of 10 **Coherence:** Moderate concerns. Although there is data supporting this finding. There is also data that explains that women considered attending antenatal care as part of their self-care. **Adequacy:** No or very minor concerns.	Moderate confidence.

		Relevance: No or very minor concerns.	
Finding 4: Women who adopted an active role in their care, and understood the value of antenatal care, tended to attend early during pregnancy. For these women, antenatal care acted as preventive measure, source of knowledge and support, and distraction.	390,391,402,403,392–398,400	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/12 had a poor justification of their choice of research design • 8/12 studies recruitment strategy poorly described. • 9/11 no reflexivity statement, 3/12 unclear reflexivity. 10/12 studies poorly reported ethical implications. • 6/12 data analysis method poorly discussed or justified. • 4/12 described their findings poorly • 0/12 studies borderline score of 10 **Coherence:** Minor concerns. Although reported in most cases, some data suggests	High confidence.

		that women adopting an active role in their care delayed care seeking as they felt confident enough in their own abilities. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	

Previous experiences summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 5: Having had previous negative experiences using the maternity services or learning about other women's negative experiences influenced women's behaviours. These negative experiences included negative encounters with healthcare professionals and traumatic pregnancies and labour.	390,392–395,401,403	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/7 had a poor justification of their choice of research design • 3/7 studies recruitment strategy poorly described. • 6/7 no reflexivity statement, 1/4 unclear reflexivity. 6/7 studies poorly reported ethical implications. • 4/7 data analysis method poorly discussed or justified. • 2/7 described their findings poorly • 1/7 studies borderline score of 10	High confidence.

		Coherence: No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	

Healthcare professionals' attitudes summary of review finding	**Studies contributing to the review finding**	**Explanation of CERQual assessment**	**CERQual assessment of confidence in the evidence**
Finding 6: Some women perceived healthcare professionals held certain biases towards them which led women to disengage from their care. These include judgements, stigmatisation, racism and inequality.	390,392–396,398,404	**Methodological limitations:** Minor concerns regarding methodological limitations. • 3/8 had a poor justification of their choice of research design • 4/8 studies recruitment strategy poorly described. • 5/8 no reflexivity statement, 3/8 unclear reflexivity. 6/8 studies poorly reported ethical implications. • 3/8 data analysis method poorly discussed or justified.	Moderate confidence.

		• 2/8 described their findings poorly • 0/11 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. Most of the studies contributing to this finding explore women of low socioeconomic backgrounds.	
Finding 7: Some women perceived their healthcare professionals to be insensitive and lacked empathy, which made women feel disrespected.	392–395,402,404,686	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/7 had a poor justification of their choice of research design • 4/7 studies recruitment strategy poorly described. • 6/7 no reflexivity statement. 5/7 studies poorly reported ethical implications.	High confidence.

		• 2/7 data analysis method poorly discussed or justified. • 0/7 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	
Finding 8: Other women found non-judgemental, supportive and honest healthcare professionals which made for a very positive experience during their care, acting as a facilitator for engagement.	395,396,398,399,403,686	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/6 had a poor justification of their choice of research design • 4/6 studies recruitment strategy poorly described. • 5/6 no reflexivity statement. 4/6 studies poorly reported ethical implications. • 4/6 data analysis method poorly discussed or justified. • 0/6 studies borderline score of 10 **Coherence:** No or very minor concerns.	High confidence.

		Adequacy: No or very minor concerns. **Relevance:** No or very minor concerns.	

Communication with healthcare professionals summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 9: Women spoke about feeling confused and poorly informed due to communication issues. These included contradictive messages, having concerns dismissed and feeling uninformed.	395,396,398,401–403,686	**Methodological limitations:** Serious concerns regarding methodological limitations. • 5/7 had a poor justification of their choice of research design • 5/7 studies recruitment strategy poorly described. • 6/7 no reflexivity statement, 1/7 unclear reflexivity. 5/7 studies poorly reported ethical implications. • 3/7 data analysis method poorly discussed or justified. • 0/7 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** Minor concerns. Most of the data comes from 3 of the studies. The other studies contributions is quite limited. **Relevance:** No or very minor concerns.	Moderate confidence.

Finding 10: Women valued positive communication with healthcare professionals such as engaging in active listening, taking time to address women's concerns and encouraging women to participate in the decision making process.	391,393,395,396,398,403	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 2/6 had a poor justification of their choice of research design • 4/6 studies recruitment strategy poorly described. • 5/6 no reflexivity statement. 4/6 studies poorly reported ethical implications. • 3/6 data analysis method poorly discussed or justified. • 0/6 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	High confidence.

Unsatisfactory clinical experiences summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 11: Some women had negative experiences during their use of the antenatal services which led to disengagement. Feeling rushed, long waits, conditional treatment and disrespectful environments made women perceive antenatal care as a waste of time.	392–396,403	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 2/6 had a poor justification of their choice of research design • 3/6 studies recruitment strategy poorly described. • 4/6 no reflexivity statement. 4/6 studies poorly reported ethical implications. • 3/6 data analysis method poorly discussed or justified. • 0/6 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. This finding refers mostly to how women from low socioeconomic backgrounds.	Moderate confidence.

Organisation of antenatal care summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 12: Women reported structural barriers when trying to access antenatal care such as administrative delays and problems with the referral systems. For others, the application process was too complex and women reported a lack of information regarding the organization and functioning of the antenatal care system.	378,390,402,404,391,393–395,398–401	**Methodological limitations:** Serious concerns regarding methodological limitations. • 7/11 had a poor justification of their choice of research design • 7/11 studies recruitment strategy poorly described. • 3/11 showed issues related to the data collection method. • 8/11 no reflexivity statement. 9/11 studies poorly reported ethical implications. • 5/11 data analysis method poorly discussed or justified. • 1/11 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. Although applicable to all of high-income countries, these barriers were accentuated in the	Moderate confidence.

		USA due to the characteristics of their health system.	
Finding 13: Some women reported issues related to the quality and continuity of their care. Women perceived that their care was not tailored to their individual needs, and complained about the lack of continuity of carer.	390,393–395,398,400,403	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 4/7 had a poor justification of their choice of research design • 4/7 poor reporting of recruitment strategy. • 6/7 no reflexivity statement. • 3/7 data analysis method poorly discussed or justified. • 0/7 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	High confidence.

Cultural aspects summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 14: Women from different cultural backgrounds reported issues related to cultural inappropriateness or language barriers. This included discrimination and the perceived need to justify the right to care due to immigration status.	390,392,402,403,393–396,398–401	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 6/12 had a poor justification of their choice of research design • 7/12 studies recruitment strategy poorly described. • 9/12 no reflexivity statement. • 7/12 data analysis method poorly discussed or justified. • 1/12 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. This finding applies to minority groups in the pregnant population.	High confidence. Despite the minor concern regarding relevance, this finding explicitly states what type of population it applies to.
Finding 15: Immigrant women delayed care seeking due to fear of deportation and increased costs of care. This made them believe that they	399,400	**Methodological limitations:** Serious concerns regarding methodological limitations. • 2/2 had a poor justification of their choice of research design	Moderate concerns. Despite the reported concerns in most of the parameters measured, these studies focused on a very particular sample and so we

should only access care when in obstetric emergencies or they go into labour.		• 1/2 studies recruitment strategy poorly described. • 2/2 showed issues with data collection method • 1/2 studies poorly reported ethical implications. • 2/2 data analysis method poorly discussed or justified. • 0/3 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** Moderate concerns. Only two studies contributing data to this finding. **Relevance:** Minor concerns. The sample of these studies is very specific and hence this finding is not generalisable to the whole pregnant population	consider that the finding is relevant for the overall findings of this meta-synthesis.
Finding 16: Immigrant women found that having dedicated clinics for them was crucial for their care, as they felt safe and reassured when attending.	399,400	**Methodological limitations:** Serious concerns regarding methodological limitations. • 2/2 had a poor justification of their choice of research design • 1/2 studies recruitment strategy poorly described. • 2/2 showed issues with data collection method	Moderate confidence. Despite the reported concerns in most of the parameters measured, this studies focused on a very particular sample and so we consider that the finding is relevant for the overall findings of this meta-synthesis.

		• 1/2 studies poorly reported ethical implications. • 2/2 data analysis method poorly discussed or justified. • 0/3 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** Moderate concerns. Only two studies contributing data to this finding. **Relevance:** Minor concerns. The sample of this studies is very specific and hence this finding is not generalisable to the whole pregnant population.	

Social position and lifestyle factors summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 18: Certain issues related to social position and lifestyle factors also acted as barriers to access antenatal care. In these cases, women did not prioritise antenatal care as they did not have their basic needs of survival covered. These included immigrant status, homelessness, intimate partner violence, childcare difficulties, incompatibility with work hours, living in deprived areas or having low economic resources.	390–395,401–403	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 4/9 had a poor justification of their choice of research design • 5/9 studies recruitment strategy poorly described. • 8/9 no reflexivity statement. 8/9 studies poorly reported ethical implications. • 5/9 data analysis method poorly discussed or justified. • 0/6 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. This finding only applied to women from certain socioeconomic backgrounds.	Moderate confidence
Finding 19: The discrimination of certain collectives and the institutionalised racism	390,392,394,395,398,404	**Methodological limitations:** Minor concerns regarding methodological limitations.	High confidence.

at a societal level acted as barriers to access antenatal care due to fear of repercussions or discrimination. These included teenage pregnancies, substance users, women from ethnic minorities and women with lower socioeconomic status.		• 2/6 had a poor justification of their choice of research design • 3/6 studies recruitment strategy poorly described. • 4/6 no reflexivity statement. 6/6 studies poorly reported ethical implications. • 2/6 data analysis method poorly discussed or justified. • 0/6 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** Minor concerns. This finding does not apply to the whole pregnant population.	
Finding 20: Women with more favourable socioeconomic positions were more likely to attend antenatal care in a timely manner, as they were able to prioritise their care. This included higher economic status, higher education level,	390,392,403	**Methodological limitations:** Minor concerns regarding methodological limitations. • 3/3 no reflexivity statement. 3/3 studies poorly reported ethical implications. • 3/3 data analysis method poorly discussed or justified. • 0/3 studies borderline score of 10 **Coherence:** No or very minor concerns.	Moderate confidence.

steady living situation, older age and being a first time mother.		**Adequacy:** Moderate concerns. Only three studies contributing data to this finding. **Relevance:** Minor concerns. This finding only applies to women with middle and high socioeconomic status.	

Social support and community network summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 21: For many women, the lack of social support acted as a barrier to access antenatal care. This was associated with a lack of disclosure to avoid disapproval withing the women's community or family.	390,392,394,399–401	**Methodological limitations:** Serious concerns regarding methodological limitations. • 3/6 had a poor justification of their choice of research design • 3/6 studies recruitment strategy poorly described. • 3/6 showed issues regarding data collection method. • 3/6 no reflexivity statement. • 5/6 data analysis method poorly discussed or justified. • 1/6 studies borderline score of 10 **Coherence:** No or very minor concerns. **Adequacy:** No or very minor concerns. **Relevance:** No or very minor concerns.	Moderate confidence.

Finding 22: Having the support of the partner, family and friends and a positive reaction to the disclosure of the pregnancy acted as a facilitator for women to seek and engage with antenatal care.	390,392,394,399–401,686	**Methodological limitations:** Serious concerns regarding methodological limitations. • 4/7 studies recruitment strategy poorly described. • 4/7 no reflexivity statement. • 6/7 data analysis method poorly discussed or justified. • 1/7 studies borderline score of 10 **Coherence:** No or very minor concerns **Adequacy:** No or very minor concerns **Relevance:** No or very minor concerns	Moderate confidence

Transportation issues summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 23: The lack of availability and cost of transportation acted as a environmental barrier for women to attend antenatal care.	391,395,398,399	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/4 had a poor justification of their choice of research design • 3/4 studies recruitment strategy poorly described. • 4/4 no reflexivity statement. 3/4 studies poorly reported ethical implications. • 2/4 data analysis method poorly discussed or justified. • 0/4 studies borderline score of 10 **Coherence:** No or very minor concerns **Adequacy:** No or very minor concerns **Relevance:** No or very minor concerns	High confidence.

Lack of health literacy and reproductive knowledge summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 24: Many women reported delays in the recognition of their pregnancy due to a lack of health literacy and reproductive knowledge.	390,392,393,395,398	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/5 had a poor justification of their choice of research design • 2/5 studies recruitment strategy poorly described. • 3/6 showed issues regarding data collection method. • 5/5 no reflexivity statement. 4/5 studies poorly reported ethical implications. • 2/5 data analysis method poorly discussed or justified. • 1/6 studies borderline score of 10 **Coherence:** Minor concerns. In some instances, the data suggested that the lack of recognition could have been due to psychological reasons, such as lack of acceptance, rather than lack of knowledge. **Adequacy:** No or very minor concerns **Relevance:** No or very minor concerns	High confidence.

Government level issues summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 25: Some of the limitations related to the healthcare system can be attributed to limitations of financial resources at a government level. Additionally, the cost of care in some countries and the limitations within the government insurance programs were reported as barriers.	392,394,395,398,399,401,404	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/6 had a poor justification of their choice of research design • 3/6 studies recruitment strategy poorly described. • 3/6 showed issues regarding data collection method. • 3/6 no reflexivity statement. 5/6 studies poorly reported ethical implications. • 3/6 data analysis method poorly discussed or justified. • 1/6 studies borderline score of 10 **Coherence:** Moderate concerns. Although there is plenty of data supporting the limitations within the healthcare system, the association with the limited funding is less clear. **Adequacy:** No or very minor concerns **Relevance:** No or very minor concerns	Moderate confidence.

Callaghan, M., Buller, A.M., Nightingale, F., Nightingale, F., 2011. Understanding 'late bookers' and their social circumstances. Br. J. Midwifery 19, 7–13.

Daniels, P., Noe, G.F., Mayberry, R., 2006. Barriers to prenatal care among black women of low socioeconomic status. Am. J. Health Behav. 30, 188–198. https://doi.org/10.5993/AJHB.30.2.8

Docherty, A., Bugge, C., Watterson, A., 2012. Engagement: An indicator of difference in the perceptions of antenatal care for pregnant women from diverse socioeconomic backgrounds. Heal. Expect. https://doi.org/10.1111/j.1369-7625.2011.00684.x

Downe, S., Finlayson, K., Walsh, D., Lavender, T., 2009. "Weighing up and balancing out": A meta-synthesis of barriers to antenatal care for marginalised women in high-income countries. BJOG An Int. J. Obstet. Gynaecol. 116, 518–529. https://doi.org/10.1111/j.1471-0528.2008.02067.x

Funge, J.K., Boye, M.C., Johnsen, H., Nørredam, M., 2020. "No Papers. No Doctor": A Qualitative Study of Access to Maternity Care Services for Undocumented Immigrant Women in Denmark. Int. J. Environ. Res. Public Health 17, 6503. https://doi.org/10.3390/ijerph17186503

Haddrill, R., Jones, G.L., Mitchell, C.A., Anumba, D.O.C., 2014. Understanding delayed access to antenatal care: A qualitative interview study. BMC Pregnancy Childbirth. https://doi.org/10.1186/1471-2393-14-207

Hatherall, B., Morris, J., Jamal, F., Sweeney, L., Wiggins, M., Kaur, I., Renton, A., Harden, A., 2016. Timing of the initiation of antenatal care: An exploratory qualitative study of women and service providers in East London. Midwifery 36, 1–7. https://doi.org/10.1016/j.midw.2016.02.017

Johnson, J.L., Primas, P.J., Coe, M.K., 1994. Factors that Prevent Women of Low Socioeconomic Status from Seeking Prenatal Care. J. Am. Acad. Nurse Pract. 6, 105–111. https://doi.org/10.1111/j.1745-7599.1994.tb00924.x

Larsson, Å., Wärnå-Furu, C., Näsman, Y., 2017. The meaning of caring in prenatal care from Swedish women's perspectives. Scand. J. Caring Sci. 31, 702–709. https://doi.org/10.1111/scs.12383

Mehta, P.K., Carter, T., Vinoya, C., Kangovi, S., Srinivas, S.K., 2017. Understanding High Utilization of Unscheduled Care in Pregnant Women of Low Socioeconomic Status. Women's Heal. Issues 27, 441–448. https://doi.org/10.1016/j.whi.2017.01.007

Meyer, E., Hennink, M., Rochat, R., Julian, Z., Pinto, M., Zertuche, A.D., Spelke, B., Dott, A., Cota, P., 2016. Working Towards Safe Motherhood: Delays and Barriers to Prenatal Care for Women in Rural and Peri-Urban Areas of Georgia. Matern. Child Health J. https://doi.org/10.1007/s10995-016-1997-x

Murphy Tighe, S., 2010. An exploration of the attitudes of attenders and non-attenders towards antenatal education. Midwifery. https://doi.org/10.1016/j.midw.2008.06.005

Roman, L.A., Raffo, J.E., Dertz, K., Agee, B., Evans, D., Penninga, K., Pierce, T., Cunningham, B., VanderMeulen, P., 2017. Understanding Perspectives of African American Medicaid-Insured Women on the Process of Perinatal Care: An Opportunity for Systems Improvement. Matern. Child Health J. https://doi.org/10.1007/s10995-017-2372-2

Ross, E., 2012. Maternal – fetal attachment and engagement with antenatal advice. Br. J. Midwifery 20.

Sami, J., Quack Lötscher, K.C., Eperon, I., Gonik, L., Martinez de Tejada, B., Epiney, M., Schmidt, N.C., 2019. Giving birth in Switzerland: a qualitative study exploring migrant women's experiences during pregnancy and childbirth in Geneva and Zurich using focus groups. Reprod. Health 16, N.PAG-N.PAG. https://doi.org/10.1186/s12978-019-0771-0

Tighe, S.M., 2010. An exploration of the attitudes of attenders and non-attenders towards antenatal education. Midwifery 26, 294–303. https://doi.org/10.1016/j.midw.2008.06.005

Ward, T.C.S., Mazul, M., Ngui, E.M., Bridgewater, F.D., Harley, A.E., 2013. "You learn to go last": Perceptions of prenatal care experiences among African-American women with limited incomes. Matern. Child Health J. 17, 1753–1759. https://doi.org/10.1007/s10995-012-1194-5

Supplementary file 4.4 - Adapted from Noblit and Hare (1988)

Phases of meta-ethnography(1,2)	
Phase 1: Getting started	Identify intellectual interest
Phase 2: Deciding what is relevant to the initial interest.	Deciding what studies or accounts are relevant by doing an exhaustive search.
Phase 3: Reading the studies	This phase involves the repeated reading of the accounts and the noting of metaphors.
Phase 4: Determining how the studies are related	Determining the relationships between the studies to be synthesised. The authors suggest to create a list of key metaphors, phrases, ideas and/or concepts (and their relations) used in each account and to juxtapose them.
Phase 5: Translating the studies into one another	This process involves maintaining the central concepts or metaphors of each account and comparing both metaphors and their interactions in one account with the metaphor or concepts and their interactions in the other accounts.
Phase 6: Synthesizing translations.	Making a whole into something more than the parts. This is a second level of analysis, where the various translations can be compared with one another with the aim of providing a fresh interpretation.
Phase 7: expressing the synthesis	Choosing the ways to communicate the research.

1. Britten N, Campbell R, Pope C, Donovan J, Morgan M, Pill R. Using meta ethnography to synthesis qualitative research. J Heal Serv Res Policy 2002; 7: 209–15.

2. Noblit GW, Hare DR. Meta-ethnography: synthesizing qualitative studies. Newbury Park, California: Sage, 1988.

Supplementary file 5.1 – eMERGE reporting criteria for meta-ethnographies

No.	Criteria Headings	Reporting Criteria
Phase 1—Selecting meta-ethnography and getting started		
Introduction		
1	Rationale and context for the meta-ethnography	Describe the gap in research or knowledge to be filled by the meta-ethnography, and the wider context of the meta-ethnography Introduction – paragraph 1,2,3
2	Aim(s) of the meta-ethnography	Describe the meta-ethnography aim(s) Introduction – paragraph 5
3	Focus of the meta-ethnography	Describe the meta-ethnography review question(s) (or objectives) Introduction – paragraph 5
4	Rationale for using meta-ethnography	Explain why meta-ethnography was considered the most appropriate qualitative synthesis methodology Introduction – paragraph 5
Phase 2—Deciding what is relevant		
Methods		
5	Search strategy	Describe the rationale for the literature search strategy Section 2.1 – paragraph 1
6	Search processes	Describe how the literature searching was carried out and by whom Section 2.2 – Paragraph 1
7	Selecting primary studies	Describe the process of study screening and selection, and who was involved Section 2.2 – Paragraph 1
Findings		
8	Outcome of study selection	Describe the results of study searches and screening Results - Section 3.1
Phase 3—Reading included studies		

No.	Criteria Headings	Reporting Criteria
Methods		
9	Reading and data extraction approach	Describe the reading and data extraction method and processes Section 2.3.1 Section 2.3.3 – Phase 3
Findings		
10	Presenting characteristics of included studies	Describe characteristics of the included studies Results – Section 3.2
Phase 4—Determining how studies are related		
Methods		
11	Process for determining how studies are related	Describe the methods and processes for determining how the included studies are related: - Which aspects of studies were compared AND - How the studies were compared Section 2.3.3 – Phase 4
Findings		
12	Outcome of relating studies	Describe how studies relate to each other Section 4 - Synthesis
Phase 5—Translating studies into one another		
Methods		
13	Process of translating studies	Describe the methods of translation**:** - Describe steps taken to preserve the context and meaning of the relationships between concepts within and across studies- Describe how the reciprocal and refutational translations were conducted- Describe how potential alternative interpretations or explanations were considered in the translations Section 2.3.3 – Phase 5
Findings		
14	Outcome of translation	Describe the interpretive findings of the translation.

No.	Criteria Headings	Reporting Criteria
		Section 4 – Synthesis
Phase 6—Synthesizing translations		
Methods		
15	Synthesis process	Describe the methods used to develop overarching concepts ("synthesised translations")Describe how potential alternative interpretations or explanations were considered in the synthesis Section 2.3.3 – Phase 6
Findings		
16	Outcome of synthesis process	Describe the new theory, conceptual framework, model, configuration, or interpretation of data developed from the synthesis Section 4 – Synthesis Section 5 – Discussion
Phase 7—Expressing the synthesis		
Discussion		
17	Summary of findings	Summarize the main interpretive findings of the translation and synthesis and compare them to existing literature Section 5 – Paragraph 1
18	Strengths, limitations, and reflexivity	Reflect on and describe the strengths and limitations of the synthesis: - Methodological aspects—for example, describe how the synthesis findings were influenced by the nature of the included studies and how the meta-ethnography was conducted.- Reflexivity—for example, the impact of the research team on the synthesis findings Section 5 – Paragraph 9
19	Recommendations and conclusions	Describe the implications of the synthesis Section 5 – Paragraph 10,11,12

Supplementary file 5.2 – Example of Search

Database: Web of Science **Date: 29/03/2019**	
	Search Terms
Concept 1 Facilitators and Barriers	
21.	TO Facilitator*
22.	TO Barrier*
23.	TO Promoter*
24.	TO Benefit*
25.	TO Attitude*
26.	TO Opportunit*
27.	TO Determinant*
28.	TO Promotion
29.	TO Intention
30.	TO Education
31.	TO Initiative
32.	TO Prevention
33.	TO Pregnancy
34.	**S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 AND S13**
Concept 2 Smoking	
35.	TO Smoking Cessation
36.	TO Smoking
37.	TO Nicotine
38.	**S15 AND S16 AND S17**
39.	**S14 AND S18**
Concept 3 Alcohol Consumption	
40.	TO Alcohol
41.	[redacted]
42.	TO Alcohol Abuse
43.	TO Alcohol drinking
44.	**S20 OR S21 OR S22 OR S23**
45.	**S14 AND S24**
Concept 4 Drug consumption	
46.	TO “Drug abuse”
47.	TO Drug*
48.	TO “Oral drugs”
49.	TO “Intravenous drugs”
50.	**S26 AND S27 OR S28 OR S29**
51.	**S14 AND S30**
Notes Limiters: English Language	

Supplementary file 5.3 – Characteristics of included studies

Reference	Arborelius, et al. (1997)	Barbosa-Leiker, C. et al. (2020)	Bauld, L., et al., (2017)	Borland, T., et al. (2013)	Britton, G. R., et al., (2017)	Crawford-Williams, F., et al. (2015)	Dunn, C. L., et al. (1998)	Fergie L, et al (2019)
Country	Sweden	USA	UK	Canada	USA	Australia	USA	UK
Year of publication	1997	2019	2017	2013	2017	2015	1998	2019
Aims	Understand how women with low educational attainment perceived and experienced the fact of their smoking during pregnancy	Identify women's perceptions of risks and benefits of cannabis use during pregnancy and postpartum, in a state that has legalized recreational cannabis.	Explore the barriers to and facilitators of smoking cessation experienced by women during pregnancy and post-partum	Improving Ontario's cessation system to better meet the needs of pregnant and postpartum women who smoke	Increase our understanding of the expriences of pregnant smokers, including motivation to quit	Identify gaps in knowledge about the effects of alcohol use in pregnancy among pregnant women.	Gain further insight into issues underlying smoking during pregnancy in a low-income, ethnically diverse population	To gain pregnant women's opinions on techniques which they found to be, or think could be, useful in helping overcome barriers and enhancing facilitators towards achieving cessation
Design	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study	Qualitative study
Data collection method	Semi-structured interviews.	Semi-structured interviews	Semi-structured interviews.	Semi-structured interviews. Face to face and telephone.	Focus groups.	Focus groups.	Nine focus groups.	Telephone semi-structured interviews

Recruitment	Purposive-sampling of smoking women from one maternity clinic.	Online forums and community flyers were used.	Purposive sampling from smoking cessation service.	Purposive sampling from smoking cessation service.	Purposive-sampling of smoking women from one maternity unit.	Purposive-sampling of smoking women from one children's and university hospital.	Local residents were hired and trained to recruit women from their own background within the community	Participants were identified through antenatal clinics
Consent process	Consent obtained. Form not specified.	Written informed consent	Written informed consent	No info	Both verbal and written informed consent	Written informed consent	Written informed consent	Written informed consent
Nº of participants	13	19	41	29	21	21	57	12
Age	20-38	18-39	16-42	15-49	17-36	23-40	<15 to 35	19-40
Pregnant	Post-partum	Pregnant or up to 3 months postpartum	Pregnant	Pergnant or up to one year postpartum	Pregnant	Pregnant or up to one year postpartum	Pregnant or delivered within last 6 months before study	Pregnant
Substance	Smoking	Cannabis	Smoking	Smoking	Smoking	Smoking	Smoking	Smoking
Timing of data collection	Two month period in 1994	No info	Dember 2013	February - May 2011	No info	February-May 2014	Not reported	No info
Method of data analysis	Thematic analysis	Qualitative content analysis	Thematic analysis	Thematic interpretive analysis	Delphi consensus process with results from six independent analysis.	Thematic analysis	Theme identification and agreement on coding scheme.	Thematic analysis
CASP score	15	15	17	17	16	15	16	17

Supplementary file 5.4a – Summary of barriers

BARRIERS					Alcohol	Illicit drug use	Smoking
Internal factors	Perceived incentives	Perceived disadvantages of remaining substance free	Fear of failure, frustration				(1)
			Fear of gaining too much weight				(2,3)
			Idea that increased stress from abstaining can harm the baby		(4,5)		
			Loss of stress coping strategy		(6)		
			Negative effects over relationships/feeling excluded		(1,7–9)		
			Withdrawal symptoms / high cravings		(5)		
			Mental health issues as consequence of abstinence			(2,3,8,9)	
			Perception that life is dull without substances		(10)		
		Reported reasons for substance use	Lack of motivation to quit		(11)		
			Boredom		(3,9,12)		
			Lack of alternative coping strategy		(10)		
			Peer pressure		(6,11)		
			Having other users in the household		(8)		
			Physiological addiction			(11)	
			Perceived benefits obtained from substance use				
					(11)		
						(7,8,12–14)	
						(2,8,9,12)	

				(4)			
	Feelings of shame and guilt						(5,6,15)
External factors	Knowledge and information	Using anecdotal evidence as to justify substance use				(3,9)	
		Poor understanding of risks or recommendations				(4,16)	
		Misconceptions about risk	Belief that consequences are fixable			(8,16)	
			Belief that using the substance in moderation is safe				(1,2,8,14,17)
			Belief that substances are only dangerous during the first trimester of pregnancy			(6,11)	
			Considering some substances more dangerous than others		(2,7,8,10,12–14)		
			Belief that consequences of quitting are worse than consequences of use		(4,5,10)		
			Perceiving risk as exaggerated/not having enough evidence of risk			(18)	
			Relaxing attitude towards substance use while pregnancy progresses		(4,5)		
			Greater concerns about dramatic short-time effects than long-term permanent ones		(19)		
			Belief that soothing properties of substances will benefit the baby		(2,3,7–10,12–14,17,20)		
			Not feeling susceptible to potential harms of substance abuse		(5)		
		Sources and quality of information	Receiving inconsistent information			(7–9,12,13,18)	
			Superficial discussion with healthcare professionals		(5)		
			Lack of sources of information		(19)		
			Perceived lack of scientific evidence		(13)		

	Experiences within healthcare system	Negative experiences during antenatal care	Perceived judgement and stigmatization from healthcare professionals	(4,16)		
			Authoritarian and coercive style	(16)		
			Lack of encouragement / insensitive attitude	(3,9)		
			Perceived prioritising of baby's needs and health			(3,9)
			Communication issues	(4)		
		Limitation of cessation treatments	Limit in number of children they can go with them	(21)		
			Being rejected due to pregnancy	(2,3,9,12,13)		
			Pre-admission requirements and cost	(4,5)		
			Perceived as not helpful	(6,11,16,19)		
			Source of additional stress	(2,8,9,12,13,18)		
			Transportation issues	(4,5,10)		
			Not tailored to individual needs	(11,16,19)		
			Difficult referal process	(3,12)		
		Relationship with staff at cessation treatment centres	Bias towards patients		(6)	
			Patients perceived as challenging	(8,12)		
			Perceived as hopeless and beyond help	(4,5,10)		
			Judgemental attitudes	(6,11)		
	Negative influence of social environment	Disadvantages of social disapproval	Feeling questioned, judged, labelled and stigmatised	(2,9,18)		
			Substance use socially perceived as lack of will / not care for baby	(4)		
			Being pressured causing additional stress			
			Pressure perceived as a challenge	(4,6,16)		
			Perceived need to hide and avoid disclosure	(2,6,11,16,19)		
			Conducting the behaviour in public seen as less acceptable than in private	(8,9)		

		Influence of partner, family and social circle	Family and friends as source of information		(6,11)	
			Social substance use	(8,9)		
			Substance use as normalised event at work	(5)		
			Substance use as normalised event at home	(6,11,21)		
			Light substance abuse encouraged	(2,8,9,12,13,18)		
			Partner substance abuse behaviour	(4)		
		Socioeconomic factors – competing priorities				
		Concerns about legal consequences of substance use				(6)

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2. Dunn CL, Pirie PL, Lando HA. Attitudes and perceptions related to smoking among pregnant and postpartum women in a low-income, multiethnic setting. Am J Heal Promot. 1998;12(4):267–74.

3. Nichter M, Nichter M, Adrian S, Goldade K, Tesler L, Muramoto M. Smoking and Harm-Reduction Efforts Among Postpartum Women. Qual Health Res. 2008;18(9):1184–94.

4. Crawford-Williams F, Steen M, Esterman A, Fielder A, Mikocka-Walus A. “My midwife said that having a glass of red wine was actually better for the baby”: a focus group study of women and their partner’s knowledge and experiences relating to alcohol consumption in pregnancy Fiona. BMC Pregnancy Childbirth. 2015;15(79).

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6. Van Scoyoc A, Harrison JA, Fisher PA. Beliefs and Behaviors of Pregnant Women with Addictions Awaiting Treatment Initiation. Child Adolesc Soc Work J. 2017;34:65–79.

7. McLeod D, Benn C, Pullon S, Viccars A, White S, Cookson T, et al. The midwife’s role in facilitating smoking behaviour change during pregnancy. Midwifery. 2003;19:285–97.

8. Britton GR, Collier R, McKitrick S, Sprague LM, Rhodes-Keefe J, Feeney A, et al. CE: Original research: The experiences of pregnant smokers and their providers. Vol. 117, American Journal of Nursing. 2017. p. 24–34.

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12. Bauld L, Graham H, Sinclair L, Flemming K, Naughton F, Ford A, et al. Barriers to and facilitators of smoking cessation in pregnancy and following childbirth: Literature review and qualitative study. Health Technol Assess (Rockv). 2017;21(36):V–158.

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14. Gamble J, Grant J, Tsourtos G. Missed opportunities: A qualitative exploration of the experiences of smoking cessation interventions among socially disadvantaged pregnant women. Women and Birth [Internet]. 2015;28(1):8–15. Available from: http://dx.doi.org/10.1016/j.wombi.2014.11.003

15. Jessup MA, Humphreys JC, Brindis CD, Lee KA. Extrinsic barriers to substance abuse treatment among pregnant drug dependent women. J Drug Issues. 2003;33(2):285–304.

16. Sheridan K. Substance-Using Pregnant and Parenting Adolescent Girls: Considerations for Preventive Intervention Using a Resilience Lens. J Soc Work Pract Addict [Internet]. 2018;18(2):128–52. Available from: https://doi.org/10.1080/1533256X.2018.1448278

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18. Grant A, Morgan M, Gallagher D, Mannay D. Smoking during pregnancy, stigma and secrets: Visual methods exploration in the UK. Women and Birth [Internet]. 2018;33(1):70–6. Available from: https://doi.org/10.1016/j.wombi.2018.11.012

19. Barbosa-Leiker C, Burduli E, Smith CL, Brooks O, Orr M, Gartstein M. Daily Cannabis Use During Pregnancy and Postpartum in a State With Legalized Recreational Cannabis. J Addict Med. 2020 Dec;14(6):467–74.

20. Pletsch PK, Morgan S, Pieper AF. Context and beliefs about smoking and smoking cessation. MCN, Am J Matern Nurs. 2003;28(5).

21. Morris M, Seibold C, Webber R. Drugs and having babies: An exploration of how a specialist clinic meets the needs of chemically dependent pregnant women. Midwifery. 2012;28:163–72.

22. Herbec A, Beard E, Brown J, Gardner B, Tombor I, West R. The needs and preferences of pregnant smokers regarding tailored internet-based smoking cessation interventions: A qualitative interview study. Vol. 14, BMC Public Health. 2014.

Supplementary file 5.4b – Summary of facilitators

Facilitators				Alcohol	Illicit drugs	Smoking
Internal facilitators	Incentives to remain substance free	Perceived benefits of remaining substance free	Benefit for baby's and woman's health	(1,2)		
			Increased perception of willpower and self-efficacy	(3)		
			Hygiene benefits	(4–9)		
			Relief from guilt of social pressure		(3)	
			Money saving	(4,8,10)		
			Raising kids in a substance-free environment			(11)
		Perceived disadvantages of substance use	Cost			(5,8)
			Addiction			(4,6)
			Risk for baby's health			(7)
			Negative example for children			(11)
		Pregnancy and motherhood as motivation to quit	Concerns about the health of the baby			(6,7,11–13)
			Putting baby's interest first	(1,2)		
			Perceiving abstinence as a time limited experience		(7,11)	
			Feeling moral responsibility for the baby			(7)
			Pregnancy sickness as barrier for substance use	(1,3)		

			Having children as motivation to quit	(3,14–16)		
External facilitators	Knowledge and information	Having accurate knowledge about the risks			(1,3,8,9,11,17)	
		Making efforts to minimise harms of substance use	Trying to compensate with other health behaviours	(2,13)		
			Trying to cut down or quit	(3)		
			Switching substances to what it's considered safer			
			Complying with antenatal care, child welfare	(2)		
			Seeking care			
			Using distractions	(1,2)		
			Limiting relationships with other users	(3)		
			Information seeking behaviour	(1,2,8,18)		
	Experiences within healthcare system	Being encouraged to remain substance free			(13)	
		Trusting healthcare providers				(4,8,9,13)
		Empathetic, personalised approach				
		Supportive, non-judgemental relationship			(1–3,13)	
		Women centred experience			(3,14,15)	
		Finding help in replacement therapies			(3,4,7–9,11–13,18,19)	
			Social disapproval as motivation to quit	(1)		

	Influence of social environment	Benefits of social disapproval	Work environment that discourages substance use	(14)		
			Quitting understood as family decision, not only the women's	(8,9,17)		
		Supportive close environment	Support from partner		(14,16)	
			Being encouraged positively by social circle	(4,6–9,17)		

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9. Fergie L, Coleman T, Ussher M, Cooper S, Campbell KA. Pregnant Smokers' Experiences and Opinions of Techniques Aimed to Address Barriers and Facilitators to Smoking Cessation: A Qualitative Study. Int J Environ Res Public Health. 2019 Aug;16(15).

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15. Leppo A. " Subutex is safe": Perceptions of risk in using illicit drugs during pregnancy. Int J Drug Policy [Internet]. 2012;23(5):365–73. Available from: http://dx.doi.org/10.1016/j.drugpo.2012.01.004

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21. Herbec A, Beard E, Brown J, Gardner B, Tombor I, West R. The needs and preferences of pregnant smokers regarding tailored internet-based smoking cessation interventions: A qualitative interview study. Vol. 14, BMC Public Health. 2014.

22. Morris M, Seibold C, Webber R. Drugs and having babies: An exploration of how a specialist clinic meets the needs of chemically dependent pregnant women. Midwifery. 2012;28:163–72.

Supplementary file 5.5 – Qualitative Appraisals

Reference	Clear aims	Qualitative methodology appropiate	Research design appropiate to meet aims	Recruitment strategy appropiate	Data collected in a way that addressed the research issue	Relationship between researcher and participants considered	Ethical issues	Data analysis rigorous	clear statement of findings	How valuable is the research	Total Score
Herbec, A., et al (2014).	2	2	1	1	1	1	1	2	2	2	15
Barbosa-Leiker C, et al (2020)	2	2	1	2	1	0	1	2	2	2	15
Fergie L, et al. (2019)	2	2	1	2	2	1	1	2	2	2	17
Bauld, L., et al, (2017).	2	2	2	2	2	0	1	2	2	2	17
Dunn, C. L., et al. (1998)	2	2	2	2	2	0	0	2	2	2	16
Van Scoyoc, et al, (2017)	2	2	2	2	2	2	2	2	2	2	20
Leppo, A. (2012).	2	2	2	2	2	0	2	2	2	2	18

Jessup, M. A., et al. (2003).	2	2	2	1	2	0	1	2	2	1	15
Sheridan, K. (2018).	2	2	2	2	2	0	2	2	1	1	16
Morris, M., et al. (2012).	2	2	2	2	2	2	2	2	2	1	19
Arborelius, E., & Nyberg, K. (1997).	2	2	1	2	2		2	2	1	1	15
Borland, T., et al. (2013).	2	2	2	2	2	0	1	2	2	2	17
Britton, G. R., et al (2017).	2	2	2	2	2	0	1	1	2	2	16
Crawford-Williams, et al. (2015).	2	2	2	1	2	0	1	2	2	1	15
Gamble, J., Grant, J., et al. (2015).	2	2	2	1	2	2	2	2	2		17
Grant A, et al. (2018)	2	2	2	2	2	2	2	1	2	1	18
Grant, A., et al. (2019).	2	2	2	1	2	2	1	2	2	1	17
McLeod, D., et al. (2003).	2	2	0	2	2	0	1	2	2	2	15

Raymond, N., et al. (2009).	2	2	2	1	1	1	2	2	1	2	16
Nichter M, et al. (2008).	2	2	2	2	2	2	1	1	1	2	17
Nichter M, et al. (2007). Ethnographic analysis	2	2	2	1	2	1	1	2	2	2	17
Pletsch P.K., et al. (2003)	2	2	1	1	2	0	1	2	2	2	15

Supplementary file 5.6 – Grade CerQual Analysis

Perceived Incentives summary of review finding	Studies contributing to the review finding	Explanation of CERQual assessment	CERQual assessment of confidence in the evidence
Finding 1: Women utilised substance use as a coping strategy with psychological and social benefits.	(Arborelius and Nyberg, 1997; Barbosa-Leiker et al., 2020; Bauld et al., 2017; Britton et al., 2017; Crawford-Williams et al., 2015; Dunn et al., 1998; Fergie et al., 2019; Gamble et al., 2015; Grant et al., 2019, 2018; Jessup et al., 2003; McLeod et al., 2003; Nichter et al., 2008, 2007; Pletsch et al., 2003; Raymond et al., 2009; Sheridan, 2018; Van Scoyoc et al., 2017)	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 7/18 studies recruitment strategy poorly described. • 10/18 no reflexivity statement, 3/18 unclear reflexivity. 11/18 studies poorly reported ethical implications. • 5/18 data analysis method poorly discussed or justified. • 5/18 studies borderline score 15. **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. This finding is very descriptive of the studies related to smoking and alcohol, but this conclusion can only be reached interpretatively from the illicit drug studies.	High confidence
Finding 2: Remaining abstinent meant a big commitment for some	(Borland et al., 2013; Britton et al., 2017; Grant et al., 2019; Leppo, 2012; McLeod et al., 2003;	**Methodological limitations:** Moderate concerns regarding methodological limitations.	High confidence

women, loss of coping-strategy, increased craving and loss of social circle.	Nichter et al., 2008, 2007; Raymond et al., 2009; Sheridan, 2018; Van Scoyoc et al., 2017)	• 3/12 recruitment strategy poorly reported. 7/12 no reflexivity statement, 2/11 unclear reflexivity. • 7/12 studies reported ethical implications poorly. • 2/12 data analysis poorly described. • 1 study borderline score 15. **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns. (all substances, both pregnant and post-partum)	
Finding 3: Women obtained benefits from remaining abstinent. Health, hygiene, money saving, reducing guilt, improving baby's health and giving a good example were the benefits most commonly reported benefits of abstinence.	(Arborelius and Nyberg, 1997; Bauld et al., 2017; Crawford-Williams et al., 2015; Fergie et al., 2019; Grant et al., 2018; McLeod et al., 2003; Nichter et al., 2008, 2007; Sheridan, 2018)	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 4/8 lack reflexivity, 2/8 unclear reflexivity. 5/8 studies reported ethical implications poorly. • 2/8 recruitment poorly described • 2/8 data analysis poorly described • 3/8 borderline score 15 **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. Although very recurrent, these benefits are mostly mentioned on smoking related studies. Alcohol and drug use studies focus on health benefits of their abstinence.	High confidence.

Finding 4: For some women, being pregnant or having other children acted as a motivation to remain substance free or engage in harm-reducing actions.	(Bauld et al., 2017; Crawford-Williams et al., 2015; Fergie et al., 2019; Gamble et al., 2015; Grant et al., 2019, 2018; Nichter et al., 2007; Raymond et al., 2009; Sheridan, 2018)	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/9 study no justification of research design, 1/8 study poor justification research design. • 2/9 poor recruitment strategy reporting. • 4/9 lack reflexivity, 2/9 unclear reflexivity. 6/9 studies reported ethical implications poorly. • 2/9 data analysis poorly reported. • 3/9 borderline score 15 **Coherence:** Minor concerns. Although this was found very often, it was not the case for all women. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. Not equally representation of all substances, mostly smoking.	Moderate confidence

Feelings of shame and guilt summary of review finding	Studies contributing to the review finding	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Finding 5: Women felt guilt and shame due to their substance abuse. These negative feelings increased the women's levels of stress leading to increased substance use.	(Arborelius and Nyberg, 1997; Bauld et al., 2017; Britton et al., 2017; Crawford-Williams et al., 2015; Dunn et al., 1998; Grant et al., 2018; Leppo, 2012; Nichter et al., 2007; Raymond et al., 2009; Sheridan, 2018; Van Scoyoc et al., 2017)	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/6 poor justification research design • 2/6 poor reporting of recruitment • 5/6 lack reflexivity, 1/6 unclear reflexivity. 6/6 studies reported ethical implications poorly. • 1 poor reporting data analysis • 2/6 borderline score 15 **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** Moderate concerns. This data is only present in smoking related studies.	**Moderate confidence**

Knowledge about risks summary of review finding	Studies contributing to the review finding	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Finding 6: Many women engaged with anecdotal evidence about substance use, which may have contributed to misconceptions about risks.	3,444,468,446,447,451,453,460,462,463,467 464 465	**Methodological limitations:** Minor concerns regarding methodological limitations. • 3/13 poor justification research design. • 3/13 poor reporting recruitment strategy • 1/13 poor justification data collection methods • 8/13 lack reflexivity, 3/13 unclear reflexivity. 7/13 studies reported ethical implications poorly. • 2/13 poor reporting data analysis • 3/13 borderline score 15 **Coherence:** No concerns. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	High confidence
Finding 7: Having specific and accurate knowledge about the risks of substance use acted as a facilitators to prevent	3,444,464,466–468,446–448,451,453,460,462,463	**Methodological limitations:** Moderate concerns regarding methodological limitations.	Moderate confidence

substance use during pregnancy and facilitates help-seeking.		• 7/14 lack reflexivity, 3/14 unclear reflexivity. 6/14 studies reported ethical implications poorly. • 4/14 poor recruitment reporting • 2/14 poor justification research design • 1/14 poor justification data collection method. • 2/14 poor reporting data analysis method. • 2/14 borderline score 15 • **Coherence:** Medium concerns. Although true in many cases, some studies show that despite having knowledge, some women engage in substance use behaviours. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	
Finding 8: Having some knowledge and information about the risks of substance use led some women to engage in harm-reduction strategies.	444,446,463,466–469,447–451,453,460,462	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/15 Poor justification of research design. • 7/15 poor reporting recruitment strategy	Moderate confidence

		• 2/15 poor justification data collection methods • 8/15 lack reflexivity, 3/15 unclear reflexivity. 9/15 studies reported ethical implications poorly. • 2/15 poor reporting data analysis • 4/15 borderline score 15 • **Coherence:** Minor concerns. Although women's efforts to reduce harms are evident in most of the studies, the link with having knowledge and information is only evident in a few of them. However, many women speak about making this decisions after speaking with HCP or doing their own research. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	

Sources and quality of information summary of findings	Studies contributing to the review finding	CERQual assessment of confidence in the evidence	Explanation of CERQual assessment
Finding 9: Women perceived a lack of sources of information and evidence regarding risks associated with substance abuse, and found that information conflicting in many instances.	444,445,465–467,469,446–449,451,461–463	**Methodological limitations:** Minor concerns regarding methodological limitations. • 1/13 no justification research design, 2/13 poor justification research design. • 6/13 poor reporting recruitment strategy • 2/13 poor justification data collection methods • 7/13 lack reflexivity, 3/13 unclear reflexivity. 10/13 studies reported ethical implications poorly. • 2/13 poor reporting data analysis • 4/13 borderline score 15 **Coherence:** No concerns. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	High confidence
Finding 10: Women were not satisfied with the level and quality of discussion they had with their healthcare professionals about substance use during antenatal care.	444,446–448,461–463,465,466,468	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 2/10 studies with no justification of research design. • 3/10 poor reporting recruitment strategy	High confidence

		• 6/10 lack reflexivity, 1/10 unclear reflexivity. 8/10 studies reported ethical implications poorly. • 2/10 poor reporting data analysis • 3/10 borderline score 15 **Coherence:** No concerns. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	
Experiences within healthcare system summary of findings	**Studies contributing to the review finding**	**CERQual assessment of confidence in the evidence**	**Explanation of CERQual assessment**
Finding 11: In some occasions women perceived healthcare professionals to be judgemental and stigmatising during their antenatal care, this perception made them feel more stressed and decreased their motivation to remain abstinent.	[3,445,446,448,451,471]	**Methodological limitations:** Minor concerns regarding methodological limitations. • 1/6 poor reporting recruitment strategy • 3/6 lack reflexivity. 2/6 studies reported ethical implications poorly. • 2/6 poor reporting data analysis **Coherence:** Minor concerns. There is data supporting the idea of feeling judge in most of the studies, however the association with increase stress levels is less frequent. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. This finding does not apply to alcohol related studies.	Moderate confidence

Finding 12: Women discussed that coercive and authoritarian communication styles from healthcare professionals acted as barrier to stop using substances.	3,444,448,460,461,471	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/6 no justification research design, 1/5 poor justification research design. • 1/6 poor reporting recruitment strategy • 3/6 lack reflexivity, 3/6 unclear reflexivity. 2/6 studies reported ethical implications poorly. • 1/6 study poor reporting of data analysis. • 2/6 borderline score 15 **Coherence:** No concerns **Adequacy:** No or very limited concerns. **Relevance:** Moderate concerns. This finding relates to smoking and illicit drug use women. There are no discussions about relationship with healthcare professionals in the alcohol related studies.	Moderate confidence
Finding 13: Women reported that finding supporting, encouraging and non-judgemental healthcare professionals facilitated their quitting process.	444,446,468,471,448,449,451,460,461,463,464,466	**Methodological limitations:** Minor concerns regarding methodological limitations. • 5/13 poor justification research design. • 3/13 poor reporting recruitment strategy	Moderate confidence

		• 1/13 poor justification data collection methods • 7/13 lack reflexivity, 4/13 unclear reflexivity. 8/13 studies reported ethical implications poorly. • 2/13 poor reporting data analysis • 3/13 borderline score 15 **Coherence:** No concerns **Adequacy:** No or very limited concerns. **Relevance:** Moderate concerns. This finding relates to smoking and illicit drug use women. There are no discussions about relationship with healthcare professionals in the alcohol related studies.	
Finding 14: Issues in drug cessation treatment centres like not being able to take their children, not accepting pregnant women, not meeting pre-admission requirements and finding the referral system too complex, acted as barriers.	444,445,450	**Methodological limitations:** Minor concerns regarding methodological limitations. • 1/3 poor reporting recruitment strategy • 3/3 lack reflexivity. 3/3 studies reported ethical implications poorly. • 1/3 borderline score 15 **Coherence:** No concerns **Adequacy:** Moderate concerns. Restricted number of studies. However this finding is very specific to illicit drug use.	Low confidence

		Relevance: Moderate concerns. This finding only applies to women using drug cessation treatment for illicit drug use.	

Influence of social environment summary of findings	**Studies contributing to the review finding**	**CERQual assessment of confidence in the evidence**	**Explanation of CERQual assessment**
Finding 15: Being exposed to social disapproval made some women feel questioned, judged and labelled, and perceived as bad mothers. This increased their levels of stress and sometimes was perceived as a challenge to their individuality, acted as a barrier for substance use disclosure and promoted hiding.	3,444,464,465,468,469,471,445–448,450,451,460,461	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 3/15 poor justification research design. 1/13 no justification of research design • 5/15 poor reporting recruitment strategy • 10/15 lack reflexivity. 10/15 studies reported ethical implications poorly. • 1/15 poor reporting data analysis • 4/15 borderline score 15 **Coherence:** Minor concerns. Although generally the case, some women find benefits to the social disapproval as motivator for change. **Adequacy:** No or very limited concerns.	**High confidence**

		Relevance: No or very limited concerns. (Big number of studies and representation of different substances)	
Finding 16: For some women, the perceived social disapproval acted as a motivator for them to remain abstinent, as they wanted to be perceived as good mothers.	[447,466]	**Methodological limitations**: Moderate regarding methodological limitations. • 1/2 poor reporting recruitment strategy • 1/2 lack reflexivity. 2/2 studies reported ethical implications poorly. • 1/2 poor reporting data analysis • 1/2 borderline score 15 **Coherence:** Serious concerns. High amounts of data contradicting this statement, most of women perceived social disapproval negatively. **Adequacy:** Moderate concerns. Only two studies with relatively thin data. **Relevance:** Serious concerns. Limited representation in a small number of studies.	Low confidence

Finding 17: Women found that having a close environment (partner, family and friends) that didn't encourage or support their abstinence made it harder. Women struggled with being exposed to the substance in their normal environments and expressed frustration for being considered the sole responsible for the wellbeing of the baby.	3,444,466,468,469,445–448,451,461,463,464	**Methodological limitations:** Moderate concerns regarding methodological limitations. • 1/12 poor justification research design. • 4/12 poor reporting recruitment strategy • 6/12 lack reflexivity, 2/12 unclear reflexivity. 9/12 studies reported ethical implications poorly. • 2/12 poor reporting data analysis • 2/12 borderline score 15 **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. Studies focusing on smoking are over-represented in the data supporting this finding. However, studies involving alcohol and illicit drug use are also contributing.	**High confidence**
Finding 18: Women spoke about how having a supportive close environment acted as a facilitators for them to remain	444,446,447,461,463,464	**Methodological limitations:** Moderate concerns regarding methodological limitations.	**High confidence**

abstinent. This included supportive partners, family and friends, and environments that discouraged substance use.		• 2/6 poor justification research design. • 2/6 poor reporting recruitment strategy • 4/6 lack reflexivity. 6/6 studies reported ethical implications poorly. • 1/6 poor reporting data analysis • 2/6 borderline score 15 **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** Minor concerns. Studies focusing on smoking are over-represented in the data supporting this finding. However, studies involving alcohol and illicit drug use are also contributing.	
Finding 19: Socioeconomic factors, including lack of childcare, financial issues, food and housing insecurity and domestic violence, were sources of immediate stress for some women and so dealing with their substance abuse was less of a priority.	444,445,471,448,450,451,453,461,463,467,470	**Methodological limitations:** Minor concerns regarding methodological limitations. • 1/11 poor justification research design. 1/11 no justification research design	**High confidence**

		• 4/11 poor reporting recruitment strategy • 7/11 lack reflexivity, 1/11 unclear reflexivity. 6/11 studies reported ethical implications poorly. • 3/11 borderline score 15 **Coherence:** No or very limited concerns. **Adequacy:** No or very limited concerns. **Relevance:** No or very limited concerns.	

Supplementary file 6.1 – Example of search

ExaDatabase: Web of Science **Date:**	
	Search Terms
Concept 1 Facilitators and Barriers	
52.	TO Facilitator*
53.	TO Barrier*
54.	TO Promoter*
55.	TO Benefit*
56.	TO Attitude*
57.	TO Opportunit*
58.	TO Determinant*
59.	TO Promotion
60.	TO Intention
61.	TO Education
62.	TO Initiative
63.	TO Prevention
64.	TO Pregnancy
65.	**S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 AND S13**
Concept 5 Weight Management	
66.	TO Weight management
67.	TO Physical activity
68.	TO Nutrition
69.	TO Overweight
70.	**S33 OR S34 OR S35**
71.	**S14 AND S19**
Notes Limiters: English Language	

Supplementary file 6.2 – Summary table of facilitators and barriers

Themes	Facilitators	Barriers
Theme 1: Awareness and beliefs about weight gain and weight management	**Knowledge and awareness** • Awareness of physical activity recommendations.[1–3] • Awareness of risks of overweight and underweight during pregnancy.[4,5] • Awareness of health benefits of healthy eating and exercise[1,2,11–13,3–10] – easier labour[1–3,5,13,14], feeling of control over pregnant body[1,2], prevent excessive GWG[4], improve baby's well-being[2,5,12,15], increased energy[2], enjoyable activities[2,3], physical and mental health benefits[1–3,5,12,13], socialisation[1,2]. **Risk perception and decision balance** • Having different attitude in second and subsequent pregnancies regarding exercise and nutrition[2,16]. • Positively accepting weight gain during pregnancy[13] • Prioritising food quality[12,15]	**Knowledge and awareness** • Lack of awareness of benefits of exercise during pregnancy[5,9,12] • Lack of awareness on how to safely exercise[1–3,6–8,14] • Lack of nutrition related knowledge – benefits and what's healthy[4,5,15] • Lack of awareness of appropriate range of GWG[11,14,16] • Lack of awareness of specific risks of excessive weight gain[1,5,8,10,11,13,14,17] **Risk perception and decision balance** • Not feeling susceptible to risks – considering risks exaggerated[5,14] • Perception of higher risk associated with smoking or alcohol drinking[10,13] • Perception that pregnancy is risky by nature and complications happen randomly[10]. • Perceiving pregnancy as free pass period to eat anything or overeat[7,10,11,13–17]

	• Concerns about gaining too much weight[5,11,13]
	• Sense of responsibility towards baby[5,6,12,13,16]
	• Becoming a role children for their children[2,10,12]
	• Prioritising healthy eating because of pregnancy[12–16]
	
	Perceived control over health and weight gain
	
	• Established exercise habit before and during pregnancy[7]
	• Established healthy diet habits before and during pregnancy[3,10,15]
	• Motivation
	  ○ Intrinsic motivation[2]
	  ○ Perceived high levels of self-efficacy and confidence[12]
	• Considering oneself able despite pregnancy[11].
		• Idea that unhealthy choices can be compensated[13] – intention to delay improving health habits until postpartum period[7,10]
		• Considering household works as exercise enough[1,3,9,11,14]
		• Considering physical activity as unsafe[1–3,5,7,9,11,13,14]
		• Fear of harming the baby while doing exercise[1–3,5,7,9,11,13,14] (mention body physically challenged[3])
		• Fear of harming the baby by eating too little[14,15].
		
		Perceived control over health and weight gain
		
		• Perceived lack of control over GWG[11,13,14]
		• Acceptance that weight gain is inevitable, excuse to indulge[1,5,11,16]
		• Perception that own control of health is very limited[6,12]
		• Justifying unhealthy choices by eating in moderation[13]
		• Understanding cravings as nutritional needs of baby[10,11,13–15]

- Pregnancy related barriers to exercise
    - Being restricted by HCP / higher risk pregnancies[3,5,7]
    - Lack of energy, tiredness[1–3,5–7,9,12,13,15]
    - Reduced mobility[2,3,7]
    - Sickness and other pregnancy related pains and discomforts[1–3,5,7,9,13,17]
- Pregnancy related barriers to healthy diet
    - Nausea, vomiting and aversion to certain foods[14,16]
    - Making food choices based on cravings, appetite and taste[9,12,13,15,16]
    - Lack of energy to cook[15]
- Lack of exercise routine and poor dietary habits
    - Habit of eating out[9]
    - Snacking[15]
    - Using food as comfort in times of distress[10–12]
- Lack of established exercise routine[2,3,7,9]
- Lack of motivation[1–3,9]
    - Dislike of exercise[3,5]
    - Laziness[2,3,9]
    - Need for external motivation[9]

		Personal insecurities and sensitive nature of the topic • Feelings of shame and guilt • Shame and regret for not being active[7,10] • Guilt for choosing unhealthy foods[4] • Shame because of overweight – problems with body-image, self-steem[1–3,5,17]. • Perceived need to justify dietary behaviours[13] Weight and diet perceived as sensitive topic for both women and healthcare professionals[5,8,10,11,16,17].

Theme 2: Antenatal healthcare	**Interaction with Healthcare Professionals**
	Supportive healthcare professionals[8]
	
	Antenatal education and sources of information
	Advice from healthcare professionals
	• Receiving useful information about nutrition and exercise[2,4,8,16,17]
		Interaction with healthcare professionals
		Insensitive and judgemental attitudes[16]
		• Feeling embarrassed and ashamed by insensitive comments[16]
		  • Feeling stigmatised in dietary specialist clinics[10,14]
		  • Judgemental attitudes from HCP[10]

- Valuing and trusting providers opinions, information and advice[6,8,17].

Culturally sensitive information[8,11]

Access to different sources of information[1,4,8]

- Insensitive and inappropriate communication[16]

Avoidant or defensive attitudes

- Feeling challenged when receiving advice about nutrition[6,9,14].
- Personal disagreement with HCP advice[6].
- Rejecting standards GWG goals[14].
- Actively avoiding conversation about weight[8].

Antenatal education and sources of information

- Receiving conflicting information from different sources[4–7,11]
- Too much information-too many sources[3,6,8,13]
- Perceived lack of prioritising of exercise and nutrition advice[6,7,17] – prioritising of clinical aspects[7,8,16]
- Lack of pre-conceptual education[5]
- Using anecdotal evidence to reduce anxiety[10,14]

	Education received about physical activity • Lack of satisfaction with information received[1,2,5–7,13,17] • Perceived lack of support to become more active[2,5] • Perceiving physical activity advice as being too conservative[6,13] • Having to prompt discussion themselves[6] • Hesitant and unclear recommendations[6,7] Education received about diet and nutrition • Insufficient, scarce advice[4,8] • Lack of discussion with HCP about dietary advice[4,6–8,13] • Advice too generalised – not especific[4,6,8,13,16] • Advice not individualised[5,6,8] • Nutritional advice focused on food safety only[4,8,16,17] • Eating advice confusing and changing – difficulties trying to meet all dietary advice[6]

		• Having to prompt discussion themselves[8]
Theme 3: Social and environmental influence	**Availability of support** • Supportive and encouraging partner, friends and family[1–5,7,9,12,16] • Having positive role models[12] **Environmental and sociodemographic facilitators** • Having good cooking skills[4] • Affordable facilities[3,7] • Availability of transportation or built environment[2] • Right weather conditions[2,3] • High educational level • Higher socioeconomic status • Multiparty and older age[12]	**Influence of others, social support and norms** • Adapting own dietary habits to others[11,12] • Lack of control over shopping or cooking choices[11,14,15] • Cultural differences in dietary habits[8,11,12] • Family encouraging overeating[11,14–16] • Unhealthy habits in supportive circle[11] • Relying on family for advice[2,8,11,16] • Lay advice contradicting professional advice[8,11] • Social pressure to increase self-soothing eating[11,17] • Lack of support and role model[1–3,5,7,11,13] • Social perception of exercise being unsafe[11] • Family and partner encouraging inactivity[3] • Relying on family for advice (both) [2,8,11,16] • Lay advice contradicting professional advice (both) [8,11]

Social judgement and stigmatization[1,5,10,12,17]

- Perceived differentiated treatment compared to other pregnant women[14,17]
- Feeling questions and judged[12–14]

Being exposed to derogatory language[13]

Environmental and sociodemographic barriers

- Affordability and easy access to fast food[9,11–14]
- Healthy food perceived as more expensive and harder to access
- Lack of cooking skills[12]
- lack of nearby healthy shops[5,11,15]
- Lack of exercise programs tailored to pregnant women[1,3,7,13]
- Limited access to sports facilities – affordability issues[1–3,5,7,12,13]
- Neighbourhood safety[2,3]
- Urban environments unsuitable for outdoor exercise[12]
- Weather conditions[2,3,7,13]
- Low income environments[11]

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Supplementary file 6.3 – GRADE CerQual Analysis

Summary of Qualitative Findings Table

#	Summarised review finding	GRADE-CERQual Assessment of confidence	Explanation of GRADE-CERQual Assessment	References
1	Some women lacked knowledge regarding how to manage their weight during pregnancy. This lack of knowledge included lack of awareness of risks, and lack of awareness of recommendations regarding physical activity and dietary advice and the associated benefits.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Groth et al. undefined; Denison et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Reyes et al. undefined; Lee et al. undefined; Sui et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Keely et al. undefined; Holton et al. undefined; Kominiarek et al. undefined;
2	Understanding the value of food quality and benefits of physical activity during pregnancy, as well as being aware of the recommendations, acted as facilitator for some women.	High confidence	Minor concerns regarding methodological limitations, Minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Groth et al. undefined; Denison et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Reyes et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Ferrari et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Keely et al. undefined; Kominiarek et al. undefined;
3	Women engaged in a process of weighing up perceived benefits and potential risks to make choices about their weight management behaviours. Some women perceived the risks of	Moderate confidence	Minor concerns regarding methodological limitations, Minor concerns regarding coherence, No/Very minor concerns regarding	Faucher & Mirabito undefined; Groth et al. undefined; Denison et al. undefined; Flannery et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Keely

	overweight and obesity as exaggerated and felt that unhealthy choices could be compensated.		adequacy, and No/Very minor concerns regarding relevance	et al. undefined; Holton et al. undefined; Kominiarek et al. undefined;
4	Some women expressed concerns of harming their babies when doing exercise, especially in the cases of women with fertility issues or history of pregnancy loss. These fears led women to believe that the potential risks were stronger than the benefits of physical activity, so they decided to avoid it.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and Minor concerns regarding relevance	Groth et al. undefined; Denison et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Kominiarek et al. undefined;
5	Some women expressed feelings of shame and guilt due to their weight. These negative feelings had a pejorative effect over their body-image and self-steem which prevented them from wanting to engage in physical activities. Additionally, such feelings also acted as a barrier for communication and information transmission between women and healthcare professionals as weight management was perceived as a sensitive topic.	High confidence	Minor concerns regarding methodological limitations, Minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Denison et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Keely et al. undefined; Holton et al. undefined; Kominiarek et al. undefined;
6	Women's pre-pregnancy habits played an important role in their weight management behaviours during pregnancy. Women who lacked an exercise routine or had poor dietary habits encountered additional challenges, whereas women who had healthy pre-established habits found it easier to maintain them.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Groth et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Marquez et al. undefined; Keely et al. undefined; Kominiarek et al. undefined;
7	Lack of motivation to healthy eating or physical activity were reported as common barriers which	Moderate confidence	Moderate concerns regarding methodological limitations, Minor	Groth et al. undefined; Denison et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Marquez et al. undefined;

	could be overcome with the influence of external motivations.		concerns regarding coherence, Moderate concerns regarding adequacy, and No/Very minor concerns regarding relevance	
8	Pregnancy symtpoms like tiredness, reduced mobility, or nausea can act as barriers to physical activity and interfere in women's eating behaviours because of potential aversion to foods, cravings, or levels of appetite.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Groth et al. undefined; Denison et al. undefined; Flannery et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Ferrari et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Holton et al. undefined;
9	For some women, being pregnant acted as a motivation to make changes in their habits towards healthier diet and activity behaviours. These women reported that felt a sense of responsibility towards the wellbeing of their babies, they wanted to be healthy for their children and act as role model for their own health behaviours.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Flannery et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; Sui et al. undefined; O'Brien et al. undefined; Ferrari et al. undefined; Padmanabhan et al. undefined; Keely et al. undefined;
10	The social context of the woman has an influence in their weight management behaviours. Some women reported having very limited control over shopping or cooking options, or were exposed to family members who encouraged overeating. Additionally, family members acted as a source of information or advice that sometimes contradicted HCP recommendations.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Flannery et al. undefined; Garnweidner et al. undefined; Reyes et al. undefined; Leiferman et al. undefined; O'Brien et al. undefined; Holton et al. undefined; Kominiarek et al. undefined;

11	The lack of support from family and friends to engage in weight management behaviours acted as a barrier for many women. In some occasions, these women lacked positive role models and were encouraged to rest.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Kominiarek et al. undefined;
12	Having the support of the social context to engage in weight managements behaviours acted as facilitators, especially when women had positive role models throughout all their lifes.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Groth et al. undefined; Denison et al. undefined; Flannery et al. undefined; Lee et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Marquez et al. undefined;
13	Some women expressed feeling judged and stigmatised for being overweight and pregnant. These negative feelings contribute to the perception that weight management is a sensitive topic to discuss, preventing information exchange and advice provision with HCP.	Moderate confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Denison et al. undefined; Sui et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Keely et al. undefined; Holton et al. undefined;
14	Some women encountered insensitive or judgemental HCP during their antenatal care that made them feel embarrassed and stigmatised for being overweight. These led some women to adopt avoidant attitudes, where they avoided to speak about weight management, or defensive attitudes,	Low confidence	Minor concerns regarding methodological limitations, Minor concerns regarding coherence, Moderate concerns regarding adequacy, and Moderate concerns regarding relevance	Faucher & Mirabito undefined; Flannery et al. undefined; Keely et al. undefined;

	where they received advice regarding weight management as a challenge to their individuality.			
15	Women were not satisfied the level of education received regarding weight management during their antenatal care. Some women felt overwhelmed by the amount of information and sources available to them, especially regarding dietary behaviours, which led them to find contradictive information in some cases. Information was too hesitant or conservative, and lacked specific advice.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Ferrari et al. undefined; Padmanabhan et al. undefined; Holton et al. undefined;
16	Women who received information and advice regarding physical activity and diet during pregnancy were more able to engage in weight management behaviours. These women valued their HCP opinions and engaged actively in information seeking behaviours.	Moderate confidence	Minor concerns regarding methodological limitations, Moderate concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Flannery et al. undefined; Garnweidner et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Ferrari et al. undefined; Holton et al. undefined;
17	Environmental factors such as accessibility to healthy shops or sports facilities and affordability of fast food, and lack of time or other commitments also had a negative influence on women's physical activity and dietary habits.	High confidence	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Groth et al. undefined; Denison et al. undefined; Reyes et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Sui et al. undefined; Flannery et al. undefined; O'Brien et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Kominiarek et al. undefined;
18	Women living in low-income environments found additional barriers to manage their weigh, whereas	High confidence	Minor concerns regarding methodological limitations,	Garnweidner et al. undefined; Leiferman et al. undefined; O'Brien et al. undefined; Marquez et al. undefined; Kominiarek et al. undefined;

	women with higher educational level, higher socioeconomic status, multiparity and older age reported more facilitators.	No/Very minor concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	

GRADE-CERQual Assessment Worksheet

Evidence Profile

#	Summarized Review Finding	Methodological limitations	Coherence	Adequacy	Relevance	GRADE-CERQual assessment of confidence	References
1	Some women lacked knowledge regarding how to manage their weight during pregnancy. This lack of knowledge included lack of awareness of risks, and lack of awareness of recommendations regarding physical activity and dietary advice and the associated benefits.	**Minor concerns** Minor concerns regarding methodological limitations because poor reporting of reflexibity statement and ethical concerns.	**No/Very minor concerns** The data supporting this finding is quite descriptive, good fit between the finding and the data	**No/Very minor concerns** This finding is supported by a high number of studies with rich data	**No/Very minor concerns** A variety of high-income countries are represented in the studies contributing to this findings.	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Faucher & Mirabito undefined; Flannery et al. undefined; Garnweidner et al. undefined; Groth et al. undefined; Holton et al. undefined; Keely et al. undefined; Kominiarek et al. undefined; Lee et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
2	Understanding the value of food quality and benefits of physical activity during pregnancy, as well as being aware of the	**Minor concerns** Minor concerns regarding	**Minor concerns** Minor concerns regarding	**No/Very minor concerns**	**No/Very minor concerns**	**High confidence** Minor concerns regarding	Denison et al. undefined; Faucher & Mirabito undefined; Ferrari et al. undefined;

	recommendations, acted as facilitator for some women.	methodological limitations because poor reporting of reflexibity statement and ethical concerns.	coherence because some data shows that awareness is not always translated into behaviour change.	All studies except one contribute to this finding	All studies except one contribute to this finding	methodological limitations, Minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Flannery et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Groth et al. undefined; Keely et al. undefined; Kominiarek et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
3	Women engaged in a process of weighing up perceived benefits and potential risks to make choices about their weight management behaviours. Some women perceived the risks of overweight and obesity as exaggerated and felt that unhealthy choices could be compensated.	**Minor concerns** Minor concerns regarding methodological limitations because poor description of recruitment strategy overall and poor description of	**Minor concerns** Minor concerns regarding coherence because this finding is highly interpretative but there is enough data to support it.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** Wide range of contexts represented, direct relevance	**Moderate confidence** Minor concerns regarding methodological limitations, Minor concerns regarding coherence, No/Very minor concerns	Denison et al. undefined; Faucher & Mirabito undefined; Flannery et al. undefined; Flannery et al. undefined; Groth et al. undefined; Holton et al. undefined; Keely et al. undefined; Kominiarek et al. undefined;

		reflexivity statements and ethical issues.				regarding adequacy, and No/Very minor concerns regarding relevance	Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
4	Some women expressed concerns of harming their babies when doing exercise, especially in the cases of women with fertility issues or history of pregnancy loss. These fears led women to believe that the potential risks were stronger than the benefits of physical activity, so they decided to avoid it.	**Minor concerns** Minor concerns regarding methodological limitations because poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	**No/Very minor concerns** No or very minor concerns. No data contradicting the finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**Minor concerns** Minor concerns regarding relevance because 6 out of the 9 studies are specific to USA context.	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and Minor concerns regarding relevance	Denison et al. undefined; Flannery et al. undefined; Groth et al. undefined; Kominiarek et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
5	Some women expressed feelings of shame and guilt due to their weight. These negative feelings had a pejorative effect over their body-image and self-steem which prevented them from wanting to	**Minor concerns** Minor concerns regarding methodological	**Minor concerns** Minor concerns regarding coherence	**No/Very minor concerns** The number of studies and	**No/Very minor concerns** Wide range of contexts	**High confidence** Minor concerns regarding methodological	Denison et al. undefined; Faucher & Mirabito undefined; Flannery et al. undefined;

	engage in physical activities. Additionally, such feelings also acted as a barrier for communication and information transmission between women and healthcare professionals as weight management was perceived as a sensitive topic.	limitations because because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	because this finding is highly interpretative. Although there is enough data to support the finding, slightly different interpretations might be possible.	richness of data is enough to support finding	represented, direct relevance to review question	limitations, Minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Flannery et al. undefined; Garnweidner et al. undefined; Holton et al. undefined; Keely et al. undefined; Kominiarek et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Sui et al. undefined;
6	Women's pre-pregnancy habits played an important role in their weight management behaviours during pregnancy. Women who lacked an exercise routine or had poor dietary habits encountered additional challenges, whereas women who had healthy pre-established habits found it easier to maintain them.	**Minor concerns** Minor concerns regarding methodological limitations because because of poor description of recruitment strategy overall and poor description of reflexivity	**No/Very minor concerns** No or very minor concerns. No data contradicting the finding.	**Minor concerns** Minor concerns regarding adequacy because only 8 of 17 studies are contributing to this finding, however data is rich enough	**No/Very minor concerns** Wide range of contexts represented, direct relevance	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns	Flannery et al. undefined; Groth et al. undefined; Keely et al. undefined; Kominiarek et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Reyes et al. undefined;

		statements and ethical issues.				regarding relevance	
7	Lack of motivation to healthy eating or physical activity were reported as common barriers which could be overcome with the influence of external motivations.	**Moderate concerns** Moderate concerns regarding methodological limitations because of poor description of reflexivity statements and ethical issues	**Minor concerns** Minor concerns regarding coherence because no data contradicting the finding.	**Moderate concerns** Moderate concerns regarding adequacy because of the small number of studies supporting this finding	**No/Very minor concerns** Wide range of contexts and sociodemographic range represented, direct relevance	**Moderate confidence** Moderate concerns regarding methodological limitations, Minor concerns regarding coherence, Moderate concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Groth et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; Sui et al. undefined;
8	Pregnancy symtpoms like tiredness, reduced mobility, or nausea can act as barriers to physical activity and interfere in women's eating behaviours because of potential aversion to foods, cravings, or levels of appetite.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment	**No/Very minor concerns** No data contradicting the finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding	Denison et al. undefined; Faucher & Mirabito undefined; Ferrari et al. undefined; Flannery et al. undefined; Flannery et al. undefined; Groth et al. undefined;

		strategy overall and poor description of reflexivity statements and ethical issues.				coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Holton et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
9	For some women, being pregnant acted as a motivation to make changes in their habits towards healthier diet and activity behaviours. These women reported that felt a sense of responsibility towards the wellbeing of their babies, they wanted to be healthy for their children and act as role model for their own health behaviours.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Faucher & Mirabito undefined; Ferrari et al. undefined; Flannery et al. undefined; Keely et al. undefined; Leiferman et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
10	Having the support of the social context to engage in weight managements behaviours	**Minor concerns**	**No/Very minor concerns**	**No/Very minor concerns**	**No/Very minor concerns**	**High confidence**	Denison et al. undefined;

	acted as facilitators, especially when women had positive role models throughout all their lifes.	Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	No data contradicting the finding. Good fit between data and finding.	The number of studies and richness of data is enough to support finding	Wide range of contexts represented, direct relevance to research question	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Flannery et al. undefined; Flannery et al. undefined; Groth et al. undefined; Lee et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Sui et al. undefined;
11	The lack of support from family and friends to engage in weight management behaviours acted as a barrier for many women. In some occasions, these women lacked positive role models and were encouraged to rest.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and	Denison et al. undefined; Flannery et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Kominiarek et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; Padmanabhan et al. undefined; Sui et al. undefined;

	statements and ethical issues.				No/Very minor concerns regarding relevance	
12 Having the support of the social context to engage in weight managements behaviours acted as facilitators, especially when women had positive role models throughout all their lifes.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Flannery et al. undefined; Flannery et al. undefined; Groth et al. undefined; Lee et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Sui et al. undefined;
13 Some women expressed feeling judged and stigmatised for being overweight and pregnant. These negative feelings contribute to the perception that weight management is a sensitive topic to discuss, preventing information exchange and advice provision with HCP.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**Minor concerns** Minor concerns regarding adequacy because of the limited number of studies	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**Moderate confidence** Minor concerns regarding methodological limitations, No/Very minor	Denison et al. undefined; Faucher & Mirabito undefined; Holton et al. undefined; Keely et al. undefined; O'Brien et al. undefined;

	recruitment strategy overall and poor description of reflexivity statements and ethical issues.		supporting this finding		concerns regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Padmanabhan et al. undefined; Sui et al. undefined;
14 Some women encountered insensitive or judgemental HCP during their antenatal care that made them feel embarrassed and stigmatised for being overweight. These led some women to adopt avoidant attitudes, where they avoided to speak about weight management, or defensive attitudes, where they received advice regarding weight management as a challenge to their individuality.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	**Minor concerns** Minor concerns regarding coherence because this finding is highly interpretative. Although there is enough data to support the finding, slightly different interpretations might be possible.	**Moderate concerns** Moderate concerns regarding adequacy because very limited number of studies supporting this finding	**Moderate concerns** Moderate concerns regarding relevance because context limited to UK and Ireland	**Low confidence** Minor concerns regarding methodological limitations, Minor concerns regarding coherence, Moderate concerns regarding adequacy, and Moderate concerns regarding relevance	Faucher & Mirabito undefined; Flannery et al. undefined; Keely et al. undefined;
15 Women were not satisfied the level of education received regarding weight management during their antenatal care.	**Minor concerns**	**No/Very minor concerns**	**No/Very minor concerns**	**No/Very minor concerns**	**High confidence**	Denison et al. undefined; Ferrari et al. undefined;

	Some women felt overwhelmed by the amount of information and sources available to them, especially regarding dietary behaviours, which led them to find contradictive information in some cases. Information was too hesitant or conservative, and lacked specific advice.	Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	No data contradicting the finding. Good fit between data and finding.	The number of studies and richness of data is enough to support finding	Wide range of contexts represented, direct relevance to research question	Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Flannery et al. undefined; Garnweidner et al. undefined; Holton et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Padmanabhan et al. undefined; Sui et al. undefined;
16	Women who received information and advice regarding physical activity and diet during pregnancy were more able to engage in weight management behaviours. These women valued their HCP opinions and engaged actively in information seeking behaviours.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity	**Moderate concerns** Moderate concerns regarding coherence because of limited data contradicting trustworthiness of healthcare professionals	**Minor concerns** Minor concerns regarding adequacy because of limited number of studies supporting this finding.	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**Moderate confidence** Minor concerns regarding methodological limitations, Moderate concerns regarding coherence, Minor concerns regarding adequacy, and	Ferrari et al. undefined; Flannery et al. undefined; Garnweidner et al. undefined; Holton et al. undefined; Lee et al. undefined; Leiferman et al. undefined;

	statements and ethical issues.				No/Very minor concerns regarding relevance	
17 Environmental factors such as accessibility to healthy shops or sports facilities and affordability of fast food, and lack of time or other commitments also had a negative influence on women's physical activity and dietary habits.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of recruitment strategy overall and poor description of reflexivity statements and ethical issues.	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**No/Very minor concerns** The number of studies and richness of data is enough to support finding	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns regarding coherence, No/Very minor concerns regarding adequacy, and No/Very minor concerns regarding relevance	Denison et al. undefined; Faucher & Mirabito undefined; Flannery et al. undefined; Groth et al. undefined; Kominiarek et al. undefined; Lee et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined; Padmanabhan et al. undefined; Reyes et al. undefined; Sui et al. undefined;
18 Women living in low-income environments found additional barriers to manage their weigh, whereas women with higher educational level, higher socioeconomic status, multiparity and older age reported more facilitators.	**Minor concerns** Minor concerns regarding methodological limitations because of poor description of	**No/Very minor concerns** No data contradicting the finding. Good fit between data and finding.	**Minor concerns** Minor concerns regarding adequacy because of the limited number of studies	**No/Very minor concerns** Wide range of contexts represented, direct relevance to research question	**High confidence** Minor concerns regarding methodological limitations, No/Very minor concerns	Garnweidner et al. undefined; Kominiarek et al. undefined; Leiferman et al. undefined; Marquez et al. undefined; O'Brien et al. undefined;

	recruitment strategy overall and poor description of reflexivity statements and ethical issues.	supporting the finding	regarding coherence, Minor concerns regarding adequacy, and No/Very minor concerns regarding relevance

Supplementary file 6.4 – Quality appraisals

Reference	Clear aims	Qualitative methodology appropiate	Research design appropiate to meet aims	Recruitment strategy appropiate	Data collected in a way that addressed the research issue	Relationship between researcher and participants considered	Ethical issues	Data analysis rigorous	clear statement of findings	How valuable is the research	Total Score
Denison et al.	2	2	2	2	2	0	1	2	2	2	17
Ferrari et al.	2	2	1	2	2	0	1	2	2	2	16
Faucher et al.	2	1	2	1	2	1	1	1	2	2	15
Flannery et al.	2	2	2	1	2	2	2	2	2	2	19
Flannery et al.	2	2	1	2	2	0	1	2	2	2	16
Garnweidner et al.	2	2	2	1	2	0	1	2	2	2	16
Groth et al.	2	2	2	1	2	0	1	1	2	2	15
Holton et al.	2	2	2	1	2	0	0	2	2	2	15
Keely et al.	2	2	2	2	1	1	1	1	2	1	15
Kominiarek et al.	2	2	2	1	2	1	1	2	2	2	17
Lee et al.	2	2	2	1	1	0	1	2	2	2	15
Leiferman et al.	2	2	2	1	1	0	1	2	2	2	15
Marquez et al.	2	2	2	1	2	0	1	2	2	1	15
O'Brien et al.	2	2	1	2	2	1	1	1	2	2	16
Padmanabhan et al.	2	2	2	1	2	0	1	2	2	2	16
Reyes et al.	2	2	2	2	2	0	1	2	2	1	16
Sui et al.	2	2	2	2	2	0	1	1	2	2	16

Supplementary file 7.1 - Standards for reporting qualitative research: a synthesis of recommendations

Topic	Item	Page
Title		
S1 Title	Concise description of the nature and topic of the study Identifying	Page 1. Study title "Exploring first time mothers' experiences and knowledge about behavioural risk factors for stillbirth"
S2 Abstract	Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	Page 1.
Introduction		
S3 Problem formulation	Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	Introduction Paragraph 1: Description of problem/phenomenon. Introduction Paragraph 2 and 3: Review of previous literature. Introduction Paragraph 4: problem statement and study aims.
S4 Purpose or research question	Purpose of the study and specific objectives or questions	Introduction Paragraph 4: study aim. *The main objective of this study was to explore women's experiences of modifiable factors during pregnancy and knowledge and beliefs regarding behavioural risk factors related to stillbirth. Additionally, we aimed to examine women's experiences, if any, of being informed about such risk factors during their antenatal care.*

Methods		
S5 Qualitative approach and research paradigm	Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, onstructivist/interpretivist) is also recommended; rationale	Methods section, 2.1 Design.
S6 Researcher characteristics and reflexivity	Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	Methods section,2.7 Reflexivity statement.
S7 Context	Setting/site and salient contextual factors; rationale	Methods section. 2.3 Setting and Sample.
S8 Sampling strategy	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale	Methods section. 2.2 Recruitment and 2.4 Data collection.
S9 Ethical issues pertaining to human	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other	Methods, section 2.6. Ethical considerations

	confidentiality and data security issues	
S10 Data collection methods	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale	Methods. Section 2.4 Data collection.
S11 Data collection instruments and technologies	Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	Methods. Section 2.4 Data collection and supplementary file 2.
S12 Units of study	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	Results. Section 3.1 Sample Characteristics and Table 1.
S14 Data analysis	Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rational	Methods. Section 2.5 Data analysis

S15 Techniques to enhance trustworthiness	Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale	Methods. Section 2.5 Data analysis. "One researcher (TES) read and re-read all of the interview transcripts. Deductive open coding was then initiated which facilitated the identification of units of meanings that related to the research aims. Subsequently, those codes were categorised and grouped into themes, and re-labelled where appropriate. A record of the evolvement of the themes and the category names was always kept. Further analysis allowed the researchers (TES, KMS) to group the different categories into themes, by refining its meaning to portray the story the data tells. A second author (KMS) reviewed and followed the coding process at all stages, and discussions were held as necessary."
Results/findings		
S16 Synthesis and interpretation	Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	Results. 3.2 Findings section.
S17 Links to empirical data	Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	Results. 3.2 Findings section (quotes)).

Discussion		
S18 Integration with prior work, implications, transferability, and contribution(s) to the field	Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	Discussion. Paragraph one includes a short summary of findings. The rest of the conclusion addresses all the rest of items.
S19 Limitations	Trustworthiness and limitations of findings	Discussion. Paragraph 8.
Others		
S20 Conflicts of interest	Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	This information can be found at the bottom of the paper.
S21 Funding	Sources of funding and other support; role of funders in data collection, interpretation, and reporting	This information can be found at the bottom of the paper.

Supplementary file 7.2 – Topic guide

Introduction	
History and health habits	Did you do anything to prepare to get pregnant? What were your main worries related to health? Why? What habits did you have to change when you became pregnant? How difficult/easy did you find those changes? Why did you make those decisions? If a friend were to get pregnant, what would you recommend so she can make sure she has a healthy pregnancy?
Risk factors for stillbirth	Are you aware of any maternal behaviours that might be bad for the baby? And do you know any pregnancy issues that might be associated with those behaviours? What do you know about smoking/alcohol/drugs during pregnancy? What do you know about diet or physical activity during pregnancy? Did you learn how to manage your weight gain safely during pregnancy? What do you know about sleep position? Where did you get the information? Who approached you with this information? Have you heard of stillbirth? Can you explain it to me? When do you think it can happen?

	Have you heard of any behaviours that might increase the risk of stillbirth? Did you discuss any particular behaviours within your antenatal care? (smoking/alcohol/drugs/attendance at antenatal care/sleep position/weight management, diet exercise).
Feelings and opinion	Were you approached by your doctor/midwife to talk about stillbirth? or risk factors for stillbirth? How did you feel (how would have made you feel) when your doctor/midwife talked to you about stillbirth? Do you think it would have been useful for your doctor/midwife to talk to you about stillbirth? or the risk factors for it? What benefits could pregnant women (did you) get from discussing these risks with their healthcare providers? How or when would you have liked the doctor/nurse to approach the subject? (prompts: What gestational age? What opportunity? e.g: routine clinic, in Emergency Department for a problem, etc.?) Do you think it is important to speak about the risk of stillbirth to a healthy low risk pregnant woman?
Information sources	Among all of the sources available, which one do you trust the most? How did you use that source of information? What did you like about that source of information? Did you receive or find any conflicting information? (prompts: internet, GP, midwife, obs., family, fiends) About what?
Intervention	Have you found anything that helped you promote or support your health habits while you were pregnant? For example, text messages with health related information; educational groups; one on one sessions with healthcare professionals, etc.?

	Can you describe it to me? What elements of it did you find most helpful? Do you think you had anyone in your social environment that influenced your health during your pregnancy? How? In your opinion, what kind of support or information service would help pregnant women to improve their health habits? What are the best ways to provide this information or who should be providing it? (Prompts: information sessions, leaflets, websites, one to one sessions, suggestions?) When is the best time to deliver this intervention to women? When would you have liked to get the information?

Supplementary file 8.1 – Published protocol

HRB Open Research

HRB Open Research 2022, 4:92 Last updated: 24 JAN 2023

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STUDY PROTOCOL

REVISED A protocol for a systematic review of behaviour change techniques used in the context of stillbirth prevention [version 2; peer review: 2 approved, 1 approved with reservations]

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Abstract

Background: Stillbirth is a devastating pregnancy outcome that affects approximately 3.5 per 1000 births in high-income countries. Previous research has highlighted the importance of focusing prevention efforts on targeting risk factors and vulnerable groups. A wide range of risk factors has been associated with stillbirth before, including maternal behaviours such as back sleep position, smoking, alcohol intake, illicit drug use, and inadequate attendance at antenatal care. Given the modifiable nature of these risk factors, there has been an increase in the design of behaviour change interventions targeting such behaviours to reduce the risk of stillbirth.
Objectives: The aim of this study is to identify all behavioural interventions with a behavioural component designed and trialled for the prevention of stillbirth in high-income countries, and to identify the behaviour change techniques (BCTs) used in such interventions using the Behaviour Change Techniques Taxonomy V1 (BCTTv1).
Inclusion criteria: Interventions will be included in this review if they (1) have the objective of reducing stillbirth rates with a focus on behavioural risk factors; (2) are implemented in high-income countries; (3) target pregnant women or women of childbearing age; and (4) are published in research articles.
Methods: A systematic search of the literature will be conducted. The results of the search will be screened against our inclusion criteria by two authors. The following data items will be extracted from the selected papers: general information, study characteristics, participant

Open Peer Review

Approval Status ? ✓ ✓

	1	2	3
version 2 (revision) 11 Mar 2022		✓ view	✓ view
version 1 19 Aug 2021	? view	? view	

1. **Samah Alageel**, King Saud University, Riyadh, Saudi Arabia
2. **Rae Thomas**, Bond University, Gold Coast, Australia
3. **Andrea M. Patey**, Ottawa Hospital Research Institute, Ottawa, Canada
University of Ottawa, Ottawa, Canada

Any reports and responses or comments on the article can be found at the end of the article.

and intervention/approach details. The Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria will be used to assess the methodological quality of included studies. Intervention content will be coded for BCTs as present (+) or absent (-) by two authors using the BCTTv1, discrepancies will be discussed with a third author. A narrative synthesis approach will be used to present the results of this systematic review.

Keywords
stillbirth, intervention, behaviour change, risk factors



This article is included in the Maternal and Child Health collection.

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Author roles: Escañuela Sánchez T: Conceptualization, Methodology, Writing – Original Draft Preparation; **Byrne M**: Conceptualization, Methodology, Supervision, Writing – Review & Editing; **Meaney S**: Conceptualization, Methodology, Supervision, Writing – Review & Editing; **O'Donoghue K**: Conceptualization, Funding Acquisition, Methodology, Supervision, Writing – Review & Editing; **Matvienko-Sikar K**: Conceptualization, Investigation, Methodology, Supervision, Writing – Review & Editing

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REVISED **Amendments from Version 1**

This manuscript has been updated to reflect the reviewers' suggestions:

- Further detail has been added to the introduction section to strengthen the justification of why this review is needed and how is it different from the previous Cochrane review published by Ota et al.
- The PROSPERO ID has been added to the protocol section.
- A sentence has been added to the eligibility criteria section to strengthen the justification of focusing the review on high-income countries.
- Changes in rates of stillbirth has been added as an outcome in the PICO framework table. Studies that to not include stillbirth (or perinatal death) as one of their outcomes will not be included.
- A description of how the manual search will be conducted has been added to the selection process section.
- A second risk of bias tool has been proposed to make sure all types of studies are covered, as per one of our reviewers' suggestions.
- Further description has been added to the data synthesis section and BCT coding to explain how the data will be treated.

Any further responses from the reviewers can be found at the end of the article

Abbreviations

PRISMA: Preferred Reporting Items for Systematic Reviews and Meta-Analyses; BCT: Behaviour Change Technique.

Introduction

Stillbirth is one of the most devastating outcomes of pregnancy that expectant parents can face. In Ireland, a stillbirth is defined as an infant born weighing 500 grams or more or at a gestational age of at least 24 weeks who shows no signs of life[1]. However, this definition of stillbirth is not globally accepted, with different countries using different weight or gestational thresholds, ranging from 350 grams to 1000 grams and from 20 weeks to 28 weeks[2]. The worldwide estimates of stillbirth rates in 49 high-income countries at 28 weeks gestation was 3.5 per 1,000 total births in 2015, with country-specific rates varying from 1.3 in Iceland to 8.8 in Ukraine[3]. In Ireland, the latest data published by the National Perinatal Epidemiology Centre shows that the prevalence of stillbirth was 3.8 per 1,000 births in 2017[4].

The stillbirth series published in the Lancet in 2016 highlighted the importance of focusing future stillbirth prevention efforts in high-income countries on targeting specific causes, risk factors, and vulnerable groups[5]. In high-income countries, the majority of stillbirths occur prior to labour and are associated with placental pathology[6]. However, previous research has shown that stillbirth is associated with a wide range of risk factors, including maternal medical factors, for example hypertensive disorders or diabetes[7–9]; factors associated with the woman's obstetric history, for example having a history of previous pregnancy loss[10], primiparity[11] or multiple pregnancy[12]; pregnancy related complications such as placental insufficiency[13] or fetal growth restriction[14]; ethnic[15] and socioeconomic status[16]; maternal overweight and obesity[17–19]; and maternal age[20–22]. Maternal behavioural risk factors include factors such as sleep position[23], smoking[5,24,25], alcohol intake[26,27], illicit drug use[28–31] and inadequate antenatal care[32,33]. Such behavioural risk factors are modifiable and so provide useful targets for stillbirth prevention interventions.

Considering that some of the mentioned maternal behavioural risk factors have the potential to be modified, there has been an increase in development and implementation of antenatal behavioural interventions targeting those behaviours to reduce the risk of stillbirth. Behavioural interventions are defined here as those targeting behaviours such as smoking cessation interventions, midwifery led care, birth attendant training, alternative packages of antenatal care and diet and exercise interventions[34]. However, in an overview of 43 Cochrane reviews assessing 61 different stillbirth prevention approaches across the globe, few reviews were found to produce clear evidence as the effectiveness of interventions differed across settings, highlighting the importance to understand the intervention's context[34]. Further, this Cochrane review of reviews had a focus on a broad range of interventions (i.e., nutritional interventions, prevention and management of infection, prevention and management of morbidities, screening and management of fetal growth), not only those with a behaviour change component. To date, there has not been any systematic review of behaviour change interventions conducted in the context of stillbirth prevention. Hence, this review is important to understand what techniques have been used previous to inform the development of theoretically robust evidence based interventions.

Previous studies have been conducted to explore facilitators and barriers influencing health behaviours during pregnancy[35–37]. Identifying and synthesising all existing behaviour change interventions designed in the context of stillbirth prevention, combined with the information obtained from the literature exploring factors influencing behavioural risk factors[35–37], will inform the development of new strategies applicable in high-income countries. Given the variability across definitions, outcomes and measures of stillbirth, and the potential for differential intervention approaches to be used, there is a need to identify and synthesise the different components, or behaviour change techniques (BCTs), used in such interventions. A BCT is an observable, replicable and irreducible component of an intervention, an "active ingredient", designed to alter or redirect casual processes that regulate behaviour[38]. Identification of BCTs in interventions allows for accurate replication of interventions and faithful implementation of interventions demonstrating effectiveness[38]. Hence, knowing which specific BCTs have been used within interventions is important to build cumulative evidence towards delivering effective and replicable interventions in this context. Additionally, reviewing and identifying BCTs in stillbirth prevention interventions will also enable future investigations of links between BCTs and

mechanisms of action that will facilitate the optimisation of intervention effectiveness[39].

To our knowledge, there has been no systematic examination of BCTs used in behaviour change interventions designed with the objective to reduce stillbirth risks. Such a review is essential to informing and improving the development of future stillbirth prevention interventions. This research will build on previous reviews of general stillbirth prevention interventions (including infection management, pharmacotherapy for prevention of pre-eclampsia, screening for diabetes, ultrasound in early and late pregnancy, antenatal cardiotocography for fetal assessment), such as the one conducted by Ota and colleagues[34], by focusing on those interventions with a behavioural component and by aiming to identify the BCTs used within those interventions.

The aim of this systematic review is to identify all research studies examining behaviour change interventions used in the context of stillbirth prevention within high-income countries, and to synthesise the BCTs used in these interventions.

Objectives

- To identify behavioural interventions used for the prevention of stillbirth in high-income countries.
- To identify and code the BCTs used in such interventions using the Behaviour Change Techniques Taxonomy V1 (BCTTv1)[38].

Protocol

Details of this review have been submitted for registration to the PROSPERO database (ID CRD42021264914). Amendments made to the protocol will be acknowledged on PROSPERO and in any publications following this study. This protocol has been informed by the Preferred Reporting Items for Systematic Review and Meta-Analysis Protocols (PRISMA-P) guidelines for the reporting of systematic reviews[40].

Eligibility criteria

The "PICO" framework was used to select inclusion and exclusion criteria for this review (Table 1). For the purpose of this review, we did not select any specific definition for stillbirth because, as noted, definitions differ internationally[2].

The following study types are eligible for inclusion: case-control studies, randomised control trials, cross-sectional studies, and quasi experimental studies. Studies will be eligible for inclusion when published in high-income countries given the differences between care systems and the challenges associate with improving pregnancy outcomes amongst low-, middle- and high-income countries. Additionally, focusing this study in high-income countries only will facilitate the use of its findings to develop behaviour change strategies applicable to such contexts. High-income countries will be defined based on the World Bank Country Classification (Gross National Income per capita of $12,696 or more in 2020)[41]. The findings of this systematic review will help inform the development of an intervention to be used in a high-income country setting.

Table 1. PICO framework.

PICO framework	Eligibility criteria
Population or problem	Population: - Pregnant women - Women of reproductive age Setting: - High-income countries
Intervention or exposure	Any intervention published in research articles designed to prevent stillbirth that includes a focus on behavioural risk factors.
Comparison	Participants who were not exposed to an intervention or who receive 'standard care'
Outcome	Changes in rates of stillbirth after intervention, no specific definition for stillbirth will be used as they differ internationally. Behaviour change techniques used in the interventions, classified using the Behaviour Change Technique Taxonomy V1[38]

Information sources

A systematic literature search covering all interventions in the context of stillbirth prevention from inception to present will be performed in the following databases: CINAHL Complete, SocIndex, Web of Science, PubMed, PsycINFO and Open Grey. Searches in Open Grey and contact with authors will facilitate access to non-published articles. Additionally, we will also conduct a manual reference list searching of the identified articles.

Search strategy

Keyword searches will be used across four different concepts (1) Stillbirth, (2) Intervention, (3) Pregnancy and (4) Study design. The search strategy was developed for PubMed (see Table 2) but it will be adapted depending on the database in combination with database-specific filters.

Study records

Data management

Studies will be imported into Endnote, duplicates will be identified by using the automatic "Check for Duplicates" tool, as well as manually screening the results. The screening of titles will be conducted using Rayyan, which is a web platform for systematic reviews[42].

Selection process

Two independent reviewers (TES, KMS) will individually screen titles, abstracts, and full articles to identify eligible studies, using our inclusion and exclusion criteria. In cases of uncertainty in title and abstract screening, studies will be included in the full text review stage. When identifying systematic reviews, a manual search of their included interventions will be conducted to ensure that all relevant interventions are included

Table 2. Example of search.

Database: Pubmed Date:		
	Search Terms	**Search Results**
Concept 1 Stillbirth		
1.	TI/AB Stillbirth	
2.	TI/AB Fetal death	
3.	TI/AB Intrauterine death	
4.	TI/AB Perinatal death	
5.	**S1 OR S2 OR S3 OR S4**	
Concept 2 Intervention		
6.	TI/AB Health promotion	
7.	TI/AB Education	
8.	TI/AB Awareness	
9.	TI/AB Information	
10.	TI/AB Behaviour	
11.	TI/AB Behavior	
12.	TI/AB Behaviour Change	
13.	TI/AB Behavior change	
14.	TI/AB Prevent*	
15.	TI/AB Intervention	
16.	TI/AB Strategy	
17.	TI/AB Guidance	
18.	**S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17**	
Concept 3 Pregnancy		
19.	TI/AB Pregnant women	
20.	TI/AB Pregnancy	
21.	TI/AB Childbearing age	
22.	TI/AB Prenatal	
23.	TI/AB antenatal	
24.	**S19 OR S20 OR S21 OR S22 OR S23**	
Concept 4 Type of report		
25.	TI/AB "Randomised controlled trial"	
26.	TI/AB "Randomized controlled trial"	
27.	TI/AB RCT	
28.	TI/AB "control group"	
29.	TI/AB "controlled trial"	
30.	TI/AB "case-control study"	
31.	TI/AB "quasi-experimental"	
32.	TI/AB Initiative	
33.	TI/AB Guideline	
34.	**S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31 OR S32 OR S33**	
35.	**S5 AND S18 AND S24 AND S34**	
Limiters: - Humans		

in this review. We will also perform a further manual citation search of included studies. Discrepancies will be resolved through consensus discussion and/or recourse to a third reviewer.

A PRISMA flow diagram[43] will be used to show process of study screening and summarise the inclusion and exclusion criteria at each stage of the review.

Data collection process

Once the final list of studies to include in the review has been determined, all supplementary materials or additional studies related to the same intervention will be retrieved by TES before data extraction or quality assessments. When direct access is not provided to these supplementary materials online, the original study authors will be contacted to request these materials. Published manuscripts of trial outcomes and intervention protocols will be examined for intervention details.

Data items

A structured data extraction sheet using Microsoft Word will be used to extract the study characteristics (see *Extended data*[44]), which will be piloted in advanced. Data will be extracted by one reviewer (TES) and verified for accuracy by another reviewer (KMS).

Study characteristics to be extracted include:

(1) General information: Authors, title, name of intervention/ approach (if applicable), year of publication, country of origin.

(2) Study characteristics: Study aims/objectives, type of study/report, stillbirth definition used, recruitment strategy.

(3) Participants: cohort size, number of people in control arm (if applicable), number of people in intervention arm (if applicable), mean age, age range, socio-economic status, education level, ethnicity, pregnancy status, relationship status.

(4) Intervention/approach details: setting, investigated topic/target behaviour, description of intervention/approach and control treatments, delivery mode, intervention duration, current stage of implementation, outcomes measured in intervention (e.g. reduction stillbirth rates, increased awareness, behaviour change), effectiveness of intervention.

Outcomes and prioritization
Primary outcome:

- BCTs used (as described in the BCTTv1[38])

BCT coding
The coding of the BCTs included in the identified interventions/approached will be conducted following the BCTTv1 approach[38]. Intervention content will be coded for presence (+) or absence (-) of the BCTs. This process will be done by two authors independently (TES, KMS) and discussion and comparison will be used to address any discrepancies. A third member of the research team (MB) will be consulted in case of disagreement. The findings of the BCT coding will be presented in tabular format. To examine the use of BCTs in the interventions, the percentage of individual BCTs across studies and the total number of BCTs per intervention will be calculated. Additionally, we will tabulate the identified BCTs in order to identify patterns across the different studies.

Risk of bias in individual studies/quality assessments
The Cochrane Effective Practice and Organization of Care (EPOC) risk of bias criteria[45] will be used to assess the methodological quality of the included studies that will subsequently feed into the GRADE process[46].

The Joanna Briggs Institute (JBI) critical appraisal checklist will be used for both descriptive and analytical cross-sectional studies.

Data synthesis
We anticipate that a meta-analytic approach would be inappropriate for comparison of studies due to the potential high heterogeneity of the interventions and potential BCTs. Hence, we will use a narrative synthesis approach as recommended by Cochrane when a meta-analytical approach is not possible, to summarise the findings of BCTs. Studies will be grouped based on the behaviour they focus on. The results of the studies will be systematically examined by summarising the same features of each study using pre-designed data extraction sheets. The findings will also be presented in tabular form. This approach will facilitate the description of the interventions and BCTs used, taking into account quality appraisals.

Analysis of subgroups or subsets
While subgroup analyses may be undertaken, it is not possible to specify the groups in advance.

Dissemination
The PRISMA checklist[43] will be used to report findings of the review. We will communicate the findings by publication in a peer-reviewed journal, and by participation in scientific meetings and national and international conferences.

Study status
Study protocol has been completed.

Database search completed.

Title screening completed.

Conducting abstract screening.

Data availability
Underlying data
No underlying data are associated with this article.

Extended data
Harvard Dataverse: Study Characteristics extraction table for a protocol for a systematic review of behaviour change techniques used in the context of stillbirth prevention. https://doi.org/10.7910/DVN/0GCWXX[44].

Reporting guidelines
Harvard Dataverse: PRISMA-P Checklist for "A protocol for a systematic review of behaviour change techniques used in the context of stillbirth prevention". https://doi.org/10.7910/DVN/6WNOFO[47].

Data are available under the terms of the Creative Commons Zero "No rights reserved" data waiver (CC0 1.0 Public domain dedication).

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Supplementary file 8.2 – Example of search strategy

Database: Pubmed **Date:**		
	Search Terms	**Search Results**
Concept 1 Stillbirth		
72.	TI/AB Stillbirth	
73.	TI/AB Fetal death	
74.	TI/AB Intrauterine death	
75.	TI/AB Perinatal death	
76.	**S1 OR S2 OR S3 OR S4**	
Concept 2 Intervention		
77.	TI/AB Health promotion	
78.	TI/AB Education	
79.	TI/AB Awareness	
80.	TI/AB Information	
81.	TI/AB Behaviour	
82.	TI/AB Behavior	
83.	TI/AB Behaviour Change	
84.	TI/AB Behavior change	
85.	TI/AB Prevent*	
86.	TI/AB Intervention	
87.	TI/AB Strategy	
88.	TI/AB Guidance	
89.	**S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17**	
Concept 3 Pregnancy		
90.	TI/AB Pregnant women	
91.	TI/AB Pregnancy	
92.	TI/AB Childbearing age	
93.	TI/AB Prenatal	
94.	TI/AB antenatal	
95.	**S19 OR S20 OR S21 OR S22 OR S23**	
Concept 4 Type of report		
96.	TI/AB “Randomised controlled trial”	
97.	TI/AB “Randomized controlled trial”	
98.	TI/AB RCT	
99.	TI/AB “control group”	
100	TI/AB “controlled trial”	
101	TI/AB “case-control study”	
102	TI/AB “quasi-experimental”	

103	TI/AB Initiative	
104	TI/AB Guideline	
105	**S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31 OR S32 OR S33**	
106	**S5 AND S18 AND S24 AND S34**	
Limiters: - Humans		

Supplementary table 8.3 – Included interventions and materials

Author	Article title	Additional materials identified*
Intervention 1: The Safer Baby Bundle.		
Andrews et al.	Implementation and evaluation of a quality improvement initiative to reduce late gestation stillbirths in Australia: Safer Baby Bundle study protocol(Andrews, Ellwood, Middleton, et al., 2020) Stillbirth in Australia 2: Working together to reduce stillbirth in Australia: The Safer Baby Bundle initiative(Andrews, Ellwood, Gordon, et al., 2020)	Leaflets + website https://stillbirthcre.org.au/about-us/our-work/the-safer-baby-bundle/ Descriptive paper of development of intervention: Stillbirth in Australia 2: Working together to reduce stillbirth in Australia: The Safer Baby Bundle initiative(Andrews, Ellwood, Gordon, et al., 2020) Safer baby bundle. Handbook and resource guide. https://stillbirthcre.org.au/wp-content/uploads/2021/03/SBB-Handbook_Final-1.pdf Leaflet for women https://stillbirthcre.org.au/wp-content/uploads/2021/03/SB_POSTER_Mums-1.pdf Leaflets for HCP https://stillbirthcre.org.au/wp-content/uploads/2021/03/SB_Trifold-DL_HCP-2.pdf The safer baby bundle video https://vimeo.com/352404965

		Safer baby bundle masterclass https://stillbirthcre.org.au/wp-content/uploads/2021/04/SBB_Masterclass_Mainstream_V7_10052022.pdf Safer Baby Collaborative Summary (Victoria jurisdiction) https://www.safercare.vic.gov.au/improvement/projects/mbc/safer-baby
Intervention 2: An Integrated Randomized Intervention to Reduce Behavioural and Psychosocial Risks		
Subramanian et al.	An Integrated Randomized Intervention to Reduce Behavioural and Psychosocial Risks: Pregnancy and Neonatal Outcomes(Subramanian et al., 2012)	Additional paper describing the design, implementation, challenges faced and acceptability of the intervention: The design, implementation and acceptability of an integrated intervention to address multiple behavioural and psychosocial risk factors among pregnant African American women(Katz et al., 2008). Additional paper, RCT to assess behaviour change, no pregnancy outcomes reported: Reducing Psychosocial and Behavioural Pregnancy Risk Factors: Results of a Randomized Clinical Trial Among High-Risk Pregnant African American Women(Warland, 2013)

Intervention 3: Keeping baby SAFE		
Warland et al.	Keeping baby SAFE in pregnancy: Evaluating the brochure(Warland, 2013)	Brochure
Intervention 4: Saving Babies' Lives Care Bundle		
Widdows et al. / Tommy's	Stillbirth rates, service outcomes and costs of implementing NHS England's Saving Babies'Lives care bundle in maternity units in England: A cohort study(Kate Widdows et al., 2021) Evaluation of the implementation of the Saving Babies ' Lives Care Bundle in early adopter NHS Trusts in England(K. Widdows et al., 2018)	Saving Babies' Lives Version Two Implementation report(NHS England, 2019) Protocol: Saving babies' lives project impact and results evaluation (SPiRE): a mixed methodology study(Kate Widdows et al., 2018)
Intervention 5: Financial incentives for smoking cessation		
Tappin et al.	Financial incentives for smoking cessation in pregnancy: randomised controlled trial(Tappin et al., 2015)	Trial registration https://clinicaltrials.gov/ct2/show/NCT02606227
Intervention 6: My baby's movements		

Flenady et al.	My Baby's Movements: a stepped wedge cluster randomised controlled trial to raise maternal awareness of fetal movements during pregnancy study protocol(Flenady et al., 2019) My Baby's Movements: a stepped-wedge cluster-randomised controlled trial of afetal movement awareness intervention to reduce stillbirths[687] My Baby's Movements: An assessment of the effectiveness of the My Baby's Movements phone program in reducing late-gestation stillbirth rates[688]	Website https://stillbirthcre.org.au/about-us/our-work/my-babys-movement-trial/
Intervention 7: Count the kicks		
Saastad et al.	Fetal Movement Counting Improved Identification of Fetal Growth Restriction and Perinatal Outcomes - a Multi-Centre, Randomized, Controlled Trial(Saastad et al., 2011)	Information leaflet Trial registration: https://clinicaltrials.gov/ct2/show/NCT00513942 Website: https://countthekicks.org/
Saastad et al.	Implementation of uniform information on fetal movement in a Norwegian population reduced delayed reporting of decreased fetal movement and stillbirths in primiparous women - A clinical quality improvement(Saastad et al., 2010)	

Tveit et al.	Reduction of late stillbirth with the introduction of fetal movement information and guidelines – a clinical quality improvement(Tveit et al., 2009)	
Intervention 8: A controlled study of daily fetal movement counts in the prevention of stillbirths		
Lobb et al.	A controlled study of daily fetal movement counts in the prevention of stillbirth(Lobb et al., 1985)	NA
Intervention 9: Modifying maternal sleep position in late pregnancy through positional therapy		
Warland et al.	Modifying Maternal Sleep Position in Late Pregnancy Through Positional Therapy: A Feasibility Study(Warland et al., 2018)	Study protocol followed reported in Warland J, Dorrian J (2014) Accuracy of Self-Reported Sleep Position in Late Pregnancy. PLoS ONE 9(12): e115760. https://doi.org/10.1371/journal.pone.0115760
Kember at al.	Modifying maternal sleep position in the third trimester of pregnancy with positional herapy: a randomised pilot trial (Kember et al., 2018)	Research protocol obtained from supplementary materials. Trial registration: https://clinicaltrials.gov/ct2/show/NCT02377817

Andrews, C. J., Ellwood, D., Gordon, A., Middleton, P. F., Homer, C. S. E., Wallace, E. M., Nicholl, M. C., Marr, C., Sketcher-Baker, K., Weller, M., Seeho, S. K. M., & Flenady, V. J. (2020). Stillbirth in Australia 2: Working together to reduce stillbirth in Australia: The Safer Baby Bundle initiative. *Women and Birth*, *33*(6), 514–519. https://doi.org/10.1016/j.wombi.2020.09.006

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Supplementary file 8.4 - Quality Appraisals

The Cochrane Effective Practice and Organisation of Care (EPOC) (Part 1)								
Intervention n	Author	Title	Random sequence generation	Supportive evidence	Allocation concealment (selection bias)	Supportive evidence	Blinding of participants, personnel	Supportive evidence
2	Subramanian et al.	An Integrated Randomized Intervention to Reduce Behavioral and Psychosocial Risks: Pregnancy and Neonatal Outcomes / The design, implementation and acceptability of an integrated intervention to address multiple behavioral and psychosocial risk factors among pregnant African American women	High risk	Purposive sampling. Women are recruited based on specific criteria and availability. Quote from text: participants were recruited at six prenatal care clinics in Washington, DC from July 2001 to October 2003 and followed until July 2004. IRB approval was secured from participating institutions and informed consent obtained from participants. Pregnant AA women ages 18 years and above and less than 29 weeks gestation were asked to complete a brief computerized screening interview (Audio-Computer Assisted Self-Interview: A-CASI) to assess their eligibility and status with respect to the four psychosocial risk factors: active smoking, ETSE, depression, and IPV [...] Scores on the-CASI determined a woman's risk eligibility for the study and which risk components would be addressed should she be randomized to intervention	High risk	Authors used block randomization, this type of allocation process may be predictable, no information about blinding of the investigator or block size. Quote from text: Consenting participants who completed a baseline interview were randomized to the intervention (IG) or usual care (UCG) group using a site- and risk-specific block randomization design.	High-risk	No information about blinding presented in study
5	Tappin et al.	Financial incentives for smoking cessation in pregnancy: randomised controlled trial	Low risk	Quote from text: The stop smoking services aim to contact all smokers notified to them. Throughout the enrolment period, advisers from the services discussed the trial with referred women during their first contact. The contact details of clients who gave permission were securely passed to the trial team and helpline staff through the trial database. The helpline (a call	Low risk	Quote from text: The Glasgow Clinical Trials Unit embedded the randomisation in the trial database using randomised permuted blocks, with a block length of four, thus facilitating equal distribution of clients	High-risk	No information about blinding presented in study

				centre also commissioned to provide the NHS Smoking Helpline) contacted women, confirmed that all selection criteria had been met, enrolled participants using telephone consent, and conducted concealed random allocation		between the interventions. Allocation was concealed from staff and clients until after consent and recruitment. Helpline staff, who ascertained the primary outcome, self report of smoking status in late pregnancy, were blind to allocation status		
6		My Baby's Movements: a stepped wedge cluster randomised controlled trial to raise maternal awareness of fetal movements during pregnancy study protocol	High-risk	Non random, per cluster. It can be anticipated if a woman attending a specific hospital will be recruited or not. Quote from text: The trial will include 26 hospitals in ANZ with an aver-age of 3170 singleton births per year (range: 1400, 7000)giving 256,770 total births over 3 years.	Low risk	Quote from text: Clusters are assigned to the timing of the intervention(control and interventions periods) using a computer-generated random number table by the trial biostatistician(Michael Coory) who is not to be involved in the clinical aspects of the study.	High-risk	No information about blinding presented in study
	Flenady et al.*	My Baby's Movements: a stepped-wedge cluster-randomised controlled trial of a fetal movement awareness intervention to reduce stillbirths	High-risk	Non random, per cluster. It can be anticipated if a woman attending a specific hospital will be recruited or not. Quote from text: 27 maternity sites in ANZ were randomised in clusters.	Low risk	Quote from text: The MBM intervention was rolled out at randomly assigned time points, with sequential introduction into eight clusters of between three and five hospitals at 4-monthly intervals over a period of 3 years	High-risk	No information about blinding presented in study
	Skalecki et al.	My Baby's Movements: An assessment of the effectiveness of the My Baby's Movements phone program in reducing late-gestation stillbirth rates	High-risk	Non random, per cluster. It can be anticipated if a woman attending a specific hospital will be recruited or not. Quote from original study protocol: The trial will include 26 hospitals in ANZ with an aver-age of 3170 singleton births per year (range: 1400, 7000)giving 256,770 total births over 3 years.	Low risk	Quote from original study: The MBM intervention was rolled out at randomly assigned time points, with sequential introduction into eight clusters of between three and five hospitals at 4-monthly intervals over a period of 3 years	High-risk	No information about blinding presented in study
7	Saast et al.	Fetal Movement Counting Improved Identification of Fetal	Low risk	There is no enough information to determine if all women meeting their inclusion criteria were recruited for the study. Quote from text: Participants were	Low risk	Quote from text: Simple randomization was determined according to a compute-er-	Low risk	This type of intervention can not be blinded.

		Growth Restriction and Perinatal Outcomes - a Multi-Centre, Randomized, Controlled Trial		approached during their regular ultrasound screening in pregnancy weeks 17–19(flow-chart in figure 1). [...] Women were recruited from September 2007 through November 2009 at nine Norwegian hospitals from both urban and rural populations,		generated random allocation list with an equal probability offending up in each of the groups. The allocation sequence was concealed until participants were assigned to trial groups.		Quote from text: blinding for group assignment was not desirable neither for the participants nor their care providers, as use of a feta movement counting chart was intended to be an active tool for interaction between the woman and her midwife or physician.
8	Lobb et al.	A controlled study of daily fetal movement counts in the prevention of stillbirths	High risk	Although all women booked in participating centres were invited to intervention. the control group was formed by women who were specifically selected because of their high-risk status. Quote from text: The Cardiff 'Count to Ten Kick Chart' was introduced by two consultant obstetricians (Unit A) at the Liverpool Maternity Hospital during 1977, so that by late 1977 all women booked for confinement with Unit A were given a kick chart at about 28 weeks pregnant. [...] Four other consultant obstetricians (Unit B) at the hospital introduced kick charts for a few. selected, high-risk patients	High risk	Although all women were recruited from one clinic for the intervention arm, the control group was formed by a few, selected, high-risk patients from a second clinic. It could then be predicted to which group a recruited person would be allocated.	High-risk	No information about blinding presented in study
9	Warland et al.	Modifying Maternal Sleep Position in Late Pregnancy Through Positional Therapy: A Feasibility Study	High risk	Quote from text: Healthy pregnant women were recruited to the study via convenience and snowball sampling	Low risk	Quote from text: The intervention night was randomly al-located and the study nights were consecutive.	High risk	No information about blinding presented in study

	Kember et al.	Modifying maternal sleep position in the third trimester of pregnancy with positional therapy: a randomised pilot trial	High risk	Quote from text: Participants were recruited by a trained nurse at the IWK Health Centre	Low risk	Quote from text: Participants were allocated by computer-generated (R), one-to-one, simple randomisation into one of two possible cross-over schemes: PrenaBelt on the first PSG night followed by sham on the second PSG night and vice versa. Allocation sequence was concealed via unmarked, security-tinted, sealed envelopes by an independent statistician	Low risk	Quote from text: Participants, the recruiter and personnel involved in setting up, conducting, scoring and interpreting the PSG were blinded to allocation.

The Cochrane Effective Practice and Organisation of Care (EPOC) (Part 2)										
Intervention n	Author	Title	Blinding of outcome assessment (detection bias)	Supportive evidence	Incomplete outcome data (attrition bias)	Supportive evidence	Selective outcome reporting (reporting bias)	Supportive evidence	Other bias	Supportive evidence
2	Subramanian et al.	An Integrated Randomized Intervention to Reduce Behavioral and Psychosocial Risks: Pregnancy and Neonatal Outcomes / The design, implementation and acceptability of an integrated intervention to address multiple behavioral and psychosocial risk factors among pregnant African American women	Unclear risk	Blinding of outcome assessors was not described	Low risk	Missing data explained and balanced per outcome. Quote from text: In order to conduct an intent-to-treat (ITT) analysis, which requires that outcomes be known for all subjects, multiple imputation (MI) was used to estimate missing data, including pregnancy and infant outcomes. Levels of missing data for each outcome ranged from 12% for pregnancy result to 17% for[2 days of hospitalization.	Low risk	Findings are reported in all relevant outcomes. No published protocol available.	Low risk	No other source of bias identified
5	Tappin et al.	Financial incentives for smoking cessation in pregnancy:	Low risk	Quote from text: Helpline staff, who ascertained the primary outcome, self report of	Low risk	Flow of participants very detailed explained and withdrawal from study as well. Quote from text: Three control participants withdrew from the study immediately after randomisation and did	Low risk	All outcomes in protocol are reported in this study	Low risk	No other source of bias identified

		randomised controlled trial		smoking status in late pregnancy, were blind to allocation status.		not wish any of their data to be included in the statistical analysis. At the primary outcome assessment call, two intervention participants withdrew from the study. These participants were included in the statistical analysis, leaving 306 intervention and 303 control participants in the final analysis				
6	Flenady et al.*	My Baby's Movements: a stepped wedge cluster randomised controlled trial to raise maternal awareness of fetal movements during pregnancy study protocol	Unclear risk	Blinding of outcome assessors was not described	NA	Study protocol	NA	Study protocol	Low risk	No other source of bias identified
		My Baby's Movements: a stepped-wedge cluster-randomised controlled trial of a fetal movement awareness intervention to reduce stillbirths	Unclear risk	Blinding of outcome assessors was not described	Low risk	No missing data, no attrition in either group.	Low risk	All outcomes in protocol are reported in this study	Low risk	No other source of bias identified
	Skalecki et al.	My Baby's Movements: An assessment of the effectiveness of the My Baby's Movements phone program in reducing late-	Unclear risk	Blinding of outcome assessors was not described	Low risk	No missing data, no attrition in either group.	Low risk	Findings are reported in all relevant outcomes. No published protocol available.	Low risk	No other source of bias identified

		gestation stillbirth rates Sarah								
7	Saastad et al.	Fetal Movement Counting Improved Identification of Fetal Growth Restriction and Perinatal Outcomes - a Multi-Centre, Randomized, Controlled Trial	High-risk	Quote from text: analyses were performed by the researchers without blinding for group assignment.	Low risk	No missing data, no attrition in either group.	Unclear risk	Although more outcomes are expressed in the protocol, the publication being assessed here only focuses on Perinatal Outcomes	Low risk	No other source of bias identified
8	Lobb et al.	A controlled study of daily fetal movement counts in the prevention of stillbirths	Unclear risk	Blinding of outcome assessors was not described	Low risk	No missing data, no attrition in either group. Differences in groups explained. Quote from text: The obstetric populations attending the two units were similar except that an excess of patients with severe Rhesus iso-immunisation attended Unit B. [...] The case notes of all stillbirths were analysed retrospectively. Those which were associated with lethal fetal abnormalities, abruption, intrapartum events, Rhesus iso-immunisation and multiple pregnancies were excluded because these conditions were considered not to be preventable by the use of kick charts.	Low risk	Findings are reported in all relevant outcomes. No published protocol available.	High-risk	Base-line imbalance. Control group are formed by high-risk pregnancies which would have higher risk of experiencing the assessed outcome by definition.
9	Warland et al	Modifying Maternal Sleep Position in Late Pregnancy	High risk	No information about blinding presented in study	Low risk	Missing data is exposed and justified.	Low risk	All outcomes are reported	Low risk	No other source of bias identified

		Through Positional Therapy: A Feasibility Study								
	Kember et al.	Modifying maternal sleep position in the third trimester of pregnancy with positional therapy: a randomised pilot trial	Low risk	Quote from text: Participants, the recruiter and personnel involved in setting up, conducting, scoring and interpreting the PSG were blinded to allocation.	Low risk	No missing data	Low risk	All outcomes are reported	Low risk	No other source of bias identified

Checklist for Analytical Cross Sectional Studies – JBI (Part 1)										
Intervention n	Author	Title	Were the criteria for inclusion in the sample clearly defined?	Evidence	Were the study subjects and the setting described in detail?	Evidence	Was the exposure measured in a valid and reliable way?	Evidence	Were objective, standard criteria used for measurement of the condition?	Evidence
1	Andrews et al.	Implementation and evaluation of a quality improvement initiative to reduce late gestation stillbirths in Australia: Safer Baby Bundle study protocol.	Unclear	All women attending the participating sites will benefit from the intervention	Yes	All women attending the participating study sites are included in the sample. Settings were properly described. Quote from text: This study includes all public and private hospitals providing maternity care in NSW, VIC and QLD. Sites are grouped by level of implementation support into: (1)'targeted 'implementers, maternity hospitals recruited to and engaged with state-led SBB implementation pro-grams; (2)'non-targeted 'implementers, all other maternity hospitals in each state. For 'targeted' implementers, participation was open to maternity hospitals across the three jurisdictions, with those who expressed an interesting joining recruited. Up to August 2020, 83 sites/hospitals were recruited (VIC-	Yes	Quote from text: A mixed-methods approach will be used to assess the processes, impacts and outcomes for the SBB initiative.	Yes	Quote from text: Change in rates for the primary clinical outcome, stillbirth rate at 28 or more weeks [...] We will use a framework commonly applied to complex public health service research , adapted to suit this clinical context. This framework integrates process, impact, outcome and economic evaluations across different settings and stakeholder levels including: State level health regions,

						23 sites across all 6 health regions, NSW- 25 sites across 4 Local Health Districts andQLD- 36 sites across 17 Hospital & Health Services). These sites account for approximately half of all births in these states				maternity service providers/hospitals, quality improvement leadership teams, maternity healthcare professionals and women receiving antenatal care. Both quantitative and qualitative methods will be used to collect measures across these settings
3	Warland et al.	Keeping baby SAFE in pregnancy: Evaluating the brochure	Yes	Quote from text: Inclusion criteria were women in their final trimester of pregnancy and attending antenatal care at the participating hospital.	Unclear	Only contextual information: Women undergoing antenatal care at a small maternity hospital were recruited. This service provides a range of antenatal, birthing and post-natal care to approximately 330 women each year.	Yes	Quote from text: There was consultation regarding content, and readability from both lay and expert members of the Board of SIDS and Kids SA.	Yes	Knowledge of stillbirth was evaluated through objective questions about awareness of prevalence, risk factors, etc.
4	Widdows et al.	Saving babies' lives project impact and results evaluation (SPiRE): a mixed methodology study.	Unclear	Several NHS Trust included in the study, no specific characteristic of inclusion sample, all women attending the TRUST would be included in intervention	Yes	Quote from text: The study will be conducted in twenty NHS Hospital Trusts from eight NHS Strategic Clinical Networks. These include the South West, Thames Valley, North West Coast (formerly Lancashire and South Cumbria, and Cheshire and Merseyside), North East, Yorkshire and Humber, Greater Manchester and East Cheshire, and the East Mid-lands SCNs. Where there are multiple hospitals per NHS Trust, study sites will be asked to include data from all	Yes	Each objective has its own evaluation method and they are explicitly described in the study	Yes	Quote from text: Outcome measures (Table 1) will be collected from three primary sources; Maternity Information systems (MIS), the GROW-App™, (a subsidiary online application for the generation of the Gestation Related Optimal Weight

						relevant hospitals. The Trust will be the unit of analysis (centre) for implementation measures. However, if on re- view substantive differences are identified between sites within Trusts (e.g. restructured Trusts) sites may be split prior to evaluation of the outcome data.				(GROW) curve [18]), and targeted case note audits on selected subgroups of patients. A Data Collection Proforma (DCP) (Microsoft Excel) will be provided to all sites to facilitate collection of data by a delegated team member at each trust
	Widdows et al.	Stillbirth rates, service outcomes and costs of implementing NHS England's Saving Babies' Lives care bundle in maternity units in England: A cohort study	Unclear	We conducted a pragmatic analysis before and after the introduction of the SBL care bundle (the exposure of interest) using longitudinal data collected retrospectively in 19 NHS Trusts in England across 9 clinical networks. These trusts were purposively selected from a larger cohort of maternity units that had participated in the 2015 NHS Tracker Survey	Unclear	These trusts were purposively selected from a larger cohort ofmaternity units that had participated in the 2015 NHS Tracker Survey to provide a geo- graphically distributed sample throughout England and a range ofbirth rates and levels ofneo- natal care and to reflect a range oflevels ofimplementation at the time ofselection.	Yes	This was supplemented by clinical audits (20 sequential women per time point and Trust) ofgrowth and birth monitoring practice in unselected women (pre and post implementation), small for gestational age births (pre and post implementation) and women attending hospital with RFM (post-implementation only). Additionally, in the post intervention period women were asked to complete a survey postpartum prior to discharge	Yes	Stillbirth, defined in the UK as the death ofa baby before birth after 24 weeks' gestation

				to provide a geo- graphically distributed sample throughout England and a range of birth rates and levels of neo- natal care and to reflect a range of levels of implementation at the time of selection. Units from London were not included as they were participating in a stepped-wedge cluster trial of implementation of the Growth Assessment Protocol [12].				(informed consent) which included questions on smoking behaviour and interventions received along with experiences ofRFM		
	Widdows et al.	Evaluation of the implementation of the Saving Babies' Lives Care Bundle in early adopter NHS Trusts in England	Yes	Nineteen NHS Trusts in England took part in the evaluation, they were located across 9 clinical networks and the evaluation included both secondary and	Yes	Authors have included a table with the characteristics of all participating units.	Yes	To assess current implementation levels for SBLCB, Trusts were asked to state on a Likert scale whether each intervention in each element was implemented: a) all of the time, b) most of the time, c) half of	Yes	The primary outcome was all stillbirths, with term (≥37w) singleton stillbirths reported as a secondary outcome. We also explored the number of term stillbirths that were SGA, based on a calculated birthweight centile below the 10th

				tertiary centres. / Authors have included a table with the characteristics of all units.				the time, d) not much of the time, e) never or f) not relevant – we do not implement this intervention. A		customised centile (individual level information from electronic stillbirths submissions) or known SGA as documented in the maternal notes (case note audit).
7	Saastad et al.	Implementation of uniform information on fetal movement in a Norwegian population reduced delayed reporting of decreased fetal movement and stillbirths in primiparous women - A clinical quality improvement	No	Not explicit inclusion and exclusion criteria. Seems like all women attending clinic were eligible to participate.	Unclear	Only contextual information in text: Fourteen hospitals across both urban and rural districts, with a total of approximately 33, 000 births annually, were included in the before-and-after study.	Yes	Quote from text: Two different data collection methods were used pre- and post-intervention: 1) Prospective data collection for women presenting with DFM (DFM population), and 2) Cross-sectional studies (Cross-sectional population)	Yes	Quote from text: The primary outcome measure was maternal behavioural relation to reporting perceived absence or decreased fetal movement to the health provider; defined as threat of women waiting≥25 hours with absent feta movement or≥48 hours with DFM
	Tveit et al.	Reduction of late stillbirth with the introduction of fetal movement	Yes	Quote from text: Women with singleton pregnancies of at least 28 weeks gestation	Yes	Quote from text: In Norway, almost all pregnant women attend the antenatal program which is free of charge and covered by the public health-care services. The Norwegian antenatal care program is following	Yes	Quote from text: Pregnancy outcome were collected independently after delivery from the medical files by study	Yes	Quote from text: Main outcome was collected from medical files, secondary outcomes: severe neonatal depression,

		information and guidelines – a clinical quality improvement		or more who reported a concern for DFM (either by spontaneous reporting or upon questioning), were registered prospectively for quality-assurance purposes at 14 delivery units in eastern Norway and the city of Bergen. The registrations were a part of the international collaboration, Fetal Movement Intervention Assessment.		contemporary guidelines composed by the National Institute of Clinical Excellence [28]. The community midwives and general practitioners are in charge of the antenatal program, and without the possibility to perform an NST or ultrasound examinations locally, they usually refer the concerned mothers to the nearby hospital with a maternity ward. Hence, the pregnant women in Norway typically contact maternity wards directly with any acute concerns for DFM. There are no private delivery wards in Norway.		coordinator at each hospital.		defined as Apgar score of < 3 at 5 minutes postpartum; symptoms of multisystem organ failure and pH < 7 in the umbilical artery or fetal capillary scalp, if obtained; preterm birth (28° – 366 weeks); FGR (< 10th percentile of birthweight adjusted for gender and mother's height, weight, parity, and ethnicity) [32]; fetal heart rate tracings judged clinically as nonreasoning and leading to intervention in labour; oligohydramnios defined as an amniotic fluid index of < 5 cm or at <2.5th percentile; polyhydramnios defined as an amniotic fluid index of > 25 cm or at >97.5th percentile; investigations undertaken for reduced FM; and examinations of DFM resulting in immediate admission for

										induction of labor or caesarean section. Outcomes related to maternal behavior were: the number of women waiting more than 24 hours with an absence of FM or more than 48 hours with a decrease of FM before contacting health-care professionals.

Checklist for Analytical Cross Sectional Studies – JBI (Part 2)									
Intervention n	Author	Were confounding factors identified?	Evidence	Were strategies to deal with confounding factors stated?	Evidence	Were the outcomes measured in a valid and reliable way?	Evidence	Was appropriate statistical analysis used?	Evidence
1	Andrews et al	Unclear	No information about confounding facots	Unclear	No information about confounding factors	NA	This is a protocol study. Measures have not been undertaken yet.	NA	This is a protocol study. Measures have not been undertaken yet.
3	Warland et al.	No	Only cofounding factor considered was previous experience of stillbirth, these women were excluded from study	No	Only cofounding factor considered was previous experience of stillbirth, these women were excluded from study	Yes	Quote from text: There was no existing tool to assess knowledge of stillbirth, therefore a questionnaire specifically for this project was developed. The first part of the questionnaire included maternal demographical information such as age, level of education, parity and main care provider.	Yes	Quote from text: In this study power calculations were conducted which estimated that 23 women, who were their own controls, would detect a one sample correlation with an a of 0.05 and power of 80%. Information gained from the first and second questionnaires was entered into SPSS (statistical package social sciences version 17). Data were analysed using a repeated measures student t-test.

							Questions concerning women's knowledge of stillbirth consisted of one multiple choice and three free text 'comment' boxes.		
4	Widdows et al.	Unclear	No information about confounding factors	Unclear	No information about confounding factors	Yes	Each objective has its own evaluation method and they are explicitly described in the study	Yes	Quote from text: Electronic data on stillbirths other clinical outcomes and smoking rates was provided as, or converted to, monthly counts of outcomes and appropriate denominators. Outcomes were fit- ted using quasi-binomial models to allow for the possibility of overdispersion, except for scan rates, which were fitted using quasi-Poisson models. Generalised linear models with logarithm- mic link functions were used to estimate a linear trend over time. Trust was included as a covariate thus time trends are estimated on a within-Trust basis. From this model we derived estimates of the outcome rates at dates 2 years either side of the nominal SBL start date of April 2015 and estimated the risk- (or rate) ratio between these two time points as a measure ofthe change over the implementation period.
	Widdows et al.	Unclear	No information about confounding factors	Unclear	No information about confounding factors	Yes	Electronic data on stillbirths other clinical outcomes and smoking rates was provided as, or converted to, monthly counts of outcomes	Yes	Statistical analysis is widely described in paper.

							and appropriate denominators.		
	Widdows et al.	Unclear	No information about confounding factors	Unclear	No information about confounding factors	Yes	Data on stillbirths (defined as a baby delivered at or after 24+0 weeks gestation showing no signs of life, irrespective of when the death occurred) was primarily collected from clinical audit. Intervention outcomes and resource use associated with SBLCB were collected from clinical audit and surveys with patients and healthcare professionals. Information on implementation levels was obtained from surveys with organisational leads at each Trust.	Yes	
7	Saastad et al.	Yes	Maternal characteristics were identified as potential co-founders. Quote from text: Subgroup analyses were	Yes	Adjusted OR were calculated. Quote from text: Crude and adjusted odds ratios (ORs)with 95% confidence intervals (CIs) were estimated, and variables with associations with a $p < 0.20$	Unclear	4 of the 6 secondary outcomes rely on self-report of participants.	Yes	Quote from text: Crude and adjusted odds ratios (ORs)with 95% confidence intervals (CIs) were estimated, and variables with associations with a $p < 0.20$ in univariate analyses were included in the multivariate models

			undertaken according to: maternal age [5,26], body mass index (BMI) [5,27],smoking habits [5,28], and maternal country of origin[29] and according to subgroups of Western and non-Western origin (due to higher rates of stillbirths among on-Western women in our community)		in univariate analyses were included in the multivariate models				[43].Chi square tests were used for estimating differences between proportions of categorical variables. A p-value<0.05 was considered statistically significant. Bonferroni corrections were performed in the multiple compare-sons.
	Tveit et al.	Yes	Quote from text: In the multivariate analysis, all outcomes were adjusted for potential confounding factors – such as maternal age, body mass index (BMI), smoking habits, parity, and ethnicity – due to prior knowledge of their impact on pregnancy outcomes and health-promoting behavior.	Yes	Quote from text: All statistical analysis were performed with SPSS version 15.0. (SPSS Chicago, IL, USA) using cross tabulations, with χ2 tests and logistic regressions to find crude (unadjusted) and adjusted odds ratios (OR) with 95% confidence intervals	Yes	Main outcome was collected from medical files, secondary outcomes: severe neonatal depression, defined as Apgar score of < 3 at 5 minutes postpartum; symptoms of multisystem organ failure and pH < 7 in the umbilical artery or fetal capillary scalp, if obtained; preterm birth (28° – 366 weeks); FGR (< 10th percentile of birthweight adjusted for gender and mother's height, weight, parity, and ethnicity) [32]; fetal	Yes	Quote from text: All statistical analysis were performed with SPSS version 15.0. (SPSS Chicago, IL, USA) using cross tabulations, with χ2 tests and logistic regressions to find crude (unadjusted) and adjusted odds ratios (OR) with 95% confidence intervals (CI). The level of statistical significance was set at p < 0.05.

							heart rate tracings judged clinically as nonreasoning and leading to intervention in labour; oligohydramnios defined as an amniotic fluid index of < 5 cm or at <2.5th percentile; polyhydramnios defined as an amniotic fluid index of > 25 cm or at >97.5th percentile; investigations undertaken for reduced FM; and examinations of DFM resulting in immediate admission for induction of labour or caesarean section. Outcomes related to maternal behaviour were: the number of women waiting more than 24 hours with an absence of FM or more than 48 hours with a decrease of FM before contacting health-care professionals.		

AACODS checklist (Authority, Accuracy, Coverage, Objectivity, Date, Significance)														
Intervention n	Author	Title	Authority	Evidence	Accuracy	Evidence	Coverage	Evidence	Objectivity	Evidence	Date	Evidence	Significance	Evidence
1	Andrews et al.	Stillbirth in Australia 2: Working together to reduce stillbirth in Australia: The Safer Baby Bundle initiative	Yes	Authors are associated to reputable Universities and Research Centres	Yes	The is clear, the study has been peer reviewed, references and sources are credible, reference to original materials are accurate	Yes	Limits are clearly stated	Yes	The paper is descriptive of an intervention, no source of bias identifiable	Yes	The work is dated in 2020	Yes	The item is meaningful and it adds value to the content of this review by providing extra information regarding the Safer Baby Bundle initiative.
4	NHS	Saving Babies' Lives Version Two. A care bundle for reducing perinatal mortality	Yes	Authors are associated to reputable organisations	Yes	Aims and methodology are clear.	Yes	Limits are clearly stated	Yes	The report is descriptive of an intervention, no source of bias identifiable	Yes	The work is dated in 2020	Yes	The item is meaningful and it adds value to the content of this review by providing extra information regarding the Safer Baby Bundle initiative.

Supplementary file 8.5 – Behaviour Change Techniques coding with evidence

BCT Category	BCT	Evidence from article / Quotes from test
1.The safer baby bundle (Andrews, Ellwood, Gordon, et al., 2020; Andrews, Ellwood, Middleton, et al., 2020)		
2. Monitoring and feedkback	2.1 Monitoring of behaviour	women expect to be assessed for tobacco use; regardless of smoking status, offer all women an exhaled breath carbon monoxide (CO) reading (and their partners where available). for all women, at the 28 weeks' gestation visit, record CO reading
	2.5 Monitoring of outcome of behaviour	This includes assessing women for risk factors for FGR at the first antenatal care visit and at every subsequent antenatal visit from 24 weeks' gestation
5. Natural consequences	5.1 Information about health consequences	if smoking, provide brief advice on the benefits of quitting
		As outlined by paper 4 of this series, community campaigns in New Zealand, the UK, and Australia have recently released public health messages around the importance of women going-to-sleep on their side in late pregnancy to reduce stillbirth. [...] provision of information and advice to pregnant women in their care about the importance of going to sleep on your side from week 28 of pregnancy
11. Regulation	11.1 Pharmacological support	nicotine replacement therapy may be offered. consider offering or arranging prescriptions for NRT, following a discussion of risks and benefits and in consultation with other relevant health workers.
4. Shaping knowledge	4.1 Instructions on how to perform the behaviour	explain the best way to quit, including support sessions through Quitline, and NRT if appropriate.
1. Goals and planning	1.1 Goal setting (behaviour)	offer personalised advice on how to stop smoking (e.g. setting quit date, making quit plan)
3. Social support	3.2 Social support (practical)	refer to Quitline (or other locally appropriate smoking cessation service) if identified as smoker and/or CO level is elevated (4ppm or above). offer personalised advice on how to stop smoking and available behavioural intervention services (e.g. Quitline) to support

		quitting, including details on when, where and how to access them
12. Antecedents	12.5 Adding objects to the environment	Provide information brochure* and advice on DFM to all pregnant women by the 28th week of pregnancy and remind women of the importance of reporting DFM at subsequent contacts and discuss with women the importance of being aware of DFM and to report concerns without delay
		Information brochure on safe going-to-sleep position* based on current evidence to be provided to all pregnant women by week 28 of pregnancy, and the importance of going to sleep on their side discussed at every subsequent contact.
2. An Integrated Randomized Intervention to Reduce Behavioral and Psychosocial Risks (Katz et al., 2008; Subramanian et al., 2012)		
3. Social support	3.1 Social support (unspecified)	Individualized counselling sessions were tailored to the specific risks reported by each woman.
		Women in need of other social services were provided with referrals to specific resources.
		1) Ask about smoking status, 2) Advise to quit, 3) Assess willingness to quit, 4) Assist in ways to quit, and 5) Arrange for follow-up
		CBT approaches were used, along with motivational interviewing
1. Goals and planning	1.2 Problem solving	identifying smoking triggers
		Once identified, women were encouraged to avoid triggers and educated in the use of alternative coping and behavioral change strategies.
	1.4 Action planning	developing strategies for mood management
		3) Assess willingness to quit, 4) Assist in ways to quit, and 5) Arrange for follow-up
5. Natural consequences	5.1 Information about health consequences	This intervention included content addressing both active smoking and smoke exposure
		All women randomized to the intervention group, irrespective of risk status, received information at their first postpartum session about the risks of ETSE for their new-born infants, and how to protect their infants from harm.

12. Antecedents	12.5 Adding objects to the environment	Adapted SCRIPT materials were presented at the first visit, and included viewing a 6-minute version of the "Commit to Quit" videotape, and giving women "A Pregnant Woman's Guide to Quit Smoking" for home use.
	12.2 Reestructuring social environment	as well as skills practice and role plays for negotiation with partners and household members who smoked.
		The focus for women who reported ETSE during pregnancy included 1) ETSE avoidance and reduction, and 2) changing the surrounding environment through negotiation skills and creating household smoking bans.
2. Feedback and monitoring	2.3 Self-monitoring of behaviour	Participants were also encouraged to self-monitor smoking habits and identify common smoking triggers, including ETSE.
	2.2 Feedback on behaviour	All women randomized to the intervention group, irrespective of risk status, received information at their first postpartum session about the risks of ETSE for their newborn infants, and how to protect their infants from harm.
10. Reward and threat	10.4 Social reward	Counselors provided reinforcement for successive changes toward smoke avoidance and smoking cessation/reduction
7. Associations	7.5 Remove aversive stimulus	The focus for women who reported ETSE during pregnancy included 1) ETSE avoidance and reduction, and 2) changing the surrounding environment through negotiation skills and creating household smoking bans.
4. Shaping knowledge	4.1 Instructions on how to perform the behaviour	All women randomized to the intervention group, irrespective of risk status, received information at their first postpartum session about the risks of ETSE for their newborn infants, and how to protect their infants from harm.
3. Keeping baby SAFE in pregnancy (Warland, 2013)		
5. Natural consequences	5.1 Information about health consequences	finally gave them the stillbirth information brochure just as they left their appointment.
		The best sleeping position for you is the left side. The left side position allows maximum blood flow to your baby and helps reduce the risk of stillbirth.
		How often should my baby move? Every baby has their own pattern of activity, and there's no correct one. It is not necessary to try to count movements but to simply tune into and

		become aware of your individual baby's normal sleep and wake cycle. It is common for babies to gradually become less vigorous with their kicks closer to the birth, due to less space.
	5.2 Salience of consequences	Avoid sleeping on your back: because the baby's weight presses on the main vein that returns blood from the lower body to the heart. It also puts full weight on your back and intestines, which can cause you discomfort and increase the risk of you developing backaches, and indigestion.
12. Antecedents	12.5 Adding objects to the environment	finally gave them the stillbirth information brochure just as they left their appointment.
6. Comparison of behaviour	6.2 Social comparison	Images in brochure of woman engaging in behaviours
		Tucking a pillow behind your back, may help you if you are afraid of lying on your back when you are asleep. Some women find it helpful to place a small pillow or cushion between their knees when lying on their side.
4. Shaping knowledge	4.1 Instructions on how to perform the behaviour	You should lie down to sleep on your left each night in the last 3 months of pregnancy. Do not worry about shifting positions once you have fallen asleep, as this is a natural part of sleeping.
3. Social support	3.3 Social support (emotional)	Keeping these appointments also gives you the chance to build trust with your midwife or doctor and talk through any worries you may have and ask questions.
16. Covert learning	16.3 Vicarous consequence	Women who have experienced a stillbirth often say they felt there was something wrong but, for some reason, didn't contact their doctor or midwife until it was too late.
1. Goals and planning	1.4 Action planning	However, if your baby is moving less than they normally do then you should ring your midwife or doctor right away.
		Between your appointments you should immediately contact your Midwife or Obstetrician if you are: • spotting or bleeding, • leaking fluid, • swelling in your fingers or ankles, • having headaches, blurry vision or seeing spots, • feeling nauseous or vomiting (once morning sickness is over) • having a lot more back or stomach ache than is usual for you • excessively itchy • feeling

		unwell in any way • feeling there is something wrong even if you don't quite know what it is. Trust your maternal instincts and quickly arrange to see your midwife or obstetrician
4. Saving babies' lives project (NHS England, 2019; Warland, 2013; Widdows, Reid, et al., 2018; Widdows, Roberts, et al., 2018)		
2. Feedback and monitoring	2.6 Biofeedback	Reducing smoking in pregnancy by assessing exposure to carbon monoxide (CO) as appropriate to assist in identifying smokers (or those exposed to CO through other sources). CO testing of all pregnant women at booking appointment. 1.1 CO testing should be offered to all pregnant women at the antenatal booking appointment, with the outcome recorded. 1.2 Additional CO testing should be offered to pregnant women as appropriate throughout pregnancy, with the outcome recorded. 1.3 CO testing should be offered to all pregnant women at the 36 week antenatal appointment, with the outcome recorded.
	2.2 Feedback on behaviour	Referral pathway must include feedback and follow up processes.
	2.1 Monitoring of behaviour	
3. Social support	3.1 Social support (unspecified)	Referral of all women identified as smokers to smoking cessation services on an opt out basis. Referral for those with elevated levels (4ppm or above) for support from a trained stop smoking specialist, based on an opt-out system. Referral pathway must include feedback and follow up processes.
5. Natural consequences	5.1 Information about health consequences	3.1 Information from practitioners, accompanied by an advice leaflet (for example, RCOG or Tommy's leaflet) on RFM, based on current evidence, best practice and clinical guidelines, to be provided to all pregnant women by 28+0 weeks of pregnancy and RFM discussed at every subsequent contact.
12. Antecedents	12.5 Adding objects to the environment	Information and advice leaflet on RFM to be provided to all women by 24 weeks pregnancy
5. Financial incentives for smoking cessation in pregnancy: randomised controlled trial (Tappin et al., 2015)		
5. Natural consequencces	5.1 Information about health consequences	The advisers offer a one hour appointment to discuss smoking and cessation. This face to face contact is followed by four weekly telephone support calls

11. Regulation	11.1 Pharmacological support	and free nicotine replacement therapy from local pharmacies for 10 weeks.
10. Reward and threat	10.1 Material incentive (behaviour)	The incentives group was offered the same routine support plus up to £400 of shopping vouchers (Love2shop) for engaging with stop smoking services or for quitting during pregnancy, or both
	10.2 Maternial reward (behaviour)	Intervention participants received £50 of vouchers if they attended their face to face appointment and set a quit date. As per routine practice, clients who set a quit date were telephoned by their stop smoking services adviser four weeks later.
	10.10 Reward (outcome)	Intervention participants who reported abstinence (that is, had not smoked, even a puff, in the past two weeks) were visited at home by a research nurse to confirm smoking status (carbon monoxide breath test result <10 ppm). Confirmed quitters were sent a further £50 voucher. Twelve weeks after stopping smoking, women in the incentives group who were quitters at four weeks were contacted by stop smoking services (routine practice) and, if confirmed to be abstinent (carbon monoxide breath test result <10 ppm), were sent a £100 voucher.
1. Goals and planning	1.1 Goal setting (behaviour)	Intervention participants received £50 of vouchers if they attended their face to face appointment and **set a quit date.**
3. Social support	3.1 Social support (unspecified)	specialist pregnancy support by the stop smoking services
6. My Baby's Movements (Flenady et al., 2019)		
5. Natural consequences	5.1 Information about health consequences	The messages include facts about fetal movements and a prompt to contact her healthcare provider if concerned about her baby's movements.
7. Associations	7.1 Prompt/cues	The MBM app sends an alert to prompt the woman's awareness of her baby's movements at a time and frequency of her choice, from 28 week's gestation until birth.
3. Social support	3.1 Social support (unspecified)	If concerned, she is encouraged to contact her health care provider without delay.

2. Feedback and monitoring	2.3 Self-monitoring of behaviour 2.4 Self-monitoring of outcomes of behaviour	The app also provides a 'movement counter' option where the woman can record the number of movements she feels over a two-hour period, and a strength recorder option where she can record the strength of her baby's movements if she wishes.
7. Reduction of late stillbirth with the introduction of fetal movement information and guidelines (Saastad et al., 2010, 2011; Tveit et al., 2009)		
5. Natural consequences	5.1 Information about health consequences	We developed a brochure of information that aimed to increase maternal awareness and vigilance to significant decreases in fetal activity, and to aid health-promoting behavior.
		Your baby gets everything it needs from you through the placenta. As long as the supplies are good the baby will kick. If the placenta does not supply enough or the baby becomes ill, it must save energy to continue to grow, so there are fewer kicks. If you smoke, both the placenta and your baby are affected, and there will be less kicking. If the placenta becomes very weak, growth is inhibited, and your baby could become seriously ill or injured. Your baby also may have trouble during birth. This seldom happens, though, if your baby is kicking normally! Sometimes your baby is calm. It is important that you notice, though, when it kicks much less than usual. When you feel the normal life rhythm in there, that is a sign your baby is fine. By feeling for the kicks you are connecting with your baby. That's why it's good to count kicks!
		The brochure covered information on: expected normal fetal activity [31]; differences in perception according to different fetal movements [31], maternal position [32], the inter- and intraindividual variation between fetuses [9], maternal weight [27], and smoking [33]; interpretation of variation of fetal activity; instructions on how to use the kick chart; and when to contact health professionals if experiencing DFM [11]
3. Social support	3.1 Social support (unspecified)	In some situations the woman was advised to contact health-professionals for further examinations: 1) never to wait to the next day if the baby did not kick one day or, 2) if the baby kicked less and less in the course of a day/days, or 3) if she felt less than ten FM in 2

		hours at a time of the day when the baby was usually active, and she perceived this as a reduction
		A midwife or an obstetrician from the participating hospitals or the research study group called women in the intervention group within two weeks after counting-start to support them in the interpretation of the counting method
1. Goals and planning	1.4 Action planning	In some situations the woman was advised to contact health-professionals for further examinations: 1) never to wait to the next day if the baby did not kick one day or, 2) if the baby kicked less and less in the course of a day/days, or 3) if she felt less than ten FM in 2 hours at a time of the day when the baby was usually active, and she perceived this as a reduction
12 Antecedents	12.5 Adding objects to the environment	As a guide to help the women to identify DFM, an invitation to use a kick chart was included.
		A modified “count-to-ten” chart was chosen, as this has the highest compliance and acceptance rates.
		We developed a brochure of information that aimed to increase maternal awareness and vigilance to significant decreases in fetal activity, and to aid health-promoting behavior.
2. Feedback and monitoring	2.4 Self-monitoring of outcome of behaviour	As a guide to help the women to identify DFM, an invitation to use a kick chart was included.
4. Shaping knowledge	4.1 Instructions on how to perform a behaviour	Start counting the kicks in the 28th week of your pregnancy. Write in the date you start keeping count and your expected delivery date on the form. You should count how long it takes your baby to kick 10 times, starting with the first kick (so you know your baby is awake). All movements count as a "kick" but don't count hiccups. Several movements at the same time count as one "kick." The quickest way to do this is to relax, lie or sit down and concentrate on feeling for kicks. If your baby is asleep, you can wake it with a little clapping or gentle push on your stomach or by drinking something cold. When you're finished counting record the time in the boxes provided on the form.Count at approximately the same time of day every day - if you can. Choose a time of day when you know you have time to spare and when your baby

		usually is active - preferably in the morning before you get up. Start counting within the same two-hour period every day and be sure to write on the form whatever hours you choose. It will take most mothers less than 15 minutes to count kicks this way
		The brochure covered information on: expected normal fetal activity [31]; differences in perception according to different fetal movements [31], maternal position [32], the inter- and intraindividual variation between fetuses [9], maternal weight [27], and smoking [33]; interpretation of variation of fetal activity; instructions on how to use the kick chart; and when to contact health professionals if experiencing DFM [11]
1. Goals and planning	1.4 Action planning	The brochure covered information on: expected normal fetal activity [31]; differences in perception according to different fetal movements [31], maternal position [32], the inter- and intraindividual variation between fetuses [9], maternal weight [27], and smoking [33]; interpretation of variation of fetal activity; instructions on how to use the kick chart; and when to contact health professionals if experiencing DFM [11]
		They were instructed not to wait until the next day if they perceived complete absence of fetal activity or if they felt a significant and sustained decrease.
2. Feedback and monitoring	2.4 Self-monitoring of outcome of behaviour	A modified "count-to-ten" chart was chosen, as this has the highest compliance and acceptance rates.
	2.3 Self-monitoring of behaviour	
8. A controlled study of daily fetal movement counts in the prevention of stillbirths (Lobb et al., 1985)		
12. Antecedents	12.5 Adding objects to the environment	The Cardiff 'Count to Ten Kick Chart' was introduced by two consultant obstetricians (Unit A) at the Liverpool Maternity Hospital during 1977, so that by late 1977 all women booked for confinement with Unit A were given a kick chart at about 28 weeks pregnant.
4. Shaping knowledge	4.1 Instructions on how to perform the behaviour	The kick chart was explained to the women by an obstetrician or a midwife.
1. Goals and planing	1.4 Action planning	A standard Cardiff chart was used, but the instructions were amended verbally in that women were asked to telephone the hospital at 21.00 hours, rather than at 09.00 hours the

		following day. if they had less than 10 fetal movements for 2 days, or no fetal movements for 12 hours.
9. Modifying Maternal Sleep Position in Late Pregnancy Through Positional Therapy: A Feasibility Study (Warland, 2018)		
12. Antecedents	12.5 Adding elements to the environment	The PrenaBelt is designed to cause subtle pressure points on the pregnant user's lower back and buttocks when she lies in the supine position and thereby activate her body's natural mechanism to spontaneously reposition to maintain comfort.
7. Associations	7.1 Prompts/cues	