Ollscoil na hÉireann, Corcaigh

National University of Ireland, Cork

The Development and Piloting of a Discrete Choice Experiment to Measure the Support Preferences of People with Chronic Respiratory Diseases in Ireland

This thesis is submitted by

Selena O'Connell

for the degree of Doctor of Philosophy

October 2020

Supervisors: Professor Eileen Savage, Dr Vera McCarthy & Dr Desmond Murphy

Head of Department: Professor Josephine Hegarty

Catherine McAuley School of Nursing and Midwifery

College of Medicine and Health

University College Cork

“There is no such thing as a fixed policy, because policy, like all organic entities, is always in the making” - Lord Salisbury

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. I have read and understood the regulations of University College Cork concerning plagiarism.

Signed: Selena O’Connell

Date: 26/09/20

Selena O'Connell

BA Psychology

Abstract

Introduction: Healthcare systems are increasingly developing self-management support (SMS) frameworks to reduce the burden of chronic disease. This involves a whole-system approach which enables the person with chronic disease to actively participate in managing their wellbeing. A framework to support the implementation of SMS was launched in Ireland, with asthma and COPD among the chronic diseases targeted. As this framework is being implemented in Ireland, there is a need to ensure that patient preferences are considered.

Aim: to examine the SMS preferences of people with asthma and/or COPD in Ireland through the development and piloting of a discrete choice experiment (DCE).

Methods: Early work in the thesis identified and explored eight frameworks for SMS of chronic disease developed by national/state level health systems in different countries. Subsequently, a qualitative study was conducted with the implementation leads of three frameworks. This research indicated that greater input of people with chronic disease was important to advance SMS. Steps were taken to develop a DCE to measure the relative importance of support features from the perspectives of people with asthma/COPD in accordance with good practice. Firstly, a systematic review and thematic synthesis of qualitative studies was conducted in order to inform the attributes of the DCE. Secondly, a qualitative descriptive study using interviews and focus groups was conducted with people with asthma/COPD in Ireland (n = 20), 10 of whom had asthma. The qualitative literature along with clinical and methodological considerations informed the design of the DCE. The DCE was refined through cognitive interviews (n = 16). A pilot study was conducted (n = 38) and a multinomial logit model was used to examine the strength and direction of preferences.

Findings: Fifteen articles were included in the thematic synthesis and three themes were identified: *types of support*, *the support relationship* and

accessibility. Similar themes were identified in the qualitative descriptive study. In terms of accessibility, participants highlighted the need for both scheduled and flexible unscheduled access to support from providers with specialist knowledge. They valued comprehensive and person-centred consultations and a person-provider relationship which facilitated patient input and was characterised by continuity over time. A DCE which focused on support in managing symptoms of an asthma flare-up was developed where participants made choices between services that differed on six attributes. Pilot study data indicated that having a provider that listens to the patient's concerns was most important when experiencing symptoms that may lead to an asthma flare-up. Specialist providers who can be accessed face-to-face on the same day as contacted and have previous knowledge of the patient's history were also valued. Having a written plan to manage changes was less valued.

Conclusions: The implementation of frameworks on SMS for chronic disease requires long-term buy-in from stakeholders and increased consultation with patients and providers. Qualitative research with people with asthma/COPD illuminated valued features of support such as communication and continuity in the patient-provider relationship that warrant greater attention in SMS interventions. The findings of the DCE pilot study need to be verified through conducting a study with adequate power to compare preferences across groups and to use more advanced models to account for preference heterogeneity. The findings suggest a model of support which includes timely, face-to-face access to a healthcare professional that knows the patient and fully listens to their concerns for managing symptoms of an asthma flare-up.

Acknowledgements

Firstly, I would like to thank my supervisors - Professor Eileen Savage, Dr Vera McCarthy and Dr Desmond Murphy - who have given me wonderful support and guidance throughout my PhD journey. They provided invaluable insight from various research and clinical perspectives to help advance the work. I would like to thank Dr Sarah Barry for contributing her expertise on health policy analysis and I would like to thank Dr Michelle Queally for her guidance in discrete choice methods employed in this thesis.

I would also like to thank the Health Service Executive (HSE) Programme for Health Service Improvement for funding this PhD and for their support throughout the project. The regular group updates with students and supervisors were vital in advancing my study as well as linking it with practice and for this I am very grateful. Thanks to the staff and students at Catherine McCauley School of Nursing and Midwifery who are always so welcoming and friendly.

A big thank you to all who participated in the phases of this PhD study including implementation leads of SMS frameworks and the people with asthma and COPD in Ireland who were generous with their time and in sharing their experience and preferences, without whom this study would not be possible.

Thank you to members of the respiratory team and staff at asthma clinics in Cork University Hospital; the Asthma Society of Ireland; and COPD Support Ireland for their help in facilitating this study.

Finally, thanks to my family and friends for their support and all the little things that made my life easier on this PhD journey.

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List of Abbreviations

ACQ	Asthma Control Questionnaire
ACT	Asthma Control Test
CAT	COPD Assessment Test
COPD	Chronic obstructive pulmonary disease
COREQ	Consolidated criteria for reporting qualitative research
DCE	Discrete Choice Experiment
ED	Emergency Department
ENTREQ	Enhancing Transparency in Reporting Synthesis of Qualitative Research
GINA	Global Initiative for Asthma
GOLD	Global Initiative for Chronic Obstructive Lung Disease
GP	General practitioner
HCP	Healthcare professional
HRQoL	Health-related quality of life
IRL	Ireland
JBI	Joanna Briggs Institute
IT	Information Technology
MB	Manitoba
MMNL	Mixed Multinomial Logit Model
MNL	Multinomial Logit Model
NT	Northern Territory
OECD	Organisation for Economic Co-operation and Development
PPI	Public and patient Involvement
QLD	Queensland
SL	Scotland
SMS	Self-management support
SRQR	Standards for Reporting Qualitative Research
TAS	Tasmania
WA	Western Australia
WHO	World Health Organisation
WL	Wales

Introduction to the Research

The overarching aim of the research presented in this PhD thesis by publication was to develop and pilot a discrete choice experiment (DCE) to measure the support preferences of people with chronic respiratory diseases in Ireland. Although the research started out with a focus on both chronic obstructive pulmonary disease (COPD) and asthma, the focus over time became specific to asthma.

This study arose within the context of growing policy discourse in Ireland around chronic disease management prompted by the increasing prevalence of chronic conditions and the burden of these on the lives of individuals and on the health system. In 2013, the Irish government launched *Healthy Ireland: A Framework for Improved Health and Wellbeing 2013 – 2025.* This framework outlined a vision to enhance the health of all people in Ireland taking a life-course approach to health across society (Department of Health, 2013). In the *Healthy Ireland* framework, it was noted that "chronic diseases will undoubtedly lead Ireland toward an unhealthy and extremely costly, if not unaffordable, future" (Department of Health, 2013, p. 6). Action to create change and address negative health trends was called for in this framework. One of the actions proposed was 'empowering people and communities' stating that "it is essential to focus on effective ways to empower people and communities to improve and take responsibility for their own health and wellbeing" (Department of Health, 2013, p.24).

In Ireland, operational responsibility for implementing government policy in relation to healthcare rests with the national healthcare system, that is, the Health Service Executive (HSE). In response to the *Healthy Ireland* framework, the HSE published a national implementation plan in 2015. This plan prioritised reducing the burden of chronic disease, and the development of a national framework for self-management support (SMS) to achieve this priority (Health Service Executive, 2015a). Accordingly, a national framework was subsequently developed and launched in Ireland, titled, *Living Well with a Chronic Condition: Framework for Self-management Support* (Chronic Conditions Working Group, 2017). This framework identified approaches to SMS and actions at various levels of health system to implement SMS for chronic disease. Respiratory conditions (COPD and asthma) were among the diseases prioritised in this framework, the others being diabetes and cardiovascular diseases (Chronic Conditions Working Group, 2017). The framework for SMS espoused a 'person-centred care' approach which places service users at the centre of service delivery and involves "respecting their values, preferences and diversity and involving them in the provision of care" (Chronic Conditions Working Group, 2017, p.50).

A more detailed account of the background context for the research in this thesis is presented in Chapter 1. The research began by systematically examining frameworks on SMS for chronic disease in an international context. A document analysis of eight SMS frameworks across different countries found variation in the extent to which patients were involved in framework development at policy level. This analysis is presented in Chapter 2 of the thesis. As presented in chapter 3, a follow-up qualitative study with key

informants who led the implementation of SMS frameworks in their respective countries indicated that understanding and representing the perspectives of people with chronic disease was needed and would be key to future implementation of SMS in health services.

In an effort to advance methods to assist in understanding the preferences of people with chronic disease, a DCE was developed and piloted. A DCE is a method of measuring the relative importance of service features from the perspectives of service users. COPD and asthma were chosen as the focus of this work for a number of reasons: the prevalence and impact of asthma and COPD in Ireland (further detailed in Chapter 1); recent prioritisation of these conditions as targets for SMS in Ireland (Chronic Conditions Working Group, 2017); and international reviews of SMS and chronic disease management interventions which highlight that disease management interventions for asthma and COPD have been less frequently studied than diabetes and other cardiovascular diseases (Dineen-Griffin et al., 2019; Nolte & Hinrichs, 2012; Reynolds et al., 2018).

Three additional studies were conducted up to the point of piloting the DCE. A systematic review and thematic synthesis of qualitative studies on SMS preferences of people with asthma or COPD was conducted. This study is presented in chapter 4. Then, a qualitative descriptive study was conducted with adults who had asthma or COPD to explore their preferences for SMS. The decision to further limit the development of the DCE to the preferences of people with asthma was made following this qualitative study, as explained in Chapter 6. Methodological implications of developing and piloting a DCE study

were considered and are presented in Chapter 6. Finally, the DCE was piloted with people with asthma, as presented in Chapter 7, representing the final phase of the research in this thesis. A more detailed account of the contents of all chapters is provided in the first chapter of the thesis following a background context to situate this research.

Chapter 1. Background

This chapter provides an introduction to the literature pertinent to the thesis. It provides an account of the burden of chronic disease, particularly asthma and COPD, which are the focus of this thesis, albeit noting that the thesis eventually evolved into focusing on asthma. The development of the concept of self-management support (SMS) as a pillar of chronic disease management is detailed. It is highlighted that SMS requires a systems approach to implementation. This approach has been developed through national-level policy and framework documents. Potential challenges of implementing such frameworks are identified and literature on policy implementation is introduced. The value of the patient perspective to inform policy and healthcare relating to SMS is highlighted. The discrete choice experiment is introduced as a method to measure preferences for SMS from the perspectives of people with chronic disease. Lastly, the overall layout of the thesis is outlined.

1.1 Burden of chronic disease

Globally chronic disease is a major public health problem. Developments in healthcare have led to longer life expectancy and thus an increasing aging population. Due to the increasing aging population, the prevalence of chronic diseases is increasing worldwide (Vos et al., 2016). This was indicated by the Global Burden of Disease Study where the prevalence of years lived with disability increased from 2005 to 2015 with chronic diseases such as diabetes, asthma, COPD and mental health conditions becoming leading contributors to years lived with disability (Vos et al., 2016). Chronic diseases impact on the

wellbeing of individuals and their quality of life. People with chronic diseases experience reduced health-related quality of life (HRQoL) including both physical and mental HRQoL (Cappa et al., 2019; Singh et al., 2017; Wang et al., 2008). Furthermore, chronic conditions account for an estimated 70% of all deaths worldwide (World Health Organisation, 2017). Cardiovascular diseases, cancers, respiratory diseases and diabetes together account for just over 80% of mortalities attributed to chronic conditions (World Health Organisation, 2017).

Chronic diseases also take up a large proportion of health expenditure in many countries (Muka et al., 2015). A review of studies assessing the economic impact of non-communicable diseases found that chronic diseases influence expenditure in different ways including increases in the volume of services provided and technologies to assist service provision (Muka et al., 2015). Depending on the severity or impact of chronic disease, individuals use of health services can increase including primary care visits, specialist visits, diagnostic testing, emergency department (ED) presentations and hospitalisations (Ding et al., 2017; Shaw et al., 2018; Suruki et al., 2017). Multimorbidity also requires consideration with evidence that costs increase with every additional chronic condition that an individual has due to increased health service utilisation (Hajat & Stein, 2018). In addition, chronic diseases contribute to a loss in national income due to absenteeism and inability to work (Muka et al., 2015). Improved chronic disease management has thus become a global priority in order to address the multifaceted burden of chronic disease (World Health Organisation, 2014).

In Ireland, chronic disease is a major challenge that mirrors global trends. It has been estimated that 38% of people have a chronic disease (Health Service Executive, 2017). Figures from recent *Healthy Ireland Surveys* indicate that 32% of people have a long-term condition lasting six months or more (Department of Health, 2019b). These findings were based on 7,413 interview-administered questionnaires providing geographical representation of Ireland. It is estimated that chronic conditions contribute to two of every five hospitalisations in Ireland (Health Service Executive, 2017). Reducing the burden of chronic disease has become a policy priority in Ireland with *Tackling Chronic Disease: a Policy Framework for the Management of Chronic Diseases* released in 2008 (Department of Health and Children, 2008). National Clinical Programmes for specific chronic diseases were established within the HSE in 2010 to enhance care provided for chronic conditions (Darker et al., 2018). More recently, chronic disease management has been identified as one of three strategic priorities in the *Healthy Ireland in the Health Services: National Implementation Plan 2015-2017* (Health Service Executive, 2015a). In addition, there are plans for developing a universal healthcare system in Ireland which aims to tackle the challenge of chronic disease management - *Sláintecare* (Oireachtas Committee on the Future of Healthcare, 2017). The vision of *Sláintecare* is that patients are treated 'on the basis of health need rather than on ability to pay' (p.38), providing equitable access to healthcare. *Sláintecare* aims to incentivise people to access community and primary care, to facilitate integrated care and empower individuals to play a role in managing their health (Burke et al., 2018).

1.1.1 Burden of Asthma and COPD

Globally, the management of Asthma and COPD are recognised as key areas for improvement in healthcare (World Health Organisation, 2019). The Global Burden of Disease Study indicated that COPD and asthma account for 2.6% and 1.1% of disability adjusted life-years, respectively (Soriano et al., 2017). When this contribution is put into perspective, asthma and COPD are both among the top 20 conditions that contribute to most years lived with disability globally (Vos et al., 2016).

A recent report has sought to synthesise information on the burden of chronic respiratory disease in Ireland, acknowledging the limitations of current data sources and extrapolating from service usage statistics in the absence of disease registries for asthma and COPD in Ireland (O'Connor et al., 2018). It is estimated that 7-9% of adults in Ireland have a diagnosis of asthma (O'Connor et al., 2018) with the prevalence of 7% supported by recent data from *Healthy Ireland Surveys* (Department of Health, 2019b). It was not possible to get a reliable estimate of the prevalence of COPD (O'Connor et al., 2018). However global estimates indicate that the prevalence of asthma is approximately twice that of COPD (Soriano et al., 2017).

In terms of impact, Ireland had the 4th highest death rate from respiratory disease within Europe in 2016, when almost 1 in 5 deaths were due to these conditions (O'Connor et al., 2018). Respiratory conditions also accounted for 14% of hospitalisations in Ireland which was more than any other disease group. Age-sex standardised rates of hospitalisation were 389 per 100,000 and 46 per 100,000 for COPD and asthma, respectively (O'Connor et al.,

2018). Given, the burden of asthma and COPD in Ireland, these conditions have been prioritised as targets for chronic disease management in Ireland (Chronic Conditions Working Group, 2017).

1.1.2 Symptoms and features of asthma and COPD

Asthma and COPD are both chronic respiratory diseases characterised by airflow limitation and symptoms such as shortness of breath, wheeze, chest tightness and cough (Global Initiative for Asthma, GINA, 2019; Global Intiative for Chonic Obstructive Lung Disease, GOLD, 2019). Asthma symptoms can begin at any age and often begin in childhood. While asthma can produce symptoms on a daily basis, symptoms can often vary in intensity over time and develop into serious flare-ups, even for those who commonly have few symptoms (GINA, 2019). Flare-ups, also known as exacerbations, are periods where patients experience worse symptoms and lung function than usual (GINA, 2019). Flare-ups may be sub-acute or acute. Asthma is described in terms of severity and control. Asthma severity is determined by the level of pharmacological treatment required in order to control symptoms (GINA, 2019). Asthma control describes the extent to which symptoms are managed by treatment (GINA, 2019). The underlying physiology of asthma involves airway inflammation. However, it is a heterogeneous condition and some work has been done to characterise the phenotypes, particularly in the context of severe asthma (GINA, 2019).

COPD is a disease that manifests later in life and is most common in older adults. It is often developed where there is exposure to harmful particles or gases such as smoking or pollutants in conjunction with genetic and

environmental factors (GOLD, 2019). It is normally characterised by persistent symptoms, airflow limitation and periods of exacerbation. It is progressive in nature and exacerbations contribute to further disease progression (GOLD, 2019). Like asthma, the physiological changes underpinning symptoms are heterogeneous. The physiology of COPD also involves airway inflammation which gives rise to further structural changes (GOLD, 2019). Asthma and COPD can sometimes be difficult to distinguish, particularly in older adults and smokers (GINA, 2019). The term asthma-COPD overlap is used when people present with features of both diseases.

1.1.3 Recommended management of asthma and COPD

The day-to-day pharmacological management of asthma involves regular use of controller inhalers and use of reliever inhalers as necessary. The management of COPD also involves pharmacological treatments, often inhalers as well as vaccinations to minimise the contraction of viruses that can exacerbate COPD (GOLD, 2019). Exacerbations of asthma and COPD require adjustments to medical treatments that may be managed by the individual at home or may require the assistance of healthcare professionals and in some cases hospitalisation. Action plans are written documents recommended to support management of flare-ups through information on recognising symptoms, actions to alter medicines/access healthcare and step up/step-down approaches (Gatheral et al., 2017; GINA, 2019). Given the progressive nature of COPD, more advanced disease management involves long term oxygen therapy and non-invasive ventilation and further palliative care approaches (GOLD, 2019).

Like other chronic diseases, people with asthma and COPD require support to manage the condition in everyday life. This is recommended to involve education on the disease, side effects of medication, medication adherence, inhaler technique, management of comorbidities and action plans (GINA, 2019; GOLD, 2019). It also involves assistance with smoking cessation, exercise, the identification and management of triggers, behaviour change components and goal setting (GINA, 2019; GOLD, 2019).

1.2 Approaches to chronic disease management

Achieving recommended approaches to management and improved health outcomes for people with chronic diseases has proven challenging. A model that has gained international recognition for chronic disease management is the Chronic Care Model first developed in the United States (Wagner et al., 2001). The Chronic Care Model advocates multifaceted developments in healthcare provision including healthcare delivery redesign, education and decision support for healthcare providers, efficient clinical information systems as well as the provision of SMS for people with chronic disease. SMS for chronic disease was identified as a key component of the Chronic Care Model in order to reduce the burden of chronic disease (Bodenheimer et al., 2002). Bodenheimer et al. (2002) argued for the importance of supporting self-management, firstly, based on the rationale that people will always have a daily role to play in managing their illness. Secondly, the authors drew on early evidence that interventions which incorporated problem-solving skills and targeted patient confidence in skills had greater patient benefit compared to traditional patient education. A recent review found that interventions with SMS as the primary component of the intervention have had greatest improvements

in patient outcomes compared to other elements of the Chronic Care Model (Reynolds et al., 2018).

1.2.1 Historical and conceptual development of self-management and SMS

There have been different definitions of self-management used in the literature since its earliest application to chronic disease. Earlier definitions of self-management refer to the daily tasks that an individual must complete in order to "control or reduce the impact of disease on physical health status" (Clark et al., 1991, p.5). However, this focus on physical health presented a limited perspective on self-management given the evidence that chronic disease affects wellbeing and quality of life (Cappa et al., 2019; Singh et al., 2017; Wang et al., 2008). A later definition acknowledged the importance of psychosocial as well as physical functioning (Lorig & Holman, 2003). Self-management was seen to encompass medical, role and emotional management. More recent definitions also focused on these three areas of management (Miller et al., 2015; Van de Velde et al., 2019). Chronic disease self-management is now seen as a process through which individuals actively cope with their chronic disease in the context of their daily lives (Miller et al., 2015). Recent critical perspectives suggest that the aim of self-management should be living well in the context of managing a chronic disease rather than focusing on managing chronic conditions well (Entwistle, Cribb, & Owens, 2018; Morgan et al., 2017).

In contrast to the growing body of conceptual literature on self-management, there is little conceptual literature on support for self-management. There was a shift in the focus of literature from self-management to SMS which

acknowledged that self-management of chronic disease required healthcare system support and the need to examine the provision of support to enable people to self-manage (Kawi, 2012). An early definition of SMS was the "systematic provision of education and supportive interventions" with the purpose of enhancing patients skills and confidence in managing their health (Institute of Medicine, 2003, p.52). A later concept analysis defined SMS as a comprehensive approach with defining attributes at three levels: patient-centred level, provider level and organisational level (Kawi, 2012). At the patient-centred level, SMS involved patients as active partners in care and tailoring care, including the educational modalities, to the patient. At the provider level, SMS required providers to have appropriate knowledge, skills and attitudes. At the organisational level, SMS included a multidisciplinary team approach; an organised system of care such as provision of training to staff and clarification of roles; and linkages to community and lay-led supports. The antecedents were any factors which reinforced the need for self-management of chronic disease while the consequences of SMS were enhanced quality of care, improved patient self-management and greater patient and provider satisfaction (Kawi, 2012).

Much of the early research on support for self-management was on group-based interventions in the form of the Stanford Chronic Disease Self-management Programme (Lorig et al., 2001; Lorig et al., 1999). This programme aimed to equip people with skills to manage their physical and psychological wellbeing through education but also through enhancing self-efficacy – that is peoples' beliefs or confidence in their capabilities to engage in the required tasks (Bandura, 1998; Lorig & Holman, 2003). The programme

targeted changes in exercise and diet, treatment adherence, cognitive symptom management, emotional management and communication with healthcare professionals (Lorig et al., 2001; Lorig et al., 1999). This was typically a six-week programme with weekly sessions delivered to groups of 10-15 participants with different chronic diseases including people with stroke, heart disease, arthritis and respiratory disease. Groups were led by either healthcare professionals (HCPs) or lay-led by peers with chronic conditions.

Individual approaches to supporting self-management were also developed to enhance the collaboration between individuals and their HCPs to facilitate tailored support for self-management (Battersby et al., 2010). Evidence of this approach largely originated with the Flinders Programme of Chronic Condition Management. The programme involved education and information provision, shared decision-making, collaborative goal setting and problem solving, development of self-management plans and linkages to other resources and interventions depending on the patients' needs (Battersby et al., 2010). It also involved follow-up over 6 months with flexibility in modes of communication (Battersby et al., 2015).

Both the Stanford and Flinders programmes have shown positive effects for people with chronic diseases, but these have often pertained to specific outcome areas and have differed across studies. An early evaluation found the Stanford programme resulted in reduced health distress, reduced social/role activity limitations as well as decreased hospitalisations for people with chronic disease (Lorig et al., 1999). A review of 23 studies of the Stanford programme found improvements in areas of psychological health and cognitive symptom

management at 9-12-month follow-up (Brady et al., 2013). However, there were no significant differences in some measures of physical health including self-rated health or healthcare utilisation at 9-12 month follow-up (Brady et al., 2013). A randomised controlled trial of the Flinders programme found improvements in quality of life at 6-month follow-up though not at 12-month follow-up and there were no reported differences in physical health measures (Battersby et al., 2015). A pattern of these early interventions is that effects have been stronger for psychosocial outcomes and that some effects are reduced in the long term.

Over time, SMS interventions have been adapted such that interventions differ in terms of content and delivery. Reviews of the effectiveness of various SMS interventions for chronic diseases such as diabetes, respiratory and cardiovascular conditions have shown positive effects overall but some effects were small and some individual studies did not show significant outcomes (Chrvala et al., 2016; Panagioti et al., 2014a; Zwerink et al., 2014). The heterogeneous nature of interventions has made it difficult to isolate components contributing to effectiveness and to develop recommendations about optimal SMS interventions (Trappenburg et al., 2013; Zwerink et al., 2014).

1.2.2 Optimal approaches to providing SMS for asthma and COPD

SMS for chronic respiratory conditions has had positive outcomes. A Cochrane review in 2014 indicated that SMS interventions for COPD reduced dyspnoea, improved health-related quality of life (HRQoL) and reduced hospitalisation (Zwerink et al., 2014). However, no significant effects were found on other

outcomes such as on exercise capacity or mortality. A subsequent review of SMS interventions for COPD including an action plan also reported a positive impact of SMS on HRQoL and on respiratory-related hospital admission (Lenferink et al., 2017). However, the review found no significant effect of SMS compared to usual care on dyspnoea, ED visits, general practitioner (GP) visits or number of exacerbations. For asthma, a systematic meta-review has indicated that SMS has reduced hospitalisations, ED attendances, unscheduled consultations and improved asthma control and quality of life indicators (Pinnock et al., 2017). However, narrative reviews indicated mixed findings across studies and a meta-analysis conducted on recent studies not included in the meta-review found that some effects were small including hospitalisation rates and ED visits (Pinnock et al., 2017).

Possible explanations for the mixed findings of SMS interventions exist. Different features of SMS interventions are likely to yield mixed results. A systematic review of SMS strategies in primary care found that SMS includes varying components such as action plans, symptom monitoring, psychological coping, problem solving and focus on physical activity, diet, and smoking cessation (Dineen-Griffin et al., 2019). They also differ in their delivery such as whether they are delivered face-to-face, over phone or other technologies; whether the programme is generic or disease-specific; the expertise of the programme facilitator including HCPs with varying levels of disease-specific knowledge or peers with chronic disease (Dineen-Griffin et al., 2019; Jonkman et al., 2016).

Through reviewing and comparing quantitative outcomes of studies, Dineen-Griffin et al. (2019) identified features of SMS likely to yield positive outcomes such as face-to-face, one-to-one multicomponent tailored interventions with ongoing follow-up. A review found that interventions which included education on COPD, an action plan and exercise components were linked with more positive quality of life outcomes for people with COPD while the duration of the programme (comparing programmes less or more than five weeks) was not influential (Cannon et al., 2016). However, this review explored only a subgroup of potentially relevant intervention components and does not consider other components such as emotional coping skills (Cannon et al., 2016). Within the meta-review of SMS for asthma, factors such as intensity of education or review, the active involvement of the participants and the inclusion of self-management education with an action plan were associated with effective interventions (Pinnock et al., 2017). However, many of the reviews contributing to these findings were conducted in 2002-2003. Further research is needed to understand the components of SMS that enhance its effectiveness.

There is also evidence that efficacy of support may differ across individuals and that support should be tailored to the individual and associated context. Studies have indicated that particular groups may be more likely to benefit from SMS interventions such as those with lower baseline measures (Chrvala et al., 2016). For example, people with poor glycaemic control in diabetes showed greater improvements in outcomes compared to those with good control (Elissen et al., 2012). In addition, people may have different preferences for support which may in turn influence the extent to which they benefit from an

intervention. For example, in a study of preferences for support programmes for epilepsy, there was variation in the extent to which support provided by a peer compared to a physician with some participants more accepting of this than others (Atkinson-Clark et al., 2018). Understanding how support can be tailored to an individual's needs and preferences is an important consideration (Trappenburg et al., 2013; Van de Velde et al., 2019).

An additional consideration for advancing knowledge of optimal interventions is to achieve greater clarity on the outcome measures of intervention studies. Different outcomes have been selected to measure the effect of SMS interventions and there have been calls for consensus on the most appropriate outcomes (Boger et al., 2015; Dineen-Griffin et al., 2019). Some studies may be underpowered and are thereby unable to detect changes in outcomes such as number of flare-ups which are of a low prevalence in the target population from a statistical perspective (Vanfleteren & Fabbri, 2019). Furthermore, there have been concerns about the focus on outcomes such as reduced healthcare utilisation (Gately et al., 2007) and there has been debate over the usefulness of early indicators of cost effectiveness (Nolte & Knai, 2015). SMS may in fact result in increased healthcare utilisation for some participants which may be beneficial and/or appropriate and may have long term benefits (Gately et al., 2007; Nolte & Knai, 2015; Van de Velde et al., 2019). Greater timely healthcare utilisation in primary or community care may prevent deterioration and further costs associated with hospitalisation. Thus it is suggested that further progress can be made in terms of identifying the key outcomes of SMS interventions for measurement. This may be facilitated by taking stakeholders' views into

account. Up until recently, stakeholders' views on SMS outcomes have been infrequently elicited (Boger et al., 2015).

1.2.3 The role of the wider health system in SMS

The wider health system in which SMS is provided can also influence the outcomes of SMS initiatives. As previously described, a concept analysis illustrated that SMS is a comprehensive approach in which patient-centred support is facilitated by providers with appropriate knowledge, skills and attitudes who are in turn supported by organisational factors such as referral pathways, clinical information systems and a multidisciplinary approach to care (Kawi, 2012). Intervention literature suggests that multilevel interventions contribute to better clinical outcomes. For example, a systematic review of SMS for asthma found that interventions had greater impact on clinical outcomes where they have targeted patient, provider and organisational levels than interventions focused on one level alone (Pinnock et al., 2015).

Research has highlighted barriers to implementing SMS at different levels of the system. Interventions which sought to change HCP behaviour such as training GPs and practice nurses in shared decision-making and goal planning were found not to be implemented as recommended (Kennedy, Rogers, Bowen, et al., 2014; Robertson et al., 2013). Similarly, the Flinders Programme – a programme using individualised approaches to SMS through primary care settings – was not implemented as intended for at least 24% of participants (Battersby et al., 2015). It was reported that clinicians felt a lack of confidence in elements such as goal-setting and motivation (Battersby et al., 2015). There were also weak linkages between the programme providers and their routine

healthcare services. Despite the health policy emphasis on patient participation in healthcare decision-making, a study of preferences for involvement in treatment-decision making in asthma found that in practice 55% of participants were less involved than they preferred (Caress et al., 2005). Health system commitment and HCP training are identified as facilitators of SMS interventions for asthma (Pinnock et al., 2015).

However, patients' beliefs and preferences may also affect implementation. The study of preferences for treatment decision-making by Caress et al. (2005) also found that 40% of participants preferred a more passive role which may pose challenges for implementing shared decision-making. There is also evidence of challenges in participation and completion of SMS interventions including pulmonary rehabilitation for COPD (Hudon et al., 2016; Jones et al., 2014). Reasons for lack of completion included poor health, perceived heterogeneity across participants, and perceptions of the programme (Hudon et al., 2016; Sohanpal et al., 2015). This highlights the need for patient-centred SMS where education and the approaches to delivering care are tailored to the patient (Kawi, 2012).

The above findings highlight the influence and interaction of patient, provider and organisation factors in SMS and thus, the importance of supporting self-management at multiple levels of the system. There has been a movement toward systems thinking in relation to SMS and toward understanding how SMS can be embedded within a healthcare system (Mills et al., 2017). Policies are a key component of advancing systems of SMS and understanding policy implementation may help to advance the implementation of SMS.

1.2.4 Systems and policy development for chronic disease management

Internationally, policies and approaches have been developed in the broader area of chronic disease management which include elements of SMS (Nolte & Hinrichs, 2012; Nolte & Knai, 2015; Nolte et al., 2008). The work of Nolte and colleagues has provided important information on the progress of chronic disease management within health systems in European countries (Nolte & Hinrichs, 2012; Nolte & Knai, 2015). Key informants from 12 European countries completed a template to gather relevant information and were expected to draw on a range of data sources within their country. The report highlighted a variety of approaches taken by European countries which have resulted in positive outcomes for people with chronic disease (Nolte & Knai, 2015). The reports also reflect the heterogeneity and complexity of the evidence base used to develop effective interventions, highlighting variable results of interventions for many chronic diseases including asthma and COPD (Nolte & Knai, 2015). There was large variation in the levels of SMS activities across European countries from provision of written materials through to face-to-face coaching and follow-up approaches and it was unclear to what extent they had been implemented (Elissen et al., 2013; Nolte & Knai, 2015).

The report identified the importance of wider system changes and attention to the political context in order to realise the vision of improved chronic disease management (Nolte & Knai, 2015). The authors identified how health service contexts such as the equitable access to healthcare and processes of primary care enrolment may affect the ability to implement approaches to chronic disease management. The authors also identified the influence of centralisation and decentralisation of a system, that is the extent to which

governance operates at national or regional levels. An example of this is the Italian healthcare system where decentralisation contributed to uncoordinated implementation of policies and large variation across regions (Nolte & Knai, 2015). In some countries, policy was implemented at a national level whereas elsewhere, approaches were implemented at regional levels and planned for subsequent national roll-out (Nolte & Knai, 2015). Thus, there was considerable variation in the processes of implementation across countries and no clear indication of how to balance centralisation and decentralisation. It was highlighted that insights from implementation science are needed to facilitate the implementation of chronic disease management system reforms.

Given the evidence on the benefits of SMS and its facilitation by a system-wide approach, governments have been mobilised to develop policy to specifically address SMS for people with chronic disease. Frameworks have been developed over a decade ago in some countries (Queensland Government, 2008) while an SMS framework 'Living Well with a Chronic Condition' was more recently launched in Ireland (Chronic Conditions Working Group, 2017). The Irish framework outlines changes necessary to implement SMS at system, organisational and provider level as well as the level of the person with chronic disease (Figure 1.1). The framework also seeks to reduce previously noted geographical disparities in access to SMS across Ireland (Health Service Executive, 2016) and to improve the standard and consistency of SMS services (Mullaney et al., 2016). Thus, the frameworks provide a promising approach to developing system wide SMS.

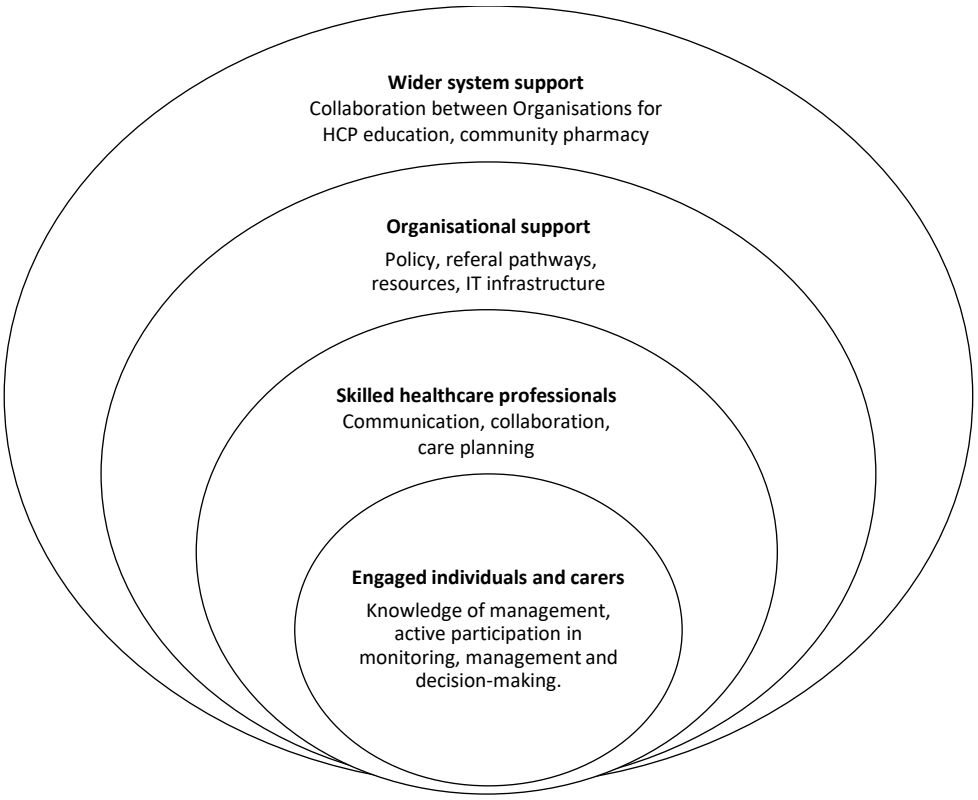


Figure 1.1 Whole systems model of SMS (Adapted from Chronic Conditions Working Group, 2017)

However, some previous healthcare policies, including those related to chronic disease management, have not achieved their aims and implementation targets in the Irish health system (May et al., 2014; McHugh et al., 2014). While research helps understand the best types of interventions and treatments for diseases, research on the policy process is needed to understand how innovations can be translated into real world practice. The following section provides a description of the policy process in order to shed light on factors that may influence SMS framework implementation.

1.3 Factors influencing the policy process

The policy process is often described as complex and there have been numerous theoretical approaches to describing and analysing policy processes (Sabatier, 2007). Stages of the policy process have been outlined including: agenda setting, policy formulation, implementation and evaluation; though this stages model is criticised for assuming linearity in stages and neat divisions which are not reflective of practice (Sabatier, 2007). The Health Policy Triangle (Walt & Gilson, 1994) is a broadly applicable framework to understand factors influencing policy (Buse et al., 2012). The framework includes the policy context, content, actors and processes (Figure 1.2). *Context* relates to the systemic factors which can affect policy such as political, economic, social as well as national and international factors. The *content* relates to the substance and constituent parts of the policy (Buse et al., 2012). The *process* concerns the ways in which policies are initiated, developed, negotiated, implemented and evaluated while the *actors* are the stakeholders which can affect policy, including individuals, organisations and government (Buse et al., 2012).

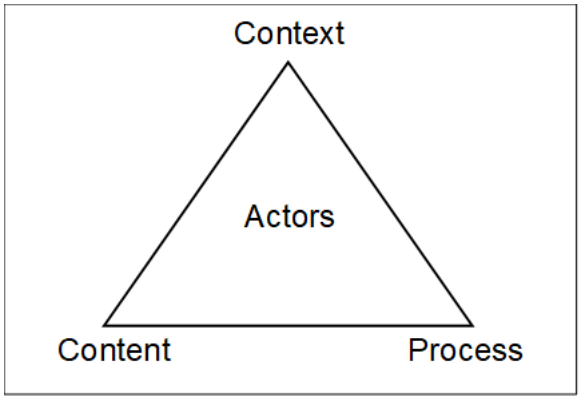


Figure 1.2 Health Policy Triangle

Theories have been developed to describe more specific elements of the policy process, particularly the interaction of policy actors (Sabatier, 2007). For example, the Advocacy Coalition Framework (Sabatier, 1988) helps to explain policy conflict and addresses a range of factors at various levels of the system that influence the behaviour of policy participants; while other perspectives focus on power dynamics and networks which influence the policy trajectory (Sabatier, 2007). The Multiple Streams Framework (Kingdon, 2014), originally outlined in 1984, has also been widely used in policy analysis particularly for understanding the process of agenda setting (Walt et al., 2008). The Multiple Streams Framework proposes that policies make it on the agenda when three elements align to open a policy window: the problem stream – the level of interest in the societal problem, the politics stream – the political environment and dominant ideology, and the policy stream – acceptability of the policy solution (Kingdon, 2014).

1.3.1 The process of policy implementation

The complexity of factors influencing policy implementation means that it has been challenging to identify a general theory of policy implementation (Nilsen et al., 2013). Policy is implemented in a real-world context where the impact of various contextual factors cannot be controlled or partitioned. Researchers have identified factors believed to increase the success of implementation such as the use of research evidence (Anderson et al., 2005); involvement of stakeholders (Pizzo et al., 2015); and inclusion of a comprehensive and strategic implementation plan which considers issues of governance and infrastructure (Walt, 1998). However, studies have also indicated that policy implementation is not a prescriptive process which is implemented as planned

but instead is a process of negotiation and exchange between policy actors responsible for implementing the policy (Buse et al., 2012).

Recently many of the ideas mentioned above have been drawn together in a multiple streams/critical juncture approach to policy implementation (Howlett, 2018; Howlett et al., 2015, 2017). This approach begins with the three streams proposed by Kingdon (2014) – problem, policy and political context – which influence agenda setting (See Figure 1.3). These three streams continue to exert influence on the subsequent policy stages including formulation, implementation and evaluation. The term process stream is used to describe the movement between various policy stages and how this shapes implementation. The programme stream captures the influences at the implementation stage where new actors who deliver, consume or administrate the policy can shape its trajectory and may raise conflicts where policy clashes with local interests or traditions.

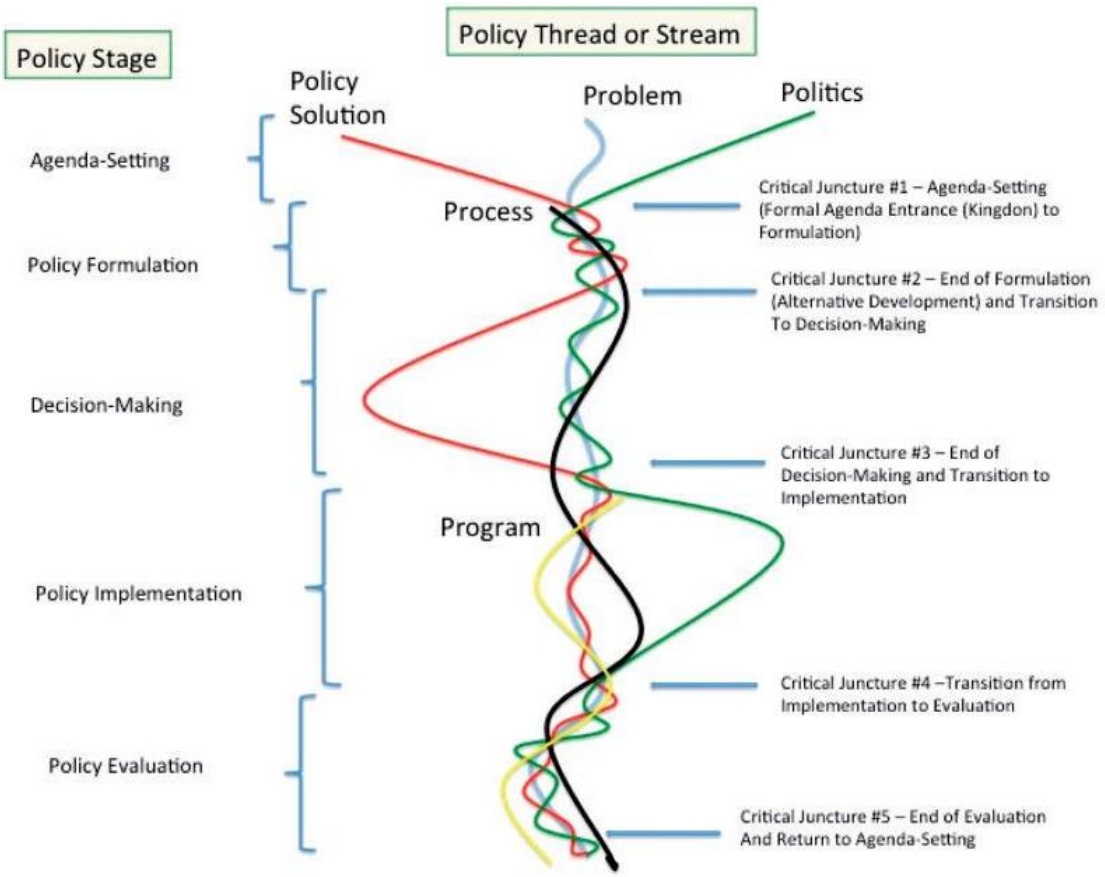


Figure 1.3 Five streams model of policy process including implementation (Howlett, 2018). Reproduced with permission.

1.3.2 Incorporating stakeholder perspectives in health policy

In the multiple streams approach to policy implementation, the new actors in the health policy implementation stage are typically HCPs, patients and managers involved in implementing the policy. However, there is now a large emphasis on involving stakeholders and those who will be actively involved in implementation from the early stages of policy development, often described as Public and Patient Involvement (PPI; Pizzo et al., 2015). Increased incorporation of the patient perspective has been noted as important for chronic disease management policy (Nolte & Knai, 2015). Different rationales

are provided for ensuring that the perspective of the patient or the person living with a disease is represented in both research and policy.

One motivation for PPI is a rights-based or emancipatory perspective which suggests that patients have a right to have a voice in the care provided to them and for there to be greater transparency in research and policy (Greenhalgh et al., 2019; Matthews et al., 2019). An additional motivation is that involving patients in research can help facilitate clearer understanding of their perspectives which can lead to the development of services which are better equipped to meet patient needs and facilitate greater service engagement (Greenhalgh et al., 2019; Matthews et al., 2019).

While there is agreement on the potential benefits of PPI, there is debate with regard to how PPI can best be achieved (Forsythe et al., 2019; Greenhalgh et al., 2019; Harris et al., 2019). There are various extents to which patients can be involved in both research and policy development. Higher levels of involvement may be described as collaboration where patients define the agenda, where information flows between researchers and patients, and decisions are shared (Forsythe et al., 2019; Harris et al., 2019). Involvement may also take the form of regular consultation processes where there is bidirectional information exchange, and people with lived experience influence designated decisions. Finally, involvement may also take the form of targeted input into research in a unidirectional manner but where people with lived experience do not actually participate in research decisions (Forsythe et al., 2019; Harris et al., 2019).

While this area of literature is rapidly developing, there is still much to be known about the most effective ways to involve patients in policy processes and ensure that their perspectives are represented. Approaches will likely need to be tailored to the target population and the aim of the project (Greenhalgh et al., 2019). There are also challenges and trade-offs that must be considered in conducting PPI particularly in the contexts of time limitations and study constraints (Forsythe et al., 2019). There is continuous caution against tokenistic involvement which could negatively influence those participating (Greenhalgh et al., 2019; Pizzo et al., 2015). Acknowledging the continuum of involvement, empirical studies of patients' preferences which are developed through research methods seeking to closely reflect patients' perspectives, may provide useful information on the patient perspective to inform healthcare decision-making (Muhlbacher & Johnson, 2016).

1.3.3 Measuring preferences through a discrete choice experiment

The discrete choice experiment (DCE) is a quantitative tool that can be used to measure the stated preferences of service users (Bridges et al., 2011). The DCE is underpinned by Lancaster's theory of consumer choice and assumes that preferences are formed based on attributes of goods or services and that this information can capture demand for services (Lancaster, 1966). DCEs ask participants to choose between alternative goods/services which differ on a number of attributes. When these data are modelled, they provide information on the relative importance of attributes of services according to the decision maker and trade-offs that participants make between service features which can ultimately assist in policy decision-making where many potential approaches are available (Mandeville et al., 2014; Mühlbacher et al., 2016).

Of the methods which seek to measure stated preferences, a DCE is more appropriate to understand the value of individual attributes rather than other methods such as contingent valuation where the emphasis is on valuing a fixed set of changes together (Johnston et al., 2017).

A DCE is a measure of stated preferences. An alternative approach to understanding consumer preferences is to examine preferences through the study of data on consumer's actual behaviour within a market. The advantages of stated preference methods over real-world measures of consumption are that they can measure the preference for hypothetical attributes which do not yet exist in services. They can measure preferences prior to intervention delivery which can be used to inform intervention or policy development (Ostermann et al., 2017) which is helpful in terms of guiding implementation of new services features (Mandeville et al., 2014; Muhlbacher & Johnson, 2016; Salloum et al., 2017). Furthermore, DCEs enable researchers to go beyond the study of end-point health/clinical outcomes and to examine the relative importance of process outcomes from the service user perspectives (Lancsar et al., 2017).

Stated preference methods have been criticised for a hypothetical bias. Some critiques suggest that choices made in DCE may not be representative of actual consumer choices (Rakotonarivo et al., 2016). Differences between DCE studies (stated preferences) and other measures of consumer behaviour ('real' preferences) have been used to support this argument (Rakotonarivo et al., 2016). However, it has been questioned to what extent controlled and laboratory based assessments of 'real' preferences are related to consumers'

actual behaviours (Rakotonarivo et al., 2016). In the area of healthcare, a review found that a limited number of studies have examined the link between DCE findings and observed real life choices, with only six suitable for inclusion in a meta-analysis (Quaife et al., 2018). DCEs were found to predict real word health choices with 88% sensitivity and 34% specificity (Quaife et al., 2018). The authors conclude that DCEs predict behaviour reasonably well and that DCE studies are useful to decision-makers when considering their advantages over other research methods such as measuring preferences for hypothetical services and process attributes prior to resource-intensive intervention.

Insights into the important features of SMS have been gained from studies which have qualitatively explored patients' perspectives and which are not possible to elucidate through quantitative studies of health outcomes (Baker & Fatoye, 2019). The DCE is a method of providing quantitative information on patients' preferences and it is recommended that qualitative research forms an integral component of its development (Bridges et al., 2011; Coast et al., 2012; Vass et al., 2017). The resulting quantitative information on the features of importance from patients' perspectives can be viewed as complimentary to qualitative research and may be more easily integrated with clinical data and policy options (Johnson & Zhou, 2016). DCEs have begun to be employed in the area of support for chronic conditions (Atkinson-Clark et al., 2018; Hertroijs et al., 2018) and may be useful in identifying the relative importance of features of SMS for asthma and COPD which require further elucidation (Baker & Fatoye, 2019; Dineen-Griffin et al., 2019).

1.4 Thesis layout

This thesis comprises eight chapters in total. Five of these chapters were prepared for publication. Of these: two are published; one paper is under review, and two papers are being prepared for submission. The trajectory of research development is presented in Figure 1.4. The layout of the remaining chapters is as follows:

Chapter 2 presents a document analysis of frameworks on SMS for chronic disease across health systems in various countries using the Health Policy Triangle (Walt & Gilson, 1994). The systematic approach to conducting this analysis is detailed. The chapter is published (O'Connell et al., 2018; Appendix 1.A).

Chapter 3 presents a qualitative study that explored the process of implementing national/state level SMS frameworks from the perspectives of key informants who had a role in the implementation of these frameworks in their respective health systems. The methods used to conduct the study including telephone interviews and thematic analysis are detailed. The findings of the study are presented. The implications for further research are discussed including the need to understand the priorities of people with chronic conditions regarding SMS. This chapter has been submitted for publication.

Following on from Chapter 3, this thesis presents the development and piloting of a DCE to measure the support preferences of people with chronic respiratory diseases in Ireland. The early stages of this research focused on both COPD and asthma. However, as the research evolved over time, the DCE

focused specifically on people with asthma. Chapters 4-7 present a number of steps recommended for DCE development (Bridges et al., 2011).

Chapter 4 presents a review and meta-synthesis of qualitative research on the SMS preferences of people with asthma or COPD. Thematic synthesis was used to synthesise qualitative studies as guided by Thomas and Harden (2008). The thematic findings are presented. This chapter is now published (O'Connell et al., 2019; Appendix 1.B).

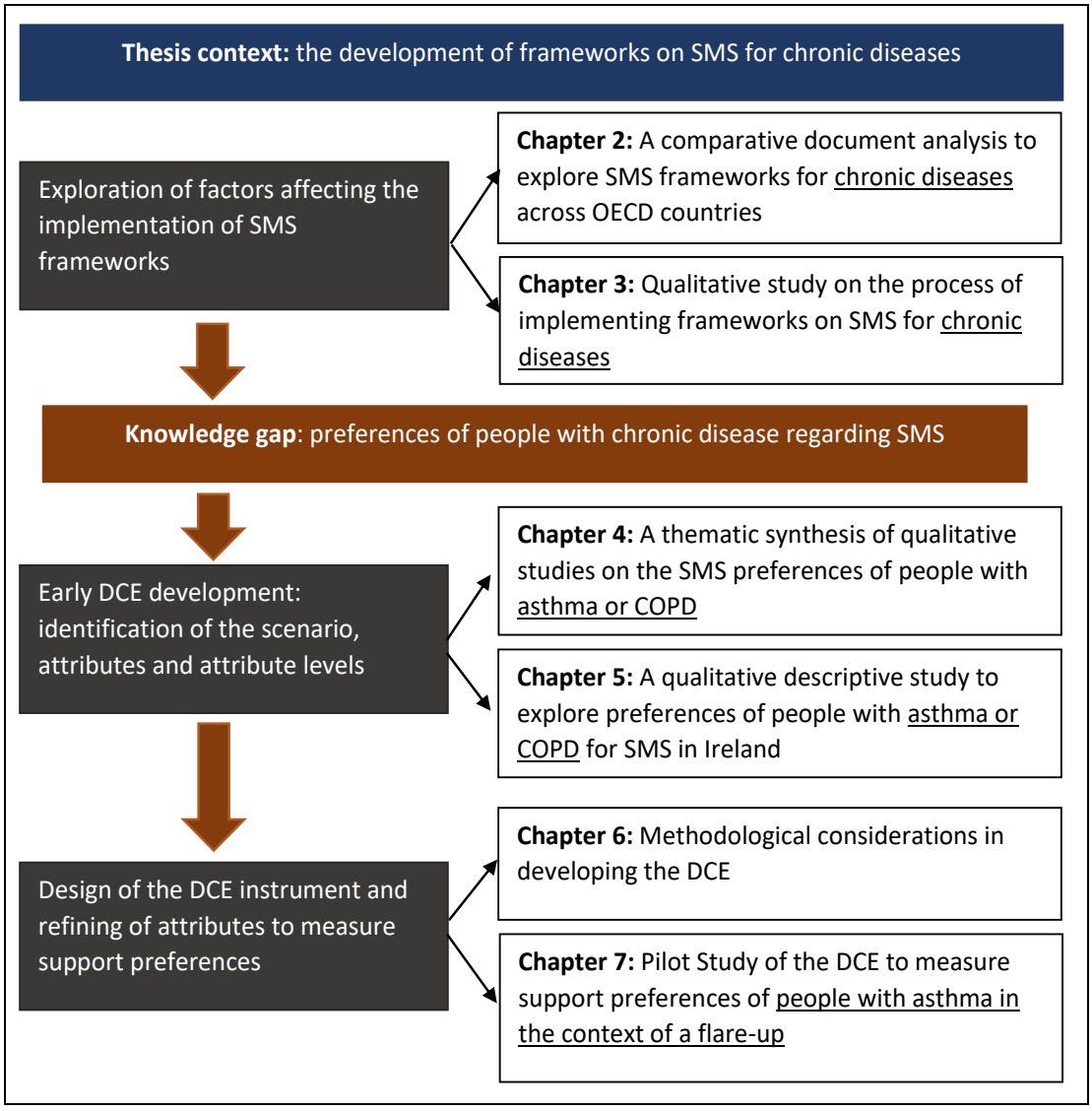
Chapter 5 is a qualitative descriptive study of the SMS preferences of people with asthma or COPD. The methods used to collect data involving individual and focus group interviews are described. The content analysis method used to analyse the data is described. Themes identified from the data analysis are presented.

Chapter 6 describes the steps in the development of the DCE, methodological considerations and the narrowing of the DCE to focus on preferences of people with asthma in the context of experiencing a flare-up.

Chapter 7 reports the pilot study of the DCE. This chapter provides an overview of the refining of the DCE instrument through cognitive interviews. Analysis of the data using a conditional logit model is described to provide evidence of the direction and strength of preferences for each attribute which can be used to further refine the DCE for future use.

Chapter 8 provides a discussion and synthesis of the findings across the thesis chapters; the strengths and limitations; and draws out implications for policy, practice and future research.

Figure 1.4. Trajectory of research development



Chapter 2. Frameworks for Self-Management Support for Chronic Disease: A Cross-Country Comparative Document Analysis

This chapter describes a document analysis of national and state-level frameworks on self-management support (SMS) for chronic disease. While chronic disease management initiatives have been studied at national and systems levels (Nolte & Knai, 2015), an analysis of policies/frameworks has not yet been carried out specific to the area of SMS. The rationale for examining framework documents is elaborated upon and the systematic search for documents from countries within the Organisation for Economic Co-operation and Development (OECD) is described. A document analysis using the Health Policy Triangle framework (Walt & Gilson, 1994) is detailed. The content of this chapter including the abstract presented at the outset is published in *BMC Health Services Research* (O'Connell et al., 2018).

Abstract

Background: In a number of countries, frameworks have been developed to improve SMS in order to reduce the impact of chronic disease. The frameworks potentially provide direction for system-wide change in the provision of SMS by healthcare systems. Although policy formulation sets a foundation for health service reform, little is currently known about the processes which underpin SMS framework development as well as the respective implementation and evaluation plans.

Methods: The aim of this study was to conduct a cross-country comparative document analysis of frameworks on SMS for chronic diseases in member countries of the Organisation for Economic Cooperation and Development. SMS frameworks were sourced through a systematic grey literature search and compared through document analysis using the Health Policy Triangle framework focusing on policy context, contents, processes of development, as well as the actors involved.

Results: Eight framework documents published from 2008 to 2017 were included for analysis from: Scotland, Wales, Ireland, Manitoba, Queensland, Western Australia, Tasmania and the Northern Territory. The number of chronic diseases identified for SMS varied across the frameworks. A notable gap was a lack of focus on multimorbidity. Common courses of action across countries included the provision of self-management programmes for individuals with chronic disease and education to health professionals, though different approaches were proposed. The 'actors' involved in policy formulation were inconsistent across countries and it was only clear from two frameworks that individuals with chronic disease were directly involved. Half of the frameworks had SMS implementation plans with timelines. Although all frameworks referred to the need for evaluation of SMS implementation, few provided a detailed plan.

Conclusions: Differences across frameworks may have implications for their success including: the extent to which people with chronic disease are involved in policy making; the courses of action taken to enhance SMS; and planned implementation processes including governance and

infrastructure. Further research is needed to examine how differences in frameworks have affected implementation and to identify the critical success factors in SMS policy implementation.

2.1 Background

Self-management is a process through which individuals actively cope with their chronic disease in the context of their daily lives (Miller et al., 2015). Supporting individuals to self-manage is an important strategy to reduce the burden of chronic disease (Bodenheimer et al., 2002; Wagner et al., 2001). SMS has had positive effects on health outcomes for people with chronic disease including increased health related quality of life (Chrvala et al., 2016; Panagioti et al., 2014b; Zwerink et al., 2014). In some countries, governments have developed health policy to support self-management of chronic disease in order to promote positive health outcomes.

SMS policy and frameworks, such as those developed in Australia (Queensland Government, 2008) and Ireland (Chronic Conditions Working Group, 2017), aim to guide system-wide changes in service delivery for chronic disease management. However, healthcare policies have not always achieved their aims and implementation targets (May et al., 2014; McHugh et al., 2014). Health policy is a complex phenomenon (Walt et al., 2008). Although there is little guidance on how to conduct analyses of health policies, the use of a framework has been recommended (Walt et al., 2008). A commonly used framework is the Health Policy Triangle which comprises of four components: context, content, actors, and processes (Buse et al., 2012; Gilson & Raphaely, 2008; Walt & Gilson, 1994). The *content* of a policy refers to the substance of

the policy while the *context* relates to the systemic factors which can affect policy such as political, economic, social, national and international influences (Buse et al., 2012). The *actors* are the stakeholders who influence policy, including individuals, organisations and government while the *process* concerns the ways in which policies are initiated, developed, negotiated, implemented and evaluated (Buse et al., 2012).

There is potential for considerable heterogeneity across chronic disease SMS policies from different countries. For example, there are contextual factors that differ across health systems including funding; organisation and governance; and the service reforms for population health priorities such as chronic disease management (Mossialos et al., 2017; Nolte & Knai, 2015; Nolte et al., 2008). The content of chronic disease management policy, such as priorities on courses of action, can vary with some countries emphasising the role of nursing in chronic disease SMS and others emphasising accessibility to specialist multidisciplinary teams in primary care (Nolte & Knai, 2015). Many approaches to providing SMS have been studied such as peer support and online programmes (Foster et al., 2007; Lorig et al., 2016; Zwerink et al., 2014). The evidence is not clear on which SMS approaches provide the greatest benefit to individuals with chronic disease (Jonkman et al., 2016; Zwerink et al., 2014) which could potentially result in countries adopting different courses of action to support self-management.

In terms of actors, the engagement of a diverse range of stakeholders in health policy making is expected (World Health Organisation, 2010). Public and patient involvement (PPI) is called for in the development of health strategies

(Pizzo et al., 2015). The potential benefits of PPI include services adapted to local needs, the possibility of identifying innovative approaches to the problem as well as giving a voice to marginalised groups (Pizzo et al., 2015). Specific to SMS, while research has examined patients' experiences of self-managing their chronic disease, few studies have directly sought patients views regarding desired outcomes (Boger et al., 2015). To date, little is known about patient involvement in SMS policy development.

The process of implementing SMS may be challenging. For example, there may be varying levels of readiness to endorse SMS as an approach to chronic disease management at the levels of the individual with chronic disease, the healthcare provider and the organisation (Gagnon et al., 2011). A clear and strategic implementation plan is recommended for health policies and strategies (Walt, 1998; World Health Organisation, 2010). The World Health Organisation (WHO) outlined the importance of including governance arrangements, consideration for current capacity and resource planning, and mechanisms for evaluation in national policy and strategy documents (World Health Organisation, 2010). Therefore, the level of detail in SMS frameworks implementation plans may affect the extent to which they achieve their intended outcomes.

Over the past decade, SMS frameworks for chronic disease have been developed as a policy initiative in various countries (Chronic Conditions Working Group, 2017; Department of Health Western Australia, 2011; Queensland Government, 2008). To the best of my knowledge, SMS frameworks have not been previously compared across countries. Therefore,

the aim of this study was to conduct a cross-country comparative document analysis of frameworks on SMS for chronic diseases. It was aimed to compare countries in terms of policy content, the contexts influencing policy development, stakeholder involvement, and processes of policy development, as well as implementation and evaluation plans. Countries in the early stages of policy development and implementation can draw on this analysis.

2.2 Methods

This study was designed as a comparative document analysis and adopted principles and procedures of systematic review methods. Document analysis, as a qualitative method, is "a systematic procedure for reviewing and evaluating documents" (Bowen, 2009, p. 27). This procedure organises document information into categories which relate to specified research questions/categories (Bowen, 2009). Data collection and analysis were guided by the Health Policy Triangle (Walt & Gilson, 1994). SMS frameworks can be considered health policy documents in line with the definition of health policy provided by Buse and colleagues, that is, 'courses of action (and inaction) that affect the set of institutions, organisations, services and funding arrangements of the health system' (Buse et al., 2012, p. 6). As presented in Table 2.1, categories of information for analysis were constructed around the four areas of the Health Policy Triangle: context, content, processes and actors (Walt & Gilson, 1994).

Table 2.1.Categories of Information for analysis

Category	Descriptor	Examples/Application
Context	Factors which can influence policy development including past provision of SMS for chronic disease	Healthcare structures and governance, health service reform agenda, burden of chronic disease, demographic and prevalence trends of chronic disease, e-health trends, information on previous delivery of SMS
Content	Scope of the framework, defining features of SMS espoused, goal statements, policy statements of action	Chronic diseases targeted and scope of population for intervention; components of SMS definition; stated goal including intended effect and outcomes; SMS priorities or courses of action
Actors	Stakeholders involved in policy making	Government representatives, clinicians, health service managers, people with chronic disease, voluntary sector representatives and community and advocacy groups.
Processes	How information and actors' interests are incorporated in policy formation; proposed process of implementation and evaluation	Evidence used and processes of stakeholder involvement such as consultation, involvement in workshops and surveys, member of working group; implementation plans, associated timelines, facilitation processes; evaluation strategies and feedback process.

2.2.1 Inclusion criteria

Eligibility criteria included documents that: exclusively targeted SMS for chronic diseases in adult populations; targeted more than one chronic disease/condition; originated in countries which are members of the OECD; were produced by/affiliated with a national government or national healthcare service (for countries such as USA, Australia, Canada and the United Kingdom, documents at province/state/territory level were searched); offered recommendations for the provision of SMS within the respective healthcare system; were published in English and in the period from October 31st 2007 to November 30th 2017. Only the most recent SMS framework document from countries was included.

2.2.2 Search process

A search of grey literature sources was conducted to retrieve relevant documents. While no gold standard exists for grey literature searching, this

study drew on the strategy used by Godin and colleagues (2015) with the aim of ensuring that the search methods used were explicit, reproducible and identified all relevant documents. The search involved the following three groups of terms: (1) self-manage, self-management, self-managing; (2) long-term condition, chronic condition, chronic illness, chronic disease, non-communicable disease; (3) policy, framework, guideline, model, strategy and standard.

The search was carried out in early December 2017 and consisted of four parts. Firstly, it included grey literature databases which contained policy and government publications: OAIster, BIREME Virtual Health Library, World Health Organisation (WHO) Information Repository for Information Sharing, OECD iLibrary, Open Grey, Grey Literature Report, Canadian Electronic Library and Analysis and Policy Observatory. As the search specifications differed between sites, different search terms were used. See Appendix 2.A for the specific search combinations used. Secondly, an advanced google search was completed using the search term combinations (Appendix 2.A). The first 10 pages of each search including different combinations of search terms were screened akin to previous work (Godin et al., 2015).

A third search was carried out on Ministry of Health/ Department of Health websites within OECD countries. Sites were searched using their relevant search box and manually where this did not exist. The search terms used were ‘self-manage’, ‘self-managing’ and ‘self-management’. Finally, documents were searched for by screening the reference lists in the relevant retrieved documents.

2.2.3 Study selection

Initial screening of the title and abstract/executive summary/overview was carried out by SOC. The titles and respective URLs of potentially relevant documents were entered into Excel and retained for full-text screening. Full texts of documents were assessed for inclusion by two authors independently (SOC, ES). Disagreements about eligibility were resolved by consensus and where necessary by a third reviewer (VMcC). The reason for exclusion was recorded.

2.2.4 Data extraction and analysis

Relevant information from documents was charted in a data extraction table which was initially piloted with two documents. The information was extracted by one author (SOC). This was then independently cross-checked by ES and VMcC. The data extracted included geographical location, author and year of publication as well as information categories guided by the Health Policy Triangle Framework (Walt & Gilson, 1994). This framework also guided the analysis with findings presented according to the components of the Health Policy Triangle: context, content, actors, and process.

2.3 Results

Eight documents on SMS met the inclusion criteria (see flowchart in Figure 2.1). The documents originated in Scotland (SL; Long-term Conditions Alliance Scotland and Scottish Government, 2008), Wales (WL; Welsh Assembly Government, 2009), Ireland (IRL; Chronic Conditions Working Group, 2017), Manitoba, Canada (MB ; Manitoba Health, 2012) and four Australian states/territories: Queensland (QLD; Queensland Government, 2008),

Western Australia (WA; Department of Health Western Australia, 2011), Tasmania (TAS; Department of Health and Human Services Tasmania, 2012) and the Northern Territory (NT; Northern Territory Department of Health, 2012). The frameworks were developed and published between 2008 and 2017. The full data extraction table is available in Appendix 2.B.

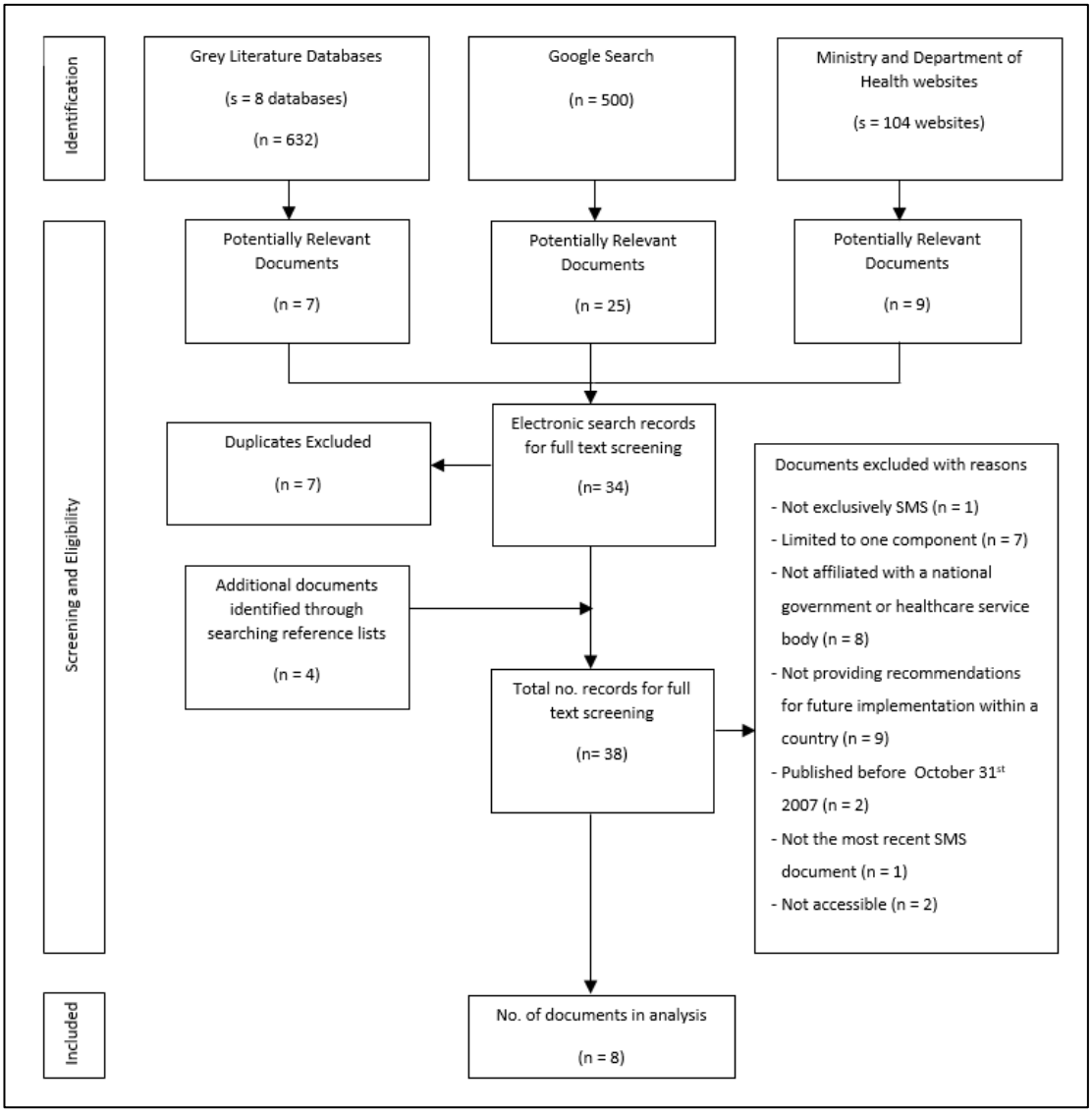


Figure 2.1. Document identification, screening and selection

2.3.1 Context

All eight frameworks highlighted the burden of chronic disease as context, referring to the impact of chronic disease in terms of: increased mortality; economic cost; use of health service resources, overburdened acute healthcare sectors; reduced participation in workforce; and impact on a person's wellbeing. Apart from MB, all frameworks identified that self-management had been made a priority of previous strategies/policies pertaining to chronic disease management generally and/or the broader policy context of healthcare improvement and reform.

There was little data on the national healthcare systems and the influence of existing structures and governance on frameworks. SMS which was provided in the countries prior to developing a national framework was noted to be inadequate due to: variability across geographical areas (WL, IRL), with particular limitations in remote areas and for certain groups (MB, NT); involvement of different organisations and not well coordinated at whole system level (WL, NT); professionals not aware of services (WL, MB); and low rates of SMS from healthcare professionals (HCPs) such as receiving written information on how to manage condition (WL, MB, IRL). For SL, it was noted that services did not place needs of individuals with chronic disease at the centre of care.

2.3.2 Content

2.3.2.1 Scope of chronic disease supported

The extent to which the continuum of health was addressed by frameworks varied. IRL and SL focused mainly on self-management for individuals with a

diagnosed chronic condition. QLD and NT addressed chronic diseases as well as risk factors while others covered the 'continuum of care' (WL, WA, MB, TAS). Reference to the 'continuum of care' differed across frameworks. It seemed to include risk identification through to disease management in MB compared to involving more active health promotion and disease prevention in WL and TAS.

As shown in Table 2.2, the range of chronic diseases differed across frameworks, although not explicit in three frameworks (SL, WL, MB). The IRL framework focused on a narrow range of conditions compared to other frameworks such as TAS which was found to target the widest range of conditions. SL, WL and IRL gave attention to multimorbidity or having more than one chronic disease.

Table 2.2 Chronic conditions addressed by frameworks

Doc.	CVD	Diabetes	Respiratory	Renal	Musculoskeletal	Mental health	Cancers	Additional Information
QLD	X	X	X	X		X		
SL	-	-	-	-	-	-	-	Multimorbidity addressed.
WL	-	-	-	-	-	-	-	Multimorbidity addressed.
WA	X	X	X	X	X			Applicable to other conditions.
MB	-	-	-	-	-	-	-	
TAS	X	X	X		X	X	X	Additional conditions listed.
NT	X	X	X	X		X	X	Applicable to other conditions.
IRL	X	X	X					Multimorbidity addressed.

Note: conditions of focus were not explicit in SL, WL and MB.

2.3.2.2 SMS defining features

All frameworks offered defining features of SMS, though the emphasis of the definitions varied somewhat. Two frameworks focused on the components of SMS: resources, programmes, services, tools (MB); education and

interventions systematically provided (IRL). The remaining six frameworks referred to SMS in terms of where responsibility lies for providing it including healthcare systems, HCPs and social care providers, the community, and carers. Although the relationship between patients and HCP was noted as integral to SMS in all frameworks, the collaborative and shared responsibility was most explicit in some definitions (SL, WL).

2.3.2.3 Goal of framework

An overarching goal was explicit in five of the frameworks. These were: empowering and preparing individuals to actively manage their own healthcare (QLD, WA, NT), better outcomes for people with chronic diseases (TAS), participation of clients in healthcare 'within their communities' (WA); access to support (SL) and improvement of health services to support self-management (TAS, NT). Though not explicit, WL referred to coordination of care.

2.3.2.4 SMS priorities/ courses of action

All eight frameworks included a range of strategies for SMS at individual, HCP, organisation and system levels. Some approaches to SMS were frequently prioritised. These included ensuring the provision of self-management skills and education programmes for people with chronic disease and providing education and training for HCPs (Table 2.3).

The approaches to these common priorities varied and some frameworks had more clear and concrete courses of action than others. In all frameworks prioritising SMS programmes to educate and develop self-management skills of people with chronic diseases, there was an emphasis on increasing the availability of these services. Some frameworks were more specific in the

content of the programmes such as MB and IRL. The extent to which specific plans around training were detailed also differed across documents such as integration into undergraduate training curricula or continuing professional development.

Various courses of action were directed toward increasing the accessibility and appropriateness of SMS for different groups and individuals. Appropriate SMS for different groups was stated broadly as an action (QLD), or targeted through specific strategies. Some frameworks sought to increase provider awareness of patient programmes (WL, MB, TAS). Others proposed broader stakeholder and public awareness raising (QLD, WA, NT, IRL). In addition, frameworks emphasised different technological resources which could be used by stakeholders to support self-management (Table 2.3).

Table 2.3. Commonly prioritised courses of action to support self-management across frameworks

	Patient Education Programmes	Training for HCPs	Awareness raising	Accessibility of SMS	Technology to support SMS
QLD	Provide evidence-based programmes	Provide education and training	Social marketing campaign	Provide suitable SMS	Consumer personal health record
WL	Generic and disease specific	Skills training	Provider awareness of programmes	SM information in various formats, signposting	Technology for reminders, self-monitoring, follow-up
WA	Coordinated SMS programmes and services	Curricula, professional development, mentoring	Marketing strategy, framework endorsement	Easy referral pathways, flexible delivery of services	Website for all stakeholders
MB	Telecare programme prioritised	-	Provider awareness of programmes	Research suitability for different groups	Online health portal
TAS	Make programmes available	A range of training options, evidence-based practice	Provider awareness of programmes	Provide range of flexible resources	Range of resources including online
NT	Build capacity	Training and access to evidence-based practice	Framework endorsement	Clear referral pathways, SM information through various mediums	Electronic client record, SMS through online and other media
IRL	Map and increase provision of generic and disease specific	Curricula, professional development	SMS communication plan	Resources to account for health literacy, signposting	IT systems to support delivery, electronic patient record

Note: SL not included as it did not explicitly prioritise actions for implementation

2.3.3 Actors in framework development

There were differences across frameworks in the extent to which actors were detailed, the extent to which actors were involved as well as variability in the composition of actors (Table 2.4). The involvement of actors was not reported in WL or MB. The remaining six frameworks included actors and some form of working group. The working group actors were unclear in QLD. Otherwise the scope of the working group was usually small involving health division managers and/or HCPs. WA was the exception to this, involving all of its listed stakeholders in the working group.

It was explicit that people with chronic disease were involved in WA and IRL whereas in other countries, it was unclear if the involvement of consumer groups actually included people with chronic disease, although this was implied in the frameworks. In SL, it was emphasised that the framework was guided by people with long term conditions, though how this was achieved was not explicit.

2.3.4 Process

2.3.4.1 Development

In six frameworks, a group or team was responsible for the development phase, as seen in Table 2.4 (QLD, SL, WA, TAS, NT, IRL). The process of framework development explicitly stated the use of consultation in all countries. Consultation involved forums (QLD, IRL), an electronic questionnaire (QLD, WA), meetings with stakeholders/focus groups (QLD, WA, NT, IRL), and feedback and refining of drafts. Explicit reference to the use of previous literature and evidence was articulated in QLD, MB, TAS, NT and IRL and most others appeared to reference previous literature, though SL appeared more limited in this area. Four frameworks mentioned drawing on frameworks already developed in other states/countries (QLD, MB, TAS, NT).

2.3.4.2 Implementation

Four frameworks included an implementation plan with specific actions associated with specific timeframes (NT, WA, MB, IRL). In addition, the implementation plan in IRL included the specific personnel responsible for achieving each action. In QLD, WL and TAS, strategies were prioritised, but these were not associated with a specific timeframe (see Table 2.4).

Table 2.4 Policy actors and processes for each framework

Framework	Actors	Development processes	Implementation Plans	Evaluation Plans
QLD	Members of strategy team and alliance, government, non-governmental organisations, professional bodies, private sector, consumer advocacy groups, HCPs universities and national and international 'experts'	Consultation through forums, electronic questionnaire, feedback on drafts, meetings with key stakeholders. Evidence and existing frameworks.	Actions with timelines not reported. Proposed to be dynamic and flexible.	Plan/outcomes not detailed. Contains recommendations.
SL	Representatives from health and chronic disease organisations.	Working group and consultation on draft strategy.	Actions with timelines not reported. Processes of funding outlined.	Plan/outcomes not detailed. Contains recommendations.
WL	Not reported.	Not reported.	Actions with timelines not reported. Information on governance, infrastructure, incentives.	Plan/outcomes not detailed. Contains recommendations.
WA	Chronic disease consumers, carers, managers, policy developers, service providers, NGOs, researchers, self-management educators.	Consultation through an electronic qualitative survey, workshops with Strategy Review Group and feedback on draft.	Actions reported with timelines. Discussion of infrastructure, governance and funding.	Plan with timelines and actors responsible for leading evaluation.
MB	Not reported.	Guided by evidence, international and local.	Actions with timelines reported. Leading organisation and mechanisms of implementation identified.	Plan/outcomes not detailed. Contains recommendations
TAS	Reference group (HCPs and programme managers) and Chronic Conditions Clinical Network.	Consultation to identify priority areas. Use of specifically developed background paper and frameworks from other areas.	Actions with timelines not reported. Dynamic process, encourage uptake of resources by services and providers.	Plan/outcomes not detailed. Contains recommendations
NT	HCPs providing SMS, government, non-government and Aboriginal community-controlled health service providers, HCPs and consumer groups.	Developed by working group. Used focus groups (urban and remote). Consultation on draft, evidence of experience and frameworks from other areas.	Actions reported with timelines.	Plan reported with performance measures for actions and timelines.
IRL	Health division and programme managers/ coordinators and researcher, HCPs, patient and chronic disease-specific organisations, people with chronic diseases, government, health service and university representation.	Working group and consultation on draft through focus groups, interviews. Evidence used from literature, international policy documents, national survey and forum for patient consultation.	Actions reported with timelines and actors responsible. Plans for governance, infrastructure and resourcing.	Early process measures detailed. Contains further recommendations.

Frameworks differed in their plans for how implementation would unfold overall and in the actors responsible for implementation (see Appendix 2.B for further detail). QLD and TAS described implementation as a 'dynamic' process which would occur in the health service over time and would be flexible to challenges arising. The TAS framework encouraged services and workers to incorporate aspects of the framework and resources provided within the document. Actors involved in implementation were not detailed in NT though it was noted that a more detailed plan was expected to follow the framework document. A subsequent document was not identified through the search strategy. In SL, the Long Term conditions Alliance Scotland was to lead implementation including oversight of SMS provision and funding. WL identified leadership roles necessary to oversee framework implementation while MB identified primary care networks as facilitating implementation through various actions.

WA and IRL frameworks provided most detail on structures to support implementation. WA prioritised establishing a reference group and steering committee and partnership and funding strategies. In IRL a national governance structure was outlined detailing participation from each geographical area, the establishment of new roles and a group to oversee implementation, toolkits for implementation and the use of financial incentives.

2.3.4.3 Evaluation

All frameworks included some recommendation for evaluation. Evaluation outcomes were outlined for specific time framed actions in WA and NT. In IRL, process measures were outlined to evaluate the initial phase of implementation. Detailed plans were not outlined for other frameworks (Table

2.4). However, some information was provided on approaches such as: evaluation using existing reporting systems (TAS, IRL) or developing additional systems (IRL); incorporating outcomes as part of other evaluations such as evaluations of primary care (MB) or the broader chronic disease management strategy (QLD, NT); using a specific recommended framework for evaluation (SL) or an evaluation strategy being developed by each service area (TAS). SL emphasised an intent to use evaluation as a criterion for funding.

2.4 Discussion

In this cross-country comparative document analysis, SMS frameworks for chronic disease in OECD countries were examined, guided by the Health Policy Triangle. A search of grey literature sources and government websites identified eight frameworks published from 2008 to 2017. Unsurprisingly, the rising burden of chronic disease was the primary driver for the development of each framework. The scope of frameworks varied with some targeting chronic disease prevention and health promotion through to complex disease management. The range of chronic diseases also differed across frameworks. Few frameworks considered multimorbidity, though many individuals have more than one chronic disease and this is understood to pose particular challenges for self-management (Liddy et al., 2014).

Commonalities across frameworks were noted in the defining features of SMS and the goals of frameworks. A consistent pattern across the frameworks was that SMS involved empowering patients to actively manage their chronic disease in collaboration with HCPs. It is encouraging to find some

commonalities on what SMS requires of HCPs and how SMS is defined given that there is little conceptual literature on SMS and in recent years it has been noted as a developing concept (Kawi, 2012). There were also similarities in the courses of action prioritised to enhance SMS such as the use of SMS programmes by people living with chronic disease, training of health professionals, and raising awareness about SMS services. These similarities may be somewhat attributed to countries drawing on previously published SMS frameworks and using literature to inform framework development. While only some frameworks explicitly stated the use of literature and evidence to inform frameworks, most appeared to reference literature. This suggests a commitment to underpin SMS health policy with evidence though it is noted that there are barriers to the use of evidence in policy making which require further attention (Orton et al., 2011).

There was also variation across frameworks. Differences were noted in the content of frameworks, including the courses of action to facilitate SMS. For example, the Irish framework placed greater emphasis than some other frameworks on the need for generic and disease-specific supports and how these would integrate to provide comprehensive SMS (Chronic Conditions Working Group, 2017). Additionally, different strategies were used to target the appropriateness and accessibility of SMS for individuals and cultural groups across frameworks. Differences in courses of action may be to some extent attributed to inconclusive evidence on SMS approaches (Jonkman et al., 2016; Zwerink et al., 2014). Both the process of tailoring SMS and the combination of disease-specific and generic supports in this process have been highlighted to require further research (Trappenburg et al., 2013). The need for further

evaluation of SMS interventions and approaches was identified in the frameworks. Thus, it is possible that some of the variation in frameworks was due to the ambiguity of evidence in relation to optimal approaches to SMS. This underscores the importance of additional research on SMS approaches.

Only half of the frameworks included a high-level implementation plan, that is, strategies associated with specific timeframes for which they need to be achieved. This is surprising given the emphasis placed on the need for a strategic implementation process relating to national health policy and health service reform (Walt, 1998; World Health Organisation, 2010) as well as research indicating that SMS involves comprehensive and sustainable approaches (Kawi, 2012). The WHO points to the importance of being comprehensive in considering how framework changes need to be supported through health workforce resources, infrastructure, financing and governance (World Health Organisation, 2010). While all frameworks attended to structural and cultural components of organisational readiness for change (Gagnon et al., 2011; Weiner, 2009) in some form, there was variation in the extent to which personnel and infrastructure for implementation were addressed, with IRL (Chronic Conditions Working Group, 2017) and WA (Department of Health Western Australia, 2011) most comprehensive in this area. Furthermore, this document analysis found that the scope of chronic diseases covered by frameworks may be linked with the specificity of the implementation plan. Two frameworks (WA (Department of Health Western Australia, 2011), IRL (Chronic Conditions Working Group, 2017)) with detailed implementation plans and timelines explicitly targeted the least number of chronic diseases. The framework targeting the widest range of conditions included comparably less

information on specific actions and timelines for implementation (TAS; Department of Health and Human Services Tasmania, 2012). It is expected that frameworks with more specific detail and associated time framed implementation plans which are strategic and comprehensive would be successfully implemented (Walt, 1998).

Some frameworks allowed for greater local flexibility than others. Two frameworks (WA (Department of Health Western Australia, 2011), IRL (Chronic Conditions Working Group, 2017)) were very specific in outlining a governance structure to facilitate implementation of SMS. Others did not specify national governance structures and emphasised local flexibility, for example, services and providers choosing the resources and models appropriate to them (QLD, Queensland Government, 2008); TAS, Department of Health and Human Services Tasmania, 2012). Research from other policy areas has identified a need for balance between allowing local flexibility in implementation and consistent mechanisms of accountability at the broader governing level (Clement & Amezaga, 2009). However, the optimal balance for the delivery of SMS through a national health system is unknown and has yet to be researched.

Differences in flexibility and courses of action may also be related to design of the healthcare system within a country as has been found in the case of chronic disease management approaches (Nolte & Knai, 2015; Nolte et al., 2008). Limited detail was provided on the healthcare systems and their influence on SMS within the framework documents. Within the policy documents, it may be useful to include information on pertinent contextual

factors which influenced framework development to allow policymakers to consider context when drawing on aspects of policy from other countries. As a follow on from this document analysis, further research was considered to examine the experience of implementing SMS frameworks and to identify lessons for implementing SMS frameworks across countries. The context specific challenges and opportunities which affect implementation were also considered.

The involvement of actors varied across frameworks. Some documents did not detail consultation processes or actors involved. Where actors were detailed, it commonly involved health professionals and managers. Contrary to the emphasis on PPI in health strategies (Pizzo et al., 2015), direct involvement of people with chronic disease appears to have been uncommon. The potential benefits of involving a diverse range of actors who will implement and benefit from the framework suggest that this is a worthwhile endeavour which may enhance the success of a health policy and intervention (Pizzo et al., 2015). It is noteworthy that two frameworks which documented the involvement of a large range of actors including consumers of care also have highly detailed plans for implementation (WA (Department of Health Western Australia, 2011), IRL (Chronic Conditions Working Group, 2017)). This points to some potential advantages of PPI in developing a policy implementation plan, though the exact role of actors and their contribution to the policy process requires further research. This will be further explored in Chapter 3 of the thesis.

2.4.1 Strengths and limitations

Use of a policy framework and information categories to guide data collection are strengths of this study. The consistent application of a framework across documents from different health systems, as in this analysis, enhances the reliability of cross-country comparisons (Cacace et al., 2013). The Health Policy Triangle (Walt & Gilson, 1994) facilitated a comprehensive document analysis of SMS frameworks as an initial step to understanding their context, content, actors and processes. Other frameworks for examining policy (Schouwstra & Ellman, 2006) and theories of the policy process (Sabatier, 2007) were considered to be more narrow in scope, focusing on either the contents of policy or the actors/processes and requiring information beyond that which is provided through policy documents. It has been recognised that there is overlap between the components of the Health Policy Triangle (Buse et al., 2012). However, specific categories for extraction within each component of the Health Policy triangle facilitated analysis of key details in this work. Thus the framework provided a useful means of classifying information.

The grey literature search strategy, which was systematic and transparent is a strength of this study. Grey literature searching can be challenging in the absence of a 'gold standard' systematic method. Standards and methods proposed in previous work (Godin et al., 2015) were drawn on, therefore conducting a rigorous and robust method of searching web-based sources of grey literature. However, there were some limitations in the grey literature search. The lack of ability to automatically export all documents meant that only one researcher, instead of two, carried out the initial phase of screening. In addition, weaknesses in archiving various details of documents as well as

the means by which some search engines develop relevance ratings may have affected the retrieval of relevant documents (Godin et al., 2015).

The search was limited to documents from OECD countries and documents available in the English language. While, eight countries might be considered a limited representation of national SMS frameworks, these may largely represent health systems which have produced system-wide SMS policy documents. Language is unlikely to have severely restricted the search given the publication of some health policy documents from non-English speaking countries through English and the dearth of documents identified in English speaking health systems such as those in the United States. Countries which are not represented may have taken different policy approaches to chronic disease management with a strategy for SMS integrated into these policies rather than having a national strategy specifically for SMS in healthcare. Thus, the findings compare national SMS frameworks for chronic disease and provide a platform for further research on the operationalisation of SMS frameworks for chronic disease management.

2.4.2 Conclusions

This study identified eight policy documents developed by national and state health departments in OECD countries over the past 10 years which aim to improve the provision of SMS for chronic diseases. This study served to illuminate and compare the contents and processes of existing policy for chronic disease SMS, an area which was not previously explored. Countries at early stages of SMS policy development and implementation can draw on this study to inform their national strategies for chronic disease healthcare.

While there was evidence that SMS frameworks in some countries drew on the work of other countries, there was little evidence of active engagement between policy makers in different countries to learn from each other. Given that health policies are not always implemented as planned; it is important to understand the success factors or barriers relating to implementation so that proposed plans for SMS can be operationalised in ways that contribute to reducing the burden of chronic disease. Factors were identified in this study which could influence the implementation of SMS policies. Further research to examine the influence of these factors is described in Chapter 3.

Chapter 3. The process of implementing system-wide frameworks on self-management support for chronic disease: a qualitative multi-country analysis

This chapter comprises a qualitative study of the implementation of three of the eight self-management support (SMS) frameworks included in the document analysis. Information on the health service contexts of the frameworks is outlined in the background to the study. Lessons for the implementation of SMS frameworks are drawn and the findings are discussed. This chapter has been submitted for publication and is under review in *Health Policy.*

Abstract

Background: SMS aims to enable individuals to manage their chronic disease through changes at patient, provider, organisational and system levels of healthcare. National and state-level policy frameworks have been developed to progress the provision of SMS for chronic disease. However, the process of implementing these system-wide SMS policies is not yet well understood.

Methods: Semi-structured interviews were used to explore the process of implementation with key informants, who had a role in directing the implementation of a framework. Thematic analysis was used, and theoretical literature was incorporated into the development of themes, drawing on a multiple streams approach to policy implementation.

Results: Six participants reported on implementation within three health systems that have produced SMS frameworks. Progress with framework implementation was often through a process of continued prioritising and adapting framework plans. Challenges for implementation included ambiguity in the definition and operationalisation of SMS; resistance from healthcare professionals; the crisis driven nature of health systems; and changing political contexts which alter health system structures and priorities.

Conclusions: Greater consultation with people at the centre of SMS, including people with chronic disease, may help overcome challenges in the definition of SMS and resistance to implementation. Communication infrastructures are needed to maintain support for implementing SMS. Global learning across countries may help to advance implementation of SMS for chronic disease.

3.1 Background

Self-management is a process through which people actively cope with chronic disease in the context of their daily lives through monitoring and responding appropriately to changes in their physical and emotional wellbeing (Miller et al., 2015). SMS involves a range of interventions which aim to enable an individual to engage in self-management (Miller et al., 2015). SMS interventions have improved health outcomes for chronic diseases such as diabetes, respiratory and cardiovascular conditions (Chrvala et al., 2016; Panagioti et al., 2014b; Zwerink et al., 2014). They have also been found to improve health-related quality of life and reduce hospitalisation for people living with chronic diseases (Panagioti et al., 2014b; Zwerink et al., 2014). While more data is needed on cost-effectiveness at health system levels, it is

generally reported that costs of interventions are low compared to the overall expenditure on chronic diseases (Health Information and Quality Authority, 2015).

SMS interventions have been delivered in many different formats and include different components such as education and information provision, goal setting and problem solving (Battersby et al., 2010). Recent studies highlight that SMS involves a comprehensive systems approach which is most effective when interventions target patient, healthcare professional (HCP) and organisational levels rather than any one level alone (Kawi, 2012; Pinnock et al., 2015). While components of SMS may be included in broader disease management strategies, frameworks specifically targeted at providing SMS for people with chronic disease have been developed within a number of national and state level health systems (O'Connell et al., 2018). A previous systematic search identified eight SMS frameworks published between 2008 and 2017 (O'Connell et al., 2018). They were developed in Ireland; Scotland and Wales in the United Kingdom; the province of Manitoba in Canada; and four Australian states, namely, Queensland, Western Australia, Tasmania and the Northern Territory. Some of the key priorities for providing SMS were providing patient education programmes, training for HCPs, raising provider awareness of programmes, increasing accessibility of SMS through referral pathways and offering resources in different formats (O'Connell et al., 2018).

However, the frameworks differed on the extent to which they had specific plans to achieve improvements in SMS including variation in timelines and governance which have the potential to influence the success of

implementation (O'Connell et al., 2018) (see Table 3.1). Furthermore, many healthcare policies including national chronic disease management strategies have not achieved their implementation targets (Begum & Por, 2010; McHugh et al., 2014). Therefore, research is needed to understand the factors influencing SMS framework implementation in order to inform future SMS implementation processes.

Table 3.1 Implementation plans for SMS frameworks

Title, Year	Proposed Implementation
Queensland, 2008	Plan with timelines not reported. Proposed to be a dynamic process aided by initiatives within different levels and sectors of health service
Scotland, 2008	Plan with timelines not reported. LTCAS leading: establishing advisory board and personnel to map SMS provision, provide feedback and oversee allocation of specific fund for SMS.
Wales, 2009	Plan with timelines not reported. Processes outlined including identifying national and local leads, an infrastructure for local training, and use of incentives to adopt training. Considers leadership and collaboration across stakeholders.
Western Australia, 2011	Includes actions for short term (2 years) and longer-term goals (2-4 years). Initial implementation plans provide infrastructure for implementation. Process includes establishing reference group and steering group as well as partnership and funding strategies. Evaluation plan included
Manitoba, 2012	Plan with timelines. Planned to achieve through Primary Care Networks and involves increasing IT connectivity, group sessions and outreach.
Tasmania, 2012	Plan with timelines not reported. Dynamic process which occurs in different ways over time across the state with services encouraged to use resources provided. Some aspects to be implemented as part of other frameworks
Northern Territory, 2012	Includes implementation plan with number of actions to be taken in short (2012-2014), medium (2015-2017) and long term (2018-2020). Evaluation plan included
Ireland, 2017	Implementation plan included with timelines. Plans for governance structure e.g. national and local leads; SMS coordinators (mapping local SMS provision and creating directories); partnerships between organisations; and financial incentives. Early process measures of implementation outlined.

Source: Adapted from previous paper (O'Connell et al., 2018)

Howlett and colleagues (Howlett, 2018; Howlett et al., 2015, 2017) recently combined and adapted theoretical concepts to provide a unifying approach to understanding the complex process of policy implementation (Figure 1.3). Within this approach, it is proposed that implementation can be analysed through the lens of the Multiple Streams model (Kingdon, 2014), that is, the

framing of the *problem* being addressed; the nature of the *policy* being developed; and the *political* context. Howlett et al. (2015, 2017) combined other theoretical perspectives to identify two further streams. The original three streams are combined in a *process* of decision making and formulation around the policy. Additionally, the *programme* stream directs attention to on-the-ground implementation of policy and the actors involved in delivering the policy (Howlett, 2018). This approach highlights how policy implementation is influenced by immediate factors such as the HCPs and organisations implementing policy (Walker & Gilson, 2004) but also how implementation of healthcare policies can be affected by the broader policy environment such as changes in political leadership (May et al., 2014; McHugh et al., 2014).

The multiple streams/critical juncture approach (Howlett, 2018) highlights the importance of the health policy environment and the context of the health system in order to understand the factors affecting implementation. Previous studies have also shown that the context of health services influences the system changes implemented as part of chronic disease management initiatives (Nolte & Knai, 2015; Nolte et al., 2008). This study invited key informants from the eight health systems with SMS frameworks (Table 3.1) to participate. The eight SMS frameworks originated from different national health system contexts (Australia, Canada, Ireland and United Kingdom) which may shape implementation (Table 3.2).

Table 3.2 National health system context for SMS frameworks for chronic disease (Mossialos et al., 2017; O'Connell et al., 2018; OECD/European Observatory on Health Systems Policies, 2017)

	Australia	Canada	Ireland	United Kingdom
Service delivery	• Primary health networks involving MDTs being developed to support after-hours care, locally tailored services. • Varying access across urban and remote regions and indigenous populations.	• Developing interdisciplinary primary care networks to increase accessibility • Varying access across large remote areas and indigenous population.	• Integrated care programmes in process of implementation • No universal coverage for primary care which presents a barrier for access.	• Growing focus on integration of health and social care. • Socioeconomic disparities and large remote areas as in Scotland. • Low levels of unmet need.
Governance	• National government sets safety and quality standards. State and territory have majority of responsibility for services. Local government play a role in community health.	• Provinces and territories primary responsibility for delivering services. Regional health authorities plan and deliver locally.	• Health Service Executive responsible for provision of care; planned to be further devolved to regional bodies	• UK government allocates money to devolved administrations such as Scotland and Wales who determine how funding will be used.
Financing	• Public and private system. Medicare scheme provides free or subsidized access. • Out-of-pocket 18% of total spending	• Public and private system. Public covers physician, diagnostic and hospital with additional varying by province. • Out-of-pocket 14% of total spending	• Public and private system. 47% qualify for free GP visit under two different schemes • Out-of-pocket 15% of total spending	• Universal access to healthcare at the point of use based solely on need. • Out-of-pocket 15% of total spending

Note: frameworks identified from each national health system: Australia - Queensland, Western Australia, Northern Territory and Tasmania; Canada - Manitoba; Ireland – national framework; United Kingdom – Scotland and Wales.

Frameworks have been developed to enhance the provision of SMS throughout a health system. However, the large scale programme-type reform required by these policies is a challenge for implementation (Walt, 1998). There is a dearth of research on system-wide implementation of SMS which is imperative to understanding how these system-wide changes can be realised in practice. This study aimed to explore experiences of implementing system-wide frameworks to support self-management of chronic disease.

3.2 Methods

3.2.1 Study Design

This study was guided by the principles of pragmatic qualitative inquiry with the focus on seeking practical applications toward a problem (Patton, 2015). Experiences of implementing SMS frameworks for chronic disease were explored through interviews with key informants. The Consolidated Criteria for Reporting Qualitative research (COREQ; Appendix 3.A) was used to guide the reporting of the research (Tong et al., 2007).

3.2.2 Participant Selection

Eligible participants were key informants, that is, individuals who were knowledgeable on the topic of interest, who were articulate in providing this knowledge and understood the focus of inquiry (Patton, 2015). For this research, individuals who had a lead role in overseeing or directing the implementation of national or state-level frameworks for SMS for chronic disease were selected. This could include individuals of various titles such as implementation leads, programme managers, coordinators at

regional/state/national levels who have a role in implementing relevant frameworks.

Key informants were identified through contacting the authors/authoring bodies of the eight previously published SMS framework documents (O'Connell et al., 2018). A process of snowball sampling was also used to identify additional potential participants meeting the criteria. Email invites were sent to prospective participants along with the study information sheet and consent form. Potential participants were not identified from two of the eight frameworks. Twelve potential participants from the remaining frameworks were contacted of which six agreed to partake in interviews. These six key informants addressed three of the eight frameworks presented in Table 3.1. The three frameworks studied are not named in the analysis in order to preserve the anonymity of key informants who would otherwise be identifiable.

The study was approved by the Social Research Ethics Committee, University College Cork (Appendix 3.B) Participants were informed of the purpose and nature of the research in a study information sheet (Appendix 3.C). Consent to participate and permission to publish data were provided through an electronically signed consent form prior to interview and was confirmed verbally at the beginning of the interview.

3.2.3 Data Collection

Telephone interviews were conducted using a piloted interview guide based on previous literature on policy implementation (Table 3.3). Each participant was interviewed once with interviews lasting between 40 and 50 minutes. All interviews were carried out by one researcher (SOC) who was a PhD student

with a background in Psychology. Notes were made during the interviews. Participants were given the opportunity to clarify and add/remove points at the end of the interview. All interviews were audio-recorded and transcribed.

Table 3.3 Topic Guide used during interviews

Story of Implementation from beginning to present
Extent to which implementation plans have been achieved
Facilitators of implementation
Barriers/ strategies to overcome barriers
Progress in evaluation of frameworks
Current impact of frameworks on health service and people with chronic disease
Advice for other countries in early stages of implementation

3.2.4 Data Analysis

A process of thematic data analysis was used which involved immersion in the data through repeated reading of transcripts and listening to recordings, coding of the data, categorisation of the codes and the identification of themes (Green et al., 2007). Coding of manuscripts was carried out by the PhD researcher (SOC) and samples were cross-checked by the research supervisor (ES). Coding was carried out by hand using notes in margins. Handwritten memos were taken throughout data collection to inform coding and category development. Codes were grounded in the participant's words and were organised into categories. Theory and literature were incorporated in the process of developing themes to conceptualise the patterns in the data and strengthen interpretations (Green et al., 2007). The data were searched for alternative themes and examination of negative cases contributed to some revision of the thematic structure (Patton, 2015).

3.2.5 Trustworthiness

For the purposes of interviewing key informants, the PhD researcher familiarised herself with the content of SMS frameworks and initiatives in the studied healthcare systems as well as the terminology used in order to facilitate interviewing and further probing (Patton, 2015; Rabionet, 2011). Credibility of the findings was enhanced through building rapport with participants prior to commencing interview and note-taking was limited to key issues during the interviews in order to ensure that note-taking did not detract from responsiveness of the interviewer, a risk that has been noted in relation to telephone interviews in particular (Irvine et al., 2013; Patton, 2015). Credibility was also enhanced through systematic field work and processes of reflexivity including notes taken directly after interview; memos during data analysis; searching for alternative patterns in the data; and the checking/ feedback on coding by a supervisor experienced in qualitative research (Patton, 2015). An audit trail of the study processes enhanced dependability.

3.3 Results

The implementation of three frameworks from three different national health service contexts was represented by six participants in this study. While the frameworks had varying contexts for implementation (Table 3.2), across all three, implementation involved prioritising certain actions over others and adapting the policy solution based on available opportunities to advance implementation. The five streams model proposed by Howlett (2018) was used as a theoretical lens to describe this process and the data are presented according to these streams below (see final coding tree in Appendix 3.D). Based on the results, lessons for implementation are presented in Table 3.4.

Table 3.4 Lessons for Implementation of SMS frameworks

Strategies for implementation	Challenges to address	Practical approaches
Understanding stakeholder priorities, particularly patients and HCPs	• Clarify objectives/purpose of SMS • Clarify SMS operational definition • Provide evidence for approach • Understand messages for different audiences for securing buy-in	• Early stage patient involvement • Drawing on patient perspective rather than data pertaining to them • Patient stories/surveys • Stakeholder planning meetings
An adaptive and collaborative approach to implementation	• Facilitate integration of SMS into everyday practice • Reduce negative impact on workers • Reduce resistance • Provide greater opportunities to move forward	• Linking with other health policy drivers • Identifying necessary infrastructures such as shared electronic medical records, tools/forms which ease HCP work • Working with partners to understand how SMS can be most easily incorporated e.g. inclusion in curriculum for HCPs
Communication infrastructure across HCPs and managers	• Ensures clarity around purpose and operationalisation of SMS • Enable champions to motivate • Enable problem solving	• Networks of primary care providers • Interdisciplinary teams • Designated meetings of personnel across regions • Engagement of HCPs through training
Ensure long-term communication infrastructure with senior/political levels	• Reduce policy-service gap • Secure support/resources in context of competing priorities	• Internal mechanisms of communication within health department
Outline deliverables and embed in evaluation systems	• Ensure SMS is a priority in practice • Clearly identify problem areas and enable problem solving	• Yearly targets, quality indicators • Component of reporting within system e.g. primary care system reporting
Embed SMS approaches in other long-term health standards at multiple levels of governance	• Facilitate integration of SMS into everyday practice • Ensure SMS approaches are priority and funding provided • Maintain support over long-term	• Components of SMS included in national, state-level and regional standards and policies • Framework elements link with close areas such as health promotion, primary care, community care standards
Balance centralised processes and decentralised tailoring to local needs	• Ensure appropriateness of service for local population and still provide mechanisms of accountability	• Common reporting mechanisms for networks but each network tailoring staff and organisation of service to meet local needs.

3.3.1 The Problem Stream

3.3.1.1 Framing of the problem

Participants frequently referred to the goal of SMS as involving better experiences or outcomes for the person with chronic disease. SMS was also presented as reducing healthcare usage or hospitalisation by some participants. However, participants cautioned about selling SMS in this way, with a risk that self-management could be seen as people being left to manage conditions alone, without the support of the health service.

> Sometimes self-management has been sold as a money saving thing or a 'this is going to stop people using health services' whereas in fact that's not what it's about, as you would know. It's about people more appropriately using health services and being able to navigate for themselves, or with support and assistance, through a system that's incredibly complicated (P03)

3.3.1.2 Competing priorities

Participants highlighted that health services are dominated by a reactive approach to healthcare provision with funding primarily flowing toward hospital care and short-term solutions, often driven by media reporting. Participants spoke of difficulties in getting support for the proactive approach of prevention and management of chronic disease through community and primary care. Across the three frameworks, funding and resources necessary for the implementation of SMS were challenging to secure in the context of competing priorities.

> The needs of the acute crisis driven or reactive services are paramount, they can't be ignored so that is where the funding goes and

> unfortunately it doesn't go to developing new ways of working or at least some of it does but not enough (P01)

3.3.2 The Policy Stream

3.3.2.1 Wide scope of policy solution

A consistent message raised was that SMS implementation was a large task and that it was difficult to progress implementation across individual, HCP, organisational and system levels simultaneously. Health systems were often more progressed at one level. For example, one system was progressing well with SMS group patient education programmes and another with education of HCPs. However, the benefits of this training were being hindered somewhat by a lack of supportive systems. In another system, services were developed more in terms of the infrastructures that would enable multidisciplinary care underlying SMS with less detail on the contents of SMS to be delivered in practice.

> We had the feedback from people that they really enjoyed the training and… but a frustration that the systems didn't change enough to support them to put it into practice in a real and consistent way (P03)
>
> I think it's the detail of how we implement and deliver self-management that has perhaps had less of a focus as we've advanced on the broader system pieces (P05)

3.3.2.2 Ambiguity of terminology

The terminology of self-management or SMS was recognised as a challenge across interviews. Participants felt that sometimes people do not know what SMS means and that different people adopt different views of self-management and SMS.

> I don't think we've done a thorough assessment of what people think self-management is. I just know in conversation at different tables when you hear people talk about self-management you very quickly realise that everyone interprets that a little bit differently. Some people will use the word self-management but still be talking very much about a top down approach to education with a patient (P05)

3.3.2.3 Acceptability of framework changes

Variation in the reactions of stakeholders implementing the policy on the ground was noted. Participants spoke of some stakeholders being very supportive of implementation while they also noted resistance to new ways of working and concerns that changes in HCPs roles could negatively impact on quality of service. There was a suggestion that the language of self-management may be threatening for HCPs and that it may be difficult to break the mind-set of doctor as expert.

> I think it's to do with the way we train health professionals still too. That we still train them, we train them to be the experts, which of course you want an expert to be looking after you, but you hope that its more in partnership (P03)

3.3.3 The Politics Stream

3.3.3.1 Service delivery context

The structural context of health services shaped implementation. Participants across the three frameworks described governance arrangements of their system including responsibilities of national, state and regional levels. They explained that funding allocation processes and geographical distributions influenced implementation. Previous system reforms seemed to have

contributed to a widening of the gap between policy makers and those delivering services.

> We had more of a service delivery role and function, and that has certainly changed and we are now much more policy oversight funding role and so self-management might be seen as more of a service delivery component. So again, we have to work within that role definition and ensure that that happens at the appropriate level (P05)

Some participants noted that although they had buy-in from senior health officials, implementation could be better facilitated by an ongoing connection with the senior health department officials. One region was noted to be progressing well, a factor thought to be a result of having greater access to senior leaders. Health services differed on the extent of implementation flexibility at regional and local levels. Most participants talked favourably of the role of leadership at regional levels and the benefits of tailoring the SMS approach and the workforce to meet the needs of the local population: '*to recognise what can be centralised and what has to be decentralised'* (P06).

3.3.3.2 Uncertain health policy environment

There was frequent reference to the unstable environment which surrounds health policy implementation. At a political level, changes in government were seen to undermine plans for health polices through changing priorities for implementation. Policies and political support which prioritised SMS were key to developing frameworks and progressing implementation. However, when these were changed it reduced the support for SMS initiatives. In one health system it was noted:

> We'd also had a significant change in the structure of our health system […] basically we had internal changes within our department which meant that there were other priorities that were named up (P02)

Changes in government also were disruptive through instigating structural changes in the health service. This required extra time and effort from workers to understand how the structure should function and where the responsibility for different tasks lies.

3.3.4 The Process Stream

3.3.4.1 Prioritising parts of implementation

In the three health systems, parts of the SMS frameworks were at an early implementation stage as planned. Prioritising was described as key to early implementation. A consistent priority across the health systems was mapping the existing services for SMS and highlighting gaps, '*creating the interest in solutions'* to facilitate planning of the local approaches to implementation. While some parts were prioritised, others were delayed compared to original timelines, as one participant noted:

> I think you know some of the things that we thought might have happened in Year 1, you know, haven't happened yet but some of the Year 2 things are moving forward (P04)

3.3.4.2 Adapting

Other components of frameworks seemed to have been adapted during the course of implementation. There were different levels of adapting from changing slightly the design of original proposals to a complete overhaul of SMS approaches. Adaptation was normally in the context of linking in with emerging health policy drivers including the emphasis on digital accessibility,

health promotion with a focus on risk factors and health literacy. It was emphasised that it was necessary to take advantage of opportunities to move forward, keeping the end focus of patient self-management in mind.

> So while we hadn't really planned it that way, we are fitting in with the overall [health service] development there and changing our parameters a little bit to suit that but it actually makes it more suitable as a service than we had planned (P01)

3.3.4.3 A long-term gradual process

While frameworks often had targets for implementing SMS within 3-8 years, it was clear from the data that implementation of SMS throughout a health service was perceived as a long term process which would require cumulative effort of different stakeholders over time to become an integrated part of care for chronic conditions.

> It's not going to be done in two or three years. It's really something that has to be … adopted … over 10/15/20 years really to transform the way we do things (P01)

3.3.5 The Programme Stream

3.3.5.1 Consumer and healthcare professional engagement

Engaging a wide range of stakeholders was seen as key to successfully implementing SMS. While user and patient experience was frequently mentioned as important in the frameworks, it was acknowledged that consultation with people with chronic disease and incorporation of their perspectives in the design of SMS approaches has not been done well and this has been 'a missing link' in the implementation of SMS.

> I think that's work that we still need to do. It's not so much the patient informing those service plans … as it is looking at the information and data that's available about the needs of the population, so I think we have work to do in bringing the patient voice to that planning process (P04)

For HCPs, communication was felt to be essential to facilitate buy-in. This was both in terms of understanding their perspectives '*to look more strongly in terms of what's the relevance of this to people working within the health system* (P02)' and to inform HCPs about proposed changes: '*Communication is where we have had any success in allaying peoples' anxieties'* (P01). Communication was often initially in the form of consultation and needed to continue throughout implementation in the form of collaboration, such that a framework could be implemented in ways that did not negatively impact on staff. Communication was noted as challenging, but some practical suggestions were identified (see Table 3.4).

Many participants supported the idea that champions were key to progressing the concept of self-management and the implementation of support. Champions often existed at regional levels and were felt to engage multiple stakeholders in implementation, including other providers as well as leaders and funders.

> We also had some champions from within the health system who had chronic conditions themselves and they were really important in kind of selling it to their colleagues and managers as saying, you know, this has helped me live my life (P03)

3.3.5.2 Integrating SMS into practice

Participants noted challenges in linking SMS into the day-to-day work of HCPs. They noted that SMS was seen as an add-on, something extra to be done in an already time-constrained environment. In addition to increasing the engagement of stakeholders, creating better linkages from a systems perspective was suggested to improve integration of SMS, e.g. linking community SMS programmes with primary care. A consistent message from participants was that embedding self-management in other health standards and often multiple policies at various levels such as regional, state and national, provided greater support for implementation.

> Where we've been more successful with the [named area of work] is that we've linked it really strongly to [national standards]. So I think that's been a real lesson because that's been a driver for the health system (P02)

3.3.5.3 Evaluation and deliverables

In addition, embedding SMS in mandated service reporting was felt to ensure that self-management remained a priority. Where participants had functioning reporting structures, they were reported to be useful in keeping track of implementation and problem solving when not meeting targets.

> So being able to identify targets; being able to measure and then report on them has just helped to identify are we on track, if we're not why not, and how do we problem solve that (P04)

3.4 Discussion

The three health systems studied had developed SMS provision and supportive infrastructures which reflect the objectives of their frameworks for

SMS for chronic disease. However, the process of implementing frameworks for SMS differed from the initial plans. The data supported the view that policy stages are neither discrete nor consecutive (Howlett, 2018; Sabatier, 2007). In the “implementation phase” there were still processes of decision-making and formulation taking place which altered the course of implementation. During implementation, certain parts of SMS were prioritised over others for early implementation and other parts were adapted to fit in with other health service drivers such as increasing accessibility through digital technologies and primary care restructuring. Thus despite varying approaches to SMS across systems, an important factor in the implementation of SMS approaches was the extent to which plans could be adapted to fit in with health service priorities, an important determinant in implementation science literature (Damschroder et al., 2009; Greenhalgh et al., 2004).

The findings indicate that it has been more difficult to advance individual, healthcare provider and systems levels of implementation simultaneously. While the frameworks had timelines ranging from 3-8 years, the findings suggest that it would take much longer for the concepts of SMS to embed and become an integral part of care delivery. Thus it may be that implementation of SMS is a slow gradual process in which developments will come together over time similar to the 20-30 year progress of policy on smoke-free environments (Currie & Clancy, 2011). This time scale suggests two points for consideration regarding the implementation of SMS frameworks. Firstly, how implementation can be sustained across the changing healthcare environment and secondly, if there are factors that could propel the implementation of SMS

forward so that it can efficiently and effectively meet the urgent needs of the growing population of people with chronic diseases.

Regarding the first point, a challenge for implementing frameworks for SMS was sustaining implementation within the unstable health policy environment. Changes in government undermined support for the policy idea and the structures underpinning implementation. Much literature highlights how directions of health policy are interrupted by changes in political agendas (May et al., 2014; McHugh et al., 2014). A critical component of implementing SMS frameworks is thus securing long-term support. The findings suggested that embedding the principles and approaches of SMS in broader healthcare reform and standards at national, state and regional levels can help to ensure that central ideas stay on the agenda. Champions were also reported to influence senior leaders. However, sustained mechanisms of communication which provided regular access to government and political leaders were found to be key to maintaining political support, as in other areas (Pelletier et al., 2011).

However, the findings also suggest that the policy process could be improved to help advance implementation of SMS. The findings suggested there is both a need and potential for increased stakeholder engagement. This study, like others (Johnston et al., 2011), also highlighted that there remains ambiguity among HCPs over what SMS means and how it is to be operationalised. A notable gap highlighted in this study and elsewhere (Pumar-Méndez et al., 2017; Reidy et al., 2016) is the lack of understanding and consideration of the priorities and preferences of people with chronic disease. Greater

understanding of what people with chronic disease value and need from SMS may help refine the understanding of how to operationalise SMS.

Though many HCPs and managers were engaged in policy development, resistance to some ideas was noted as in previous research (Joseph-Williams et al., 2017; Kennedy, Rogers, Chew-Graham, et al., 2014). This study reinforces the message of previous research regarding the need to understand how HCPs define SMS, value it and their readiness to change (Davies et al., 2018; Gagnon et al., 2011) and to include this in planning service change for SMS. However, the optimal mechanisms of feedback and collaboration present a direction for future research.

Health systems had varying contexts of care delivery and different geographical dispersions and cultural variances. It was emphasised that while it would be possible to learn from other countries, it was imperative to examine what fitted within the context of a unique health service. This finding reflects the benefits and cautions of cross-country comparisons reported elsewhere (Cacace et al., 2013; Marmor et al., 2005). Previous research which has joined the perspectives of key stakeholders across countries has helped identify directions for SMS frameworks, with recommendations that reflect the lessons identified in the current study (Mills et al., 2017). Similarly, this research suggests that cross-country initiatives can provide insight into strategies for the implementation of SMS. Common factors from implementation science literature have been identified across countries. The findings of this study point to the importance of flexibility to the local context as well as the importance of adaptability to changing health service agendas (Damschroder et al., 2009;

Greenhalgh et al., 2004). Similar to other policy areas (Clement & Amezaga, 2009), it was suggested that there needs to be a balance of common mechanisms of implementation across a health service with regional and local approaches tailored to the needs of the population.

3.4.1 Strengths and limitations

This work was exploratory in nature and incorporated theory to strengthen themes developed in the analysis, allowing for novel aspects of the implementation to emerge while also grounding the findings in theoretical work. While other theoretical frameworks were considered for describing the data (Kingdon, 2011, 2014), the recent approach of Howlett (2018) which focuses on the process of policy implementation provided optimal fit for the findings.

While pertinent contextual information has been provided as far as possible, the specific health systems represented were not named in order to preserve the anonymity of participants. This places limits on the contextual information available to the reader. The study also represented a small number of health systems that are at different time points in implementing guiding documents for SMS and thus the analysis may not be representative of implementation processes across heath systems with SMS frameworks. Nonetheless, the analysis identified facilitators and barriers across three health systems implementing SMS frameworks which are also strengthened by a theoretical framework and wider theoretical literature on policy implementation. This analysis focuses on health systems which have produced framework/policy documents specifically targeted at supporting or enabling self-management

across chronic diseases (O'Connell et al., 2018). SMS initiatives exist in other countries under policies with different emphases such as broader chronic disease management which are not covered by this analysis.

The study is based on the perspectives of individuals responsible for directing implementation and thus may be influenced by social desirability and not represent implementation issues experienced by other stakeholders such as local managers, HCPs or people receiving support. However, social desirability was found to be minimal in interviews with participants speaking openly about issues affecting implementation. Evaluation with other stakeholders and reports on the progress of framework targets can help to shed additional light on the barriers/facilitators regarding implementation.

3.4.2 Conclusions

Substantial progress has been made in enhancing the provision of SMS outlined in frameworks. Across the three SMS frameworks included in this study, implementation was a long-term process which involved prioritising certain parts of frameworks for action and adapting plans to align with other drivers in the health service. The similarities in implementation challenges arising across the three health systems implementing SMS through the frameworks examined for this study, suggests global networking can help advance SMS implementation. Securing the long-term support of funders and senior officials was key to implementation and requires consistent communication infrastructures. Future research needs to focus on understanding the priorities of people with chronic conditions in order to clarify how SMS should be operationalised in practice. The subsequent chapters of

this thesis seek to understand the SMS preferences of people with asthma/COPD.

Chapter 4. Self-management support preferences of people with asthma or COPD: a systematic review and meta-synthesis of qualitative studies

Based on the emphasis placed on the value of the patient perspective on self-management support (SMS) in Chapter 3, the focus of this thesis evolved to study the preferences of people with chronic disease. This chapter comprises a review of qualitative literature on the preferences of people with asthma and/or COPD regarding support to self-manage their conditions. This review of the literature provides an initial step toward identifying attributes of support for inclusion in a discrete choice experiment (Bridges et al., 2011). This content of this chapter is published in *Chronic illness* (O'Connell et al., 2019).

Abstract

Objectives: to synthesise findings from qualitative studies on the preferences of people with asthma or COPD for SMS.

Methods: A thematic synthesis of literature was carried out. Six databases (ASSIA, CINAHL, MEDLINE, PsycINFO, Psychology and the Behavioural Sciences, and SSCI) were used to search for qualitative studies eliciting perspectives of adults with asthma and/or COPD on SMS, published between May 2008 and April 2018.

Results: A total of 968 articles were retrieved across databases, with 15 articles included in the synthesis. Three themes were identified: *Types of Support* described the range of supports valued by participants in the studies,

particularly education provided by competent healthcare professionals; *The Support Relationship* highlighted the importance of a collaborative relationship with one's healthcare professional which was characterised by communication, trust and continuity over time; and *Accessibility* identified the considerations of participants relating to physically accessible, prompt support which is provided in a format preferred by the individual.

Conclusion: Increased understanding of patients' preferences may provide insight which can be used to enhance engagement with SMS. Further research needs to examine SMS preferences outside the context of evaluating interventions for people with asthma/COPD and needs to address the optimal means of enhancing accessibility.

4.1 Background

Self-management of chronic disease is a process where individuals with chronic disease actively manage physical, social and emotional wellbeing in the context of daily life (Miller et al., 2015). Self-management for people with COPD or asthma frequently involves lifestyle adjustments such as: smoking cessation and exercise; use of medications; inhalers, oxygen therapy; managing psychological consequences; and managing symptom exacerbations (Kaptein et al., 2014; Pinnock, 2015). Support is necessary to help individuals accomplish these self-management tasks. SMS interventions for both COPD and asthma have had beneficial outcomes for patients including better health-related quality of life, symptom reduction and reduced hospitalisation (Pinnock et al., 2017; Zwerink et al., 2014).

Early conceptual work indicates that support for self-management of chronic disease is a multilevel process involving the patient, healthcare professional (HCP) and wider organisational and healthcare system structures (Kawi, 2012). People with chronic disease are supported by HCPs through a relationship which is characterised by partnership and shared decision-making to facilitate tailored education and care (Kawi, 2012). However, while interventions to support self-management of asthma and COPD have had positive outcomes for patients, there have been inconsistencies in the results and there remains no clear indication on what components of interventions are most effective (Pinnock et al., 2017; Zwerink et al., 2014). Patient attendance (Sohanpal et al., 2015) and engagement in the programmes (Zwerink et al., 2014) may influence outcomes. Despite this, little information is available on the preferences of patients regarding SMS and whether interventions are designed taking account of these preferences.

Reviews have synthesised barriers and facilitators of self-management in asthma and COPD from the perspectives of patients and HCPs. While the input of all stakeholder groups is needed to develop SMS for implementation, there is a strong rationale for ensuring that service developments in SMS are first grounded within the perspectives of people with asthma/COPD for whom the support is intended to benefit. Using patients' preferences to inform healthcare is not only important in that people have a right to have a say in the care they receive (Rosenbaum, 2013) but also because it has the potential to improve healthcare so that it is better tailored to patients' needs at an individual and population level (Yu et al., 2017). In line with this, reviews have focused on patients' experiences of living with symptoms of breathlessness and self-

management of COPD (Clari et al., 2017). However, there remains a need to specifically examine patients' preferences in relation to support for self-management of asthma and COPD. Through aligning service delivery with patient preferences, patients may be more likely to engage with SMS which could enhance self-management outcomes.

In the past, the perspectives of people with chronic disease have been underrepresented in the development of chronic disease and SMS policies (Pumar-Méndez et al., 2017; Reidy et al., 2016), a finding also evident in the study presented in Chapter 2 of this thesis (O'Connell et al., 2018). In addition, few studies have sought patients' views on the desired outcomes of disease management (Boger et al., 2015). Research suggests that patients may place importance on characteristics of SMS that may not have previously been considered key aspects of interventions. For example, the development of positive relationships plays a key role in health interventions from the patient perspective though this may not have been emphasised as a key element in intervention design (Pearce et al., 2016; Sheridan et al., 2017; Sutcliffe et al., 2018). Patient-provider interactions which include in-depth communication and participatory decision-making may not only enable tailored SMS but may also have a direct positive impact on patients' social and emotional wellbeing (Arora et al., 2009; Street et al., 2009). Therefore, it is necessary to further understand patients' preferences in terms of SMS for asthma and COPD to ensure that support contributes to what matters to them in terms of managing their condition.

Values are typically used to describe underlying beliefs about what is desirable and are relatively stable across situations whereas preferences are guided by values but constructed in response to choices in specific contexts (Bastemeijer et al., 2017; Warren et al., 2011). Although, a taxonomy of patient values has recently been developed, further research is needed to understand the preferences for healthcare in specific contexts (Bastemeijer et al., 2017). In terms of developing supports for self-management, a review of the literature is needed to establish what is known about the preferences and perspectives of people with asthma/COPD, particularly pertaining to the support people want to help them to manage their illness. While a review of patients' perspectives on SMS has been carried out for other chronic conditions including rheumatoid, cancer or chronic kidney conditions (Dwarswaard et al., 2016), a synthesis of patients' preferences for SMS in COPD and asthma has not yet been conducted. To this end, this paper aims to explore what is known from previous research regarding the SMS preferences of people with asthma and/or COPD through synthesising findings from qualitative studies.

4.2 Methods

Thematic synthesis was used to integrate qualitative findings based on the method of Thomas and Harden (2008). This is a structured method which is suited to the synthesis of less conceptually-rich studies and reviews which seek to understand practical options in policy or intervention development (Noyes & Lewin, 2011; Snilstveit et al., 2012). The Enhancing Transparency in Reporting Synthesis of Qualitative Research (ENTREQ) statement (Tong et al., 2012) was used as a checklist to ensure transparency in the reporting of

the research (Appendix 4.A). The review was registered with PROSPERO (CRD42018100810) (O'Connell et al., 2018).

4.2.1 Search Process

Six databases were used with the aim of comprehensively identifying studies with perspectives on physical, social and emotional support. The databases included Applied Social Sciences Index and Abstracts (ASSIA), CINAHL, MEDLINE, PsycINFO, Psychology and the Behavioural Sciences, and Social Science Citation Index (SSCI). The core search strategy is detailed in Table 4.1. This was adapted to meet specifications of the different databases. The search was carried out in June 2018 and limited to articles published between May 2008 and April 2018 given the recent growth of this research area with the majority of studies in reviews of self-management of asthma and COPD published in the recent 10-year period (Miles et al., 2017; Russell et al., 2018). The search was also limited to articles available in the English language. Covidence software was used to detect duplicate studies and to screen articles. The reference lists of included papers were screened to identify additional potentially eligible articles.

Table 4.1. Key search terms and core search blocks for academic databases

Key term	Search block
Asthma or COPD	"Asthma" OR "Pulmonary Disease, Chronic Obstructive" [subject headings] OR Asthma* OR (chronic NEAR (pulmonary OR lung OR airway* OR respiratory)) OR "chronic obstructive pulmonary disease" OR COPD [title/abstract]
Preference	Prefer* OR value* OR choice* OR expect* OR attitude* OR acceptab* OR participation OR perspective* OR perce* OR view* OR priorit* [title/abstract]
Self-management	"self-care" [subject headings] OR self-manag* OR self-car* OR (self NEAR manag*) OR (self NEAR car*) [title/abstract]
Support	Support* [all text]

4.2.2 Inclusion Criteria

Studies eligible for inclusion were primary qualitative studies that explored the SMS preferences of adults (aged 18 and over) with asthma and/or COPD. Studies evaluating SMS interventions were included if participants' preferences for SMS could be gleaned e.g. telehealth interventions such as online consultations and tele-monitoring, pulmonary rehabilitation programmes, engagement with local health workers. Studies involving other stakeholders' perspectives, other chronic conditions or mixed methods were included if there was a clearly discernible section on qualitative findings from the perspectives of people living with asthma and/or COPD. Studies with participants receiving palliative or end-of-life care or residing in long-term care were not included. Reviews, conference abstracts, discussion pieces, protocols, dissertations and theses were excluded.

4.2.3 Study Selection and Data Extraction

Two reviewers independently screened the title and abstract and later screened full text for inclusion (SOC and VMcC). Disagreements were resolved by a third reviewer (ES). The data were extracted by one reviewer (SOC) and cross-checked by a second reviewer (VMcC). A standardised, piloted form was used to extract study detail under the following headings: author, publication year, country; aim and/or research questions; operationalisation of SMS/intervention; participants, method of data collection; method of data analysis; findings/results.

4.2.4 Quality Appraisal

Quality appraisal was carried out using the Joanna Briggs Institute (JBI) Checklist for Qualitative Research (The Joanna Briggs Institute, 2017) in order to capture congruency between methodology and methods, the congruency between analysis and conclusions, and the confirmability and credibility of the findings. Quality appraisal was carried out independently by two reviewers (SOC and ES) and consensus was reached through discussion.

4.2.5 Data Synthesis

A thematic synthesis of the data was carried out based on the guidance of Thomas and Harden (2008). Data encompassed portions of the text labelled as ‘findings’ or ‘results’ within the main body of the studies. The synthesis was carried out by one researcher (SOC) using NVivo (NVivo, 2012). The process initially involved line-by-line coding. Text under given codes were checked to establish consistency and additional codes/levels were added as necessary. Similarities and differences between codes were used to inform a hierarchical structure to organise descriptive themes. Descriptive themes were developed inductively and were considered in light of the research questions in order to develop analytical themes. A description of themes and supportive data was written up by the researcher and reviewed by supervisors (ES, VMC) and discussed in order to refine themes. Quotations from original studies were included to illustrate themes.

4.3 Results

A total of 968 articles were retrieved from the database search (Figure 4.1). After removal of duplicates, 594 articles were screened and 15 articles were included in the synthesis.

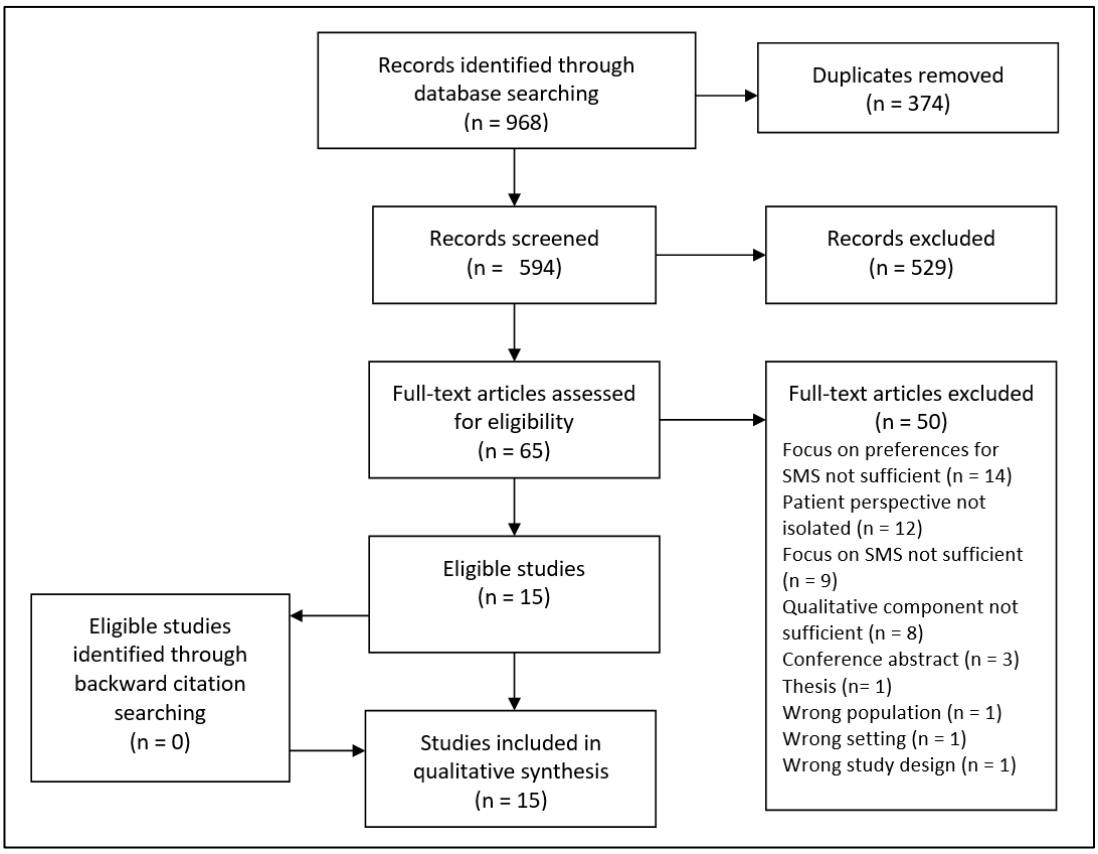


Figure 4.1. Article retrieval, screening and selection

Most studies were assessed to be of moderate to high quality (Table 4.2). One study (Patel et al., 2016) was missing information on a number of criteria for quality appraisal but given that its removal did not alter the structure of findings, it was included. Information on the philosophical perspective guiding the study and a statement locating the researcher culturally or theoretically were frequently absent.

Table 4.2 Quality appraisal of studies using JBI Checklist for Qualitative Research

	Apps et al.	Barken et al.	Burkow et al.	Cheong et al.	Gorst et al.	Halding et al.	Jackson et al.	Kielmann et al.	Langer et al.	Mathar et al.	Patel et al.	Pourlesami et al.	Vatnoy et al.	Wang et al.	Williams et al.
Congruity between stated philosophical perspective and research methodology	?	+	?	?	?	+	+	?	?	?	?	?	?	?	?
Congruity between research methodology and research question or objectives.	+	+	+	+	+	+	+	+	+	+	?	+	+	+	+
Congruity between research methodology and methods used to collect data.	+	+	+	+	+	+	+	+	+	+	?	+	+	+	+
Congruity between research methodology and representation/analysis of data.	+	+	+	+	+	+	+	+	+	+	?	+	+	+	+
Congruity between research methodology and interpretation of results.	+	+	+	+	+	+	+	+	+	+	?	+	+	+	+
Statement locating the researcher culturally or theoretically.	-	-	-	+	-	-	+	-	-	-	+	-	-	-	-
Influence of the researcher on the research, and vice-versa, is addressed.	-	+	-	+	-	+	-	+	+	-	+	-	-	-	+
Participants, and their voices, are adequately represented.	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Ethical according to current criteria or, evidence of ethical approval	+	+	+	+	?	+	+	+	+	+	+	+	+	+	+
Conclusions flow from the analysis, or interpretation, of the data.	+	+	+	+	+	+	+	+	+	+	-	+	+	+	+

Note: Yes = +, No = -, and Unclear =?

Studies provided information in relation to the SMS preferences of people with COPD (13 studies) and asthma (1 study) while an additional study included participants with one or both conditions (see Table 4.3 for summary of included studies). Three themes were identified which captured preferences for SMS from the perspectives of people with asthma or COPD. The first theme, *Types of Support,* describes the range of supports valued by participants in the studies, including their preference for tailoring support content to the individual. The next theme describes the *Support Relationship* which was seen as the foundation for providing SMS and person-centred care. The final theme of *Accessibility* identifies the importance of considering practical access to support from the patient's perspective. A sample of the coding tree is included in Appendix 4.B.

Table 4.3. Summary of included papers including original study themes

Author, year, Country	Relevant aim/ research question	Operationalisation of SMS/ intervention	Participants	Data collection and Analysis	Themes: subthemes/categories (as reported in studies)
Apps et al., 2017, United Kingdom	'to understand experiences of participation in a supported self-management programme for chronic obstructive pulmonary disease (COPD)' (p.1).	SPACE Intervention: unsupervised programme; manual for patient to follow independently; targeting activity, coping and education. 1-to-1 induction with HCP, call from HCP at 2 and 4 weeks and option to call HCP outside this.	*n* = 36 Age: mean 70.28 19 male Condition: COPD	Semi-structured interviews at 6 wk and 6 mth post intervention, 4 participants at both time points. Inductive thematic analysis	• Perceptions of the Programme • Lifestyle changes • Social support • Disrupting factors and barriers to maintaining routines
Barken et al., 2018, Norway	'To describe the lived experiences of quality of life among a group of patients living with COPD who were included in a telemedical intervention after hospitalisation for disease exacerbation' p.134	Technology for health monitoring and subjective symptom recording following hospitalisation. Nurse monitoring video communication. 30-day duration, follow-up and opportunity to contact nurse for advice over following two months.	*n* = 10 Age: 61-80 (mean 72) 7 male Condition: COPD	Interviews consisting of two open questions and probing. Descriptive phenomenological approach	• Living with chronic obstructive pulmonary disease: physical and mental limitations • Receiving care through telemedicine: accessibility, support, clinical insight and mutual clinical language • The dual chore of receiving care through telemedicine
Burkow et al., 2015, Norway	To assess 'patient acceptability of the delivery mode and components of a comprehensive pulmonary rehabilitation programme for any stage of the disease, as well as the technology usability' p.766	Home-based 9-week PR programme: daily digital health diary, pulse oximetry measure; weekly hour-long online educational sessions by professionals from different disciplines, 2 online exercise sessions, online consultation with nurse/physio.	*n* = 10 Age: mean 61.7 5 male Condition: COPD	Telephone, semi-structured interviews with open and closed questions. Descriptive Interpretation	• Acceptability: general approval; group education; supervised group exercising, exercise video and step-counting; Individual consultations and the digital health diary; comprehensive programme; supportive social environment; programme organisation; intrusiveness. • Technological usability
Cheong et al., 2015, Australia	To explore '(i) how and why patients select sources of health services, information and support, that is, their health connections and (ii) the key elements contributing to the nature and development of patients' health connections' p.2597	Exploring multidisciplinary care where multiple health workers from different disciplines collaborate with patients and carers to support Asthma management.	*n* = 47 Age: 19-75 (mean 44), 16 male Condition: asthma	Mapping of personal network before semi-structured interviews, face-to-face/phone using an interview guide. Deductive and inductive coding	Health Networks • Professional Health Connections • Personal Health connections: Symptom monitoring; emotional and physical support; information • Impersonal health connections Key elements of asthma networks: • Perceptions of the role of HCPs • Trust in relationships • Convenience of health advice • Perceptions of asthma
Gorst et al., 2015, United Kingdom	To explore COPD 'patients' beliefs and perceptions regarding home telehealth among current users of telehealth who are not enrolled in a trial' p.2	Telehealth equipment which allowed reading of vital signs or additionally for some to view data. Readings monitored shortly after by respiratory or community nurse	*n* = 8 Age 58-84 (mean 68) 3 male Condition: COPD	Semi-structured interviews after using telehealth for 6-36months. Interpretive phenomenological analysis	• Benefits of "being watched over" as providing peace of mind • Learning about the health condition and the impacts on self-management behaviour • Active engagement in health service provision and better access to healthcare • Valuing the importance of in-person care

Author, year, Country	Relevant aim/ research question	Operationalisation of SMS/ intervention	Participants	Data collection and Analysis	Themes: subthemes/categories (as reported in studies)
Halding et al., 2010, Norway	'To interpret and contextualise social relationship experiences in PR groups' p.1273	PR 12 week course (1 day per week) facilitated by MDT involving individual baseline and endpoint consultations, weekly group education sessions, open group discussion, group exercise sessions.	*n* = 18 Age: 52-81 13 male Condition: COPD	Three interviews per participant, 2 shortly after course and one within a year. Interview guides used. Qualitative content analysis	• Belonging through cheerfulness and informal settings. • Belonging through dialogue, shared understanding and fellowship. • Challenges to belonging. • Belonging through professional care and competence.
Jackson et al., 2012, Canada	To describe the patient journey of four people with COPD through the health system following hospitalisation for exacerbation	Any support provided by the healthcare system to aid management of COPD.	*n* = 4 Age 57-81 1 male Condition: COPD	Three interviews per participant (follow-up interview at 6 months with 3 participants); log of contact with health system. Thematic analysis	• Social support: physical support; informational support; emotional support; instrumental support. • System navigation: Effective Patient-Provider Communication; Effective Provider-Provider Communication; Previous Experience with the Health Care System. • Access: Physical access; Health Human Resource Access
Kielmann et al., 2010, United Kingdom	'Respiratory patients' perceptions of 'self-care', and how communication and access to professionals shaped their role in the management of their illness'p.56	Policy changes which propose patients are active partners who share responsibility for their care with health professionals	*n* = 31 Age: 40-89 20 male Condition: asthma and/or COPD	Illness diaries; two individual semi-structured telephone interviews; and focus groups. Framework approach	• Patient Self-care: advantages of self-care; disadvantages of self-care • The relationship between patients and professionals: flexible access to advice and reassurance; communication • Negotiating professional care
Langer et al., 2014, United Kingdom	'To use the accounts of patients who experienced the intervention to characterise its main features' p.2	Up to four 1-to-1 sessions with Liaison health worker in patient's home, at primary care practice or telephone. Emphasis on patient-centeredness, psychological interventions, problem solving and signposting.	*n* = 29 Age: 48–81 (mean 65) 13 male Condition: COPD	Semi-structured interviews. Inductive analysis with Constant comparative approach	• Patients' experiences: a new, motivational intervention • Listening to me • Relating to me • Investing in me • Deviant cases: not for me
Mathar et al., 2015, Denmark	'What are the experiences and preferences of COPD patients in relation to discharge from hospital with televideo consultations?' p.205	Eight 30-min televideo consultations over 2 weeks by community nurse and physiotherapist, monitoring of physical measures and verbal discussion of symptoms	*n* = 6 Age: 67-83 3 male Condition: COPD	Interviews, open exploratory questions. Systematic text-condensation method	• Control/loss of control • Security/obligations
Patel et al., 2016, United Kingdom	'to understand whether a novel system based on mhealthcare supported by specialist clinical input could improve patients' self-management' p.570	Patients completed an electronic diary with specific questions about health. Diary data monitored by the research team and could trigger contact from nurse. For exacerbation, they could contact a named nurse for a home visit	*n* = 10 Age 48-77 7 male Condition: COPD	Focus groups, Framework Method of thematic analysis	• Technology • Humanity • Interactions with the GP • All alone

Author, year, Country	Relevant aim/ research question	Operationalisation of SMS/ intervention	Participants	Data collection and Analysis	Themes: subthemes/categories (as reported in studies)
Pourlesami et al., 2017, Canada	'To investigate the feasibility and acceptability of incorporating peer supporters to PR programme; and to explore the key functions of peer support that will motivate COPD self-management in terms of sustaining long term benefits post-PR' p.58	Pulmonary rehabilitation with the addition of peer supporters	*n* = 28 15 male Condition: COPD	Focus groups and individual interviews. Inductive thematic analysis	• Peer support: Peer supporter & self-management of COPD, peer supporter selection; different methods of contacting and communicating with participants • Exercise: benefits of pulmonary rehabilitation from participants' perspectives; most helpful/challenging; confidence outside the programme; maintenance
Vatnoy et al 2017), Norway	To investigate 'how the patients experienced follow-up using a TM intervention, and the extent to which it supported and improved coping resources and independence' p.127	Technology to measure pulse and oxygen saturation, capture of subjective symptoms and symptom evaluation. Data transmitted to nurse and assessed. Daily video consultation with nurse between 10-21 days.	*n* = 10 Age: 55-83 7 male Condition: COPD	Semi-structured interview. Qualitative content analysis	• The TM solution is experienced as comprehensible and manageable and provides meaning in daily life: Handling and understanding the technology; Feelings related to technology use • The TM intervention contributes to stress reduction caused by illness burden and facilitates living as normally as possible: Confidence and trust in the health services; Impact on independence and self-management; Integrity and meaning in life.
Wang et al. 2012., Norway	'To explore COPD patients' experiences of a limited early discharge' p.2 of a hospital-at-home treatment programme	Home visit by nurse one hour daily for 3 days after discharge and took clinical measures. Offer of three outpatient consultations over following year	*n* = 9 Age: 51-79 4 male Condition: COPD (6 received intervention, 3 inpatients)	Semi-structured interviews using interview guides; 7-10 weeks after discharge Systematic text condensing	• Feeling safe • Individually adapted information • Managing strategies
Williams et al., 2014, United Kingdom	'to explore patients' expectations and experiences of using a mobile (mHealth) application to support self-management of COPD' p.392	mHealth intervention: technology based symptom diary, self-monitoring of pulse oximetry which were monitored by nurse (not daily). SM and educational multimedia resources also provided.	*n* = 19 Age: 50-85 (mean 67) 11 male Condition: COPD	2 individual interviews prior to intervention and 6 months following intervention. Grounded theory and constant comparative approach	• From 'uncertainty of ability to use technology' to 'ease of use'. • From 'disruption of care' to 'reassurance and perceived continuity of care'. • Increased awareness of variability of symptoms. • Supporting self-management behaviour.

4.3.1 Types of Support

4.3.1.1 Education through competent healthcare professionals

Participants valued education on asthma and COPD management (Apps et al., 2017; Barken et al., 2018; Cheong et al., 2015; Gorst et al., 2016; Kielmann, Huby, et al., 2010; Vatnoy et al., 2017; Wang et al., 2012). Increased knowledge provided confidence in management (Apps et al., 2017), allowed for communication with health professionals (Kielmann, Huby, et al., 2010), reduced fear of deterioration (Wang et al., 2012), and prevented hospitalisation (Vatnoy et al., 2017). Their preference for the type of information sought revolved around better understanding of one's health status and symptoms (Gorst et al., 2016; Kielmann, Huby, et al., 2010), information to deal with exacerbations and new challenges (Apps et al., 2017; Kielmann, Huby, et al., 2010), information on additional services available and new ways of managing their disease (Cheong et al., 2015; Kielmann, Huby, et al., 2010).

> The more you know, the safer you feel. You are not so frightened when you know what is what and get a proper explanation of this [disease] (Wang et al., 2012, p.3)

Self-monitoring through technologies was felt to provide greater insight into one's condition based on clinical insight over time and further education on symptoms and awareness of changes in health status (Barken et al., 2018; Gorst et al., 2016; Patel et al., 2016; Williams et al., 2014). One participant described the insight provided by self-monitoring as part of telemedicine intervention:

> Well, to get an answer yourself, to feel safer, and try to think back; what have I done now since the heart rate is so high? Also, that when it

> started to go down to normal. . . that was very reassuring, in that respect (Barken et al., 2018, p.138)

Participants preferred education provided by HCPs over other sources, with HCPs seen as competent sources of information. This competence in knowledge and advice from HCPs provided 'peace of mind' when experiencing health concerns (Apps et al., 2017; Barken et al., 2018; Burkow et al., 2015; Cheong et al., 2015; Halding et al., 2010; Kielmann, Huby, et al., 2010; Mathar et al., 2015; Patel et al., 2016; Vatnoy et al., 2017). Some studies highlighted frustrations among participants of feeling left alone to manage their own condition. There were clear indications that some participants did not feel comfortable managing alone and felt that this was beyond their capacity (Kielmann, Huby, et al., 2010; Patel et al., 2016). The support of the HCPs was particularly desired in the case of medical treatment, medications, physical examinations and tests. While various HCPs were felt to provide competent advice, in three studies, participants referred more regularly to doctors and specialists for medication and medical treatment issues (Cheong et al., 2015; Halding et al., 2010; Patel et al., 2016).

> It's not just about learning to live with COPD, learning to exercise and so on. In the PR [pulmonary rehabilitation] course, I benefited from the medical treatment, supervision and advices of experts (Halding et al., 2010, p.1276)

4.3.1.2 Psychological Support

Psychological support encompassed emotional support as well as motivational support to make changes in health behaviours. Emotional support was seen as important by people with asthma and COPD (Cheong et al., 2015; Langer

et al., 2014; Poureslami et al., 2017). They appreciated HCPs enquiring about their psychological wellbeing (Barken et al., 2018; Halding et al., 2010).

> I think that is [connection with nurse] very nice, because they understood what I was talking about . . . on everything! If I said that: oh! Today I experience breathlessness! Yes, what have you done? Then the TM nurses accept you, right, and then they ask, how do you feel? and that kind of thing. It is a completely different way to connect and I get positive effects to talk to someone like that (Barken et al., 2018, p.137)

Participants were motivated to continue with behaviour changes when they believed that HCPs acknowledged their attempts to change and commented on their progress (Apps et al., 2017; Burkow et al., 2015; Langer et al., 2014; Poureslami et al., 2017). In a study by Langer and colleagues, one participant noted of their local healthcare worker "I felt she was investing in me … and I didn't want to let her down", which was different than with his GP where "I just feel I'm one of another list of people" (Langer et al., 2014, p.5)

4.3.1.3 Person-centred Support

The studies highlighted that participants valued support which was tailored to their needs, that is, person-centred such as education and advice being tailored to the individual (Halding et al., 2010; Jackson et al., 2012; Langer et al., 2014; Wang et al., 2012). In two studies, participants reported not gaining from education when they had encountered similar information in previous healthcare encounters (Mathar et al., 2015; Wang et al., 2012), indicating that their preference was that information be tailored to their changing needs over time. In addition to the range of educational needs reported, some participants

had a particular preference for instrumental support such as transportation and the management of finances and financial claims (Jackson et al., 2012; Langer et al., 2014). In one study, participants appreciated when these areas were addressed by healthcare workers, though more frequently this type of support was provided by friends and family (Jackson et al., 2012; Langer et al., 2014).

4.3.2 The Support Relationship

The relationship between the participant and HCPs was seen as critical to SMS and enabled person-centred care. It encompassed a number of closely related features. A personal connection with one's HCP was valued because it gave participants a feeling of "being known" and was felt to make it easier to navigate through the health system (Barken et al., 2018; Jackson et al., 2012).

4.3.2.1 Collaboration and trust

Positive interactions and experiences with a HCP gave rise to a ''working relationship'' and ''trust both ways'' (Kielmann, Huby, et al., 2010). Trust was seen as a very important component of the support relationship. In the context of a trusting relationship, participants felt comfortable to share personal experiences and were more willing to try out new and alternative forms of self-management and SMS suggested by HCPs and others (Cheong et al., 2015; Halding et al., 2010; Kielmann, Huby, et al., 2010). The 'working relationship' was often preferably described as collaboration or partnership. This relationship involved the HCP acknowledging the patient's expertise and giving them an opportunity to actively participate in their care (Barken et al., 2018; Kielmann, Huby, et al., 2010). Participants sought to input because they felt best placed to provide information on their state of health (Kielmann, Huby, et

al., 2010; Vatnoy et al., 2017). As one participant noted: “I know myself best; I am the one who feels when my health condition changes” (Vatnoy et al., 2017, p.129). Participants wanted their health concerns to be taken seriously, and expressed dissatisfaction when this was not the case (Barken et al., 2018; Kielmann, Huby, et al., 2010).

4.3.2.2 Opportunities for communication

The collaboration described above was facilitated by communication. Patients appreciated the expressed interest of HCPs in how they felt and being given an opportunity to voice this. Participants placed great importance on “being listened to” across the studies and desired the opportunity to provide self-evaluations to their HCP (Jackson et al., 2012; Vatnoy et al., 2017). Monitoring health data also provided a mutual clinical language which made participants feel they were being acknowledged and their concerns were being taken seriously (Barken et al., 2018; Gorst et al., 2016). In addition, the opportunity to ask questions was strongly appreciated (Burkow et al., 2015; Halding et al., 2010; Jackson et al., 2012; Wang et al., 2012). Their preference was for open communication between HCP and themselves which was seen to reduce cultural influences such as the traditional role of doctor as expert or the influence of social desirability (Halding et al., 2010; Kielmann, Huby, et al., 2010). Some studies highlighted the importance that participants attached to having sufficient time within healthcare appointments (Halding et al., 2010; Jackson et al., 2012; Patel et al., 2016; Wang et al., 2012). This was felt to allow time to talk about how the individual feels and an opportunity to ask questions.

4.3.2.3 Continuity of care

The trusting relationships underpinning SMS were seen to be the result of positive experiences over time. Continuity of care was important for participants (Cheong et al., 2015; Jackson et al., 2012; Kielmann, Huby, et al., 2010; Vatnoy et al., 2017). They valued familiarity of their HCP which also saved them having to repeat the entire history of their condition on each visit (Cheong et al., 2015; Jackson et al., 2012).

> 'The telenurse tells me ''you are looking better today'''; 'when the nurses at central are strangers, you have to get to know them; then it feels safer'; 'you don't have to repeat yourself' (Vatnoy et al., 2017, p.129).

With regard to patient education or monitoring programmes, participants were often reluctant to see the programmes end and they expressed a preference for some form of maintenance or follow-up as a continuity for patient education and support (Barken et al., 2018; Poureslami et al., 2017; Wang et al., 2012).

4.3.3 Accessibility

4.3.3.1 Prompt, physically accessible support

Accessibility frequently arose in the data with reference to timely access to services as well as physical accessibility. Challenges in attending appointments were highlighted due to living in remote areas or physical restrictions of COPD such as use of oxygen (Burkow et al., 2015; Jackson et al., 2012; Poureslami et al., 2017). Participants' preferences placed emphasis on the benefits of immediate or instant access to care at any time of day and felt frustrations with waiting lists or delays (Barken et al., 2018; Cheong et al., 2015; Kielmann, Huby, et al., 2010; Mathar et al., 2015; Patel et al., 2016;

Wang et al., 2012). This immediate access was felt to provide security and 'peace of mind' to participants and their families particularly because it was seen as preventing deterioration.

> It would be nice actually to be able to email somebody like her [nurse] as well and just sort of say. . .remind me of the procedure of coming off a high steroid and she would be able to (Kielmann, Huby, et al., 2010, p.58)

4.3.3.2 Balancing telehealth and face-to-face modes of delivery

SMS was provided through many different modes including face-to-face, televideo, telephone and email/text. Telehealth was largely appreciated for the flexibility it provided in accessing healthcare: it was felt to reduce the time and energy spent attending appointments, allowing greater time for other daily activities (Apps et al., 2017; Burkow et al., 2015; Gorst et al., 2016; Mathar et al., 2015; Poureslami et al., 2017; Vatnoy et al., 2017). However, participants preferences were for face-to-face support and in-person care, often in addition to telehealth, which provided patients with a sense that care was personal (Apps et al., 2017; Gorst et al., 2016; Patel et al., 2016). Their preference for face-to-face interactions was particularly notable for initial meetings with HCPs or peer support groups in order for them to become familiar with the programme or their supporter before engaging in telecommunication (Apps et al., 2017; Poureslami et al., 2017).

> No [telehealth is not as good as in-person care], it's a bit personal, I think. With face to face you can see how people react when you tell them things [laughs]. You've only got to look at somebody's eyes to see (Gorst et al., 2016, p.6).

Televideo was reported to feel like being in the same room and was preferred over telephone contact because it created a feeling of personal care (Barken et al., 2018; Vatnoy et al., 2017). However, it was necessary to ensure this worked well as when it was faulty, it was disappointing for participants:

> That was nice [using videoconference]. It is okay using phones and that sort of thing, but it is. . . it is a lot better! It is just like, yeah that. . . like as we sit now, it is much more contact than it would have been by using phone and other things (Barken et al., 2018, p.137).

4.3.3.3 Support formats

There were varying preferences regarding the format of support. Peer support through groups was described to be beneficial by some participants in terms of increasing a sense of belonging as well as opportunities to share with and learn from others with similar conditions (Burkow et al., 2015; Halding et al., 2010).

> We learned from each other too… It helps to hear that other people are going through the same thing. That they struggle with things… Yes, how they do it…how they handle it (Burkow et al., 2015, p.6).

Some individuals were not interested in peer support and felt that group situations were intimidating (Halding et al., 2010). Instead, their preference was for individual support, with HCP support highly valued in this regard by the majority of participants across studies (Apps et al., 2017; Barken et al., 2018; Burkow et al., 2015; Cheong et al., 2015; Halding et al., 2010; Kielmann, Huby, et al., 2010; Mathar et al., 2015; Patel et al., 2016; Vatnoy et al., 2017).

Even though there was a large preference for individual HCP support, there were individual differences in the frequency with which support from HCPs was

needed. The preference was largely for regular support with participants highlighting benefits of regular interaction with HCPs as well as monitoring technologies which facilitated frequent information exchange with HCPs. Advantages included increased awareness of symptoms and emotions (Barken et al., 2018), reassurance regarding health concerns (Kielmann, Huby, et al., 2010; Wang et al., 2012), and also early detection of problems and prevention of deterioration (Barken et al., 2018; Gorst et al., 2016).

> It was safe, because I knew she was coming! If I did not feel 100% well, I knew that she was coming tomorrow to check me (Wang et al., 2012, p.3)

Some participants disliked the idea of regularly attending their GP (Patel et al., 2016). One reason for delaying appointments with HCPs was that participants perceived HCPs to be busy and did not want to bother them (Kielmann, Huby, et al., 2010; Williams et al., 2014). A serious implication of this was that participants waited until symptoms became more severe before seeking the support of HCPs, as evident in three studies (Gorst et al., 2016; Vatnoy et al., 2017; Williams et al., 2014). Other participants spoke of preferring healthcare appointments to be scheduled on the basis of their needs (Wang et al., 2012). The benefit of regular appointments depended largely on the current severity of illness (Barken et al., 2018; Kielmann, Huby, et al., 2010; Vatnoy et al., 2017). Participants with asthma often sought support when symptoms were obvious (Cheong et al., 2015). While people with severe COPD preferred daily contact with HCPs, people with less severe symptoms found daily contact to be a hindrance (Barken et al., 2018). Some studies highlighted how fixed appointments were felt to be obligations which interrupted other daily routines

(Mathar et al., 2015; Vatnoy et al., 2017). As one participant noted: “I don’t want to be bound to talk to her every morning. I rather call her up if I feel bad” (Vatnoy et al., 2017, p.129).

4.4 Discussion

Fifteen qualitative studies included in this review identified preferences of people with COPD and/or asthma regarding support for self-managing their disease. A synthesis of the findings highlighted the importance of HCPs and the healthcare system in supporting self-management of COPD and asthma with patients reacting strongly when feeling isolated in the journey of self-management. This reinforces the need to ensure that self-management is not framed solely as an individual process but instead as a process through which a person is supported by the healthcare system (Richard & Shea, 2011). While reviews of self-management have identified the need for greater education on management (Miles et al., 2017; Russell et al., 2018), this review highlights the importance placed on information from competent HCPs. This preference reflects the value of professionalism in the taxonomy of patient values developed by Bastemeijer et al. (2017). This review extends understanding of how patient values can be operationalised in SMS in the context of people with asthma or COPD through exploring their preferences for support.

This review also reflected the value of accessibility from the patient’s perspective, supporting the emphasis of this in policy on SMS, as was noted in a document analysis of SMS frameworks reported in Chapter 2 (O’Connell et al., 2018). The synthesis findings mapped closely to the patient considerations around accessibility in the PRISMS taxonomy of SMS (Pearce

et al., 2016; Sheridan et al., 2017). People with asthma/COPD sought support which was physically accessible without delay, preferably through telephone or technology for those living far from support hubs. There were also individual differences in preferences for accessing support. Similar to previous work (Sheridan et al., 2017) some participants desired input into the scheduling of meetings with HCPs so that frequency of appointments could be tailored to individual's needs. In addition, participants had varying attitudes toward peer support, as reported in a recent exploratory qualitative study on SMS for rheumatic disorder (Been-Dahmen et al., 2017). This suggests that peer support may only have benefit if it fits with individual preferences of people with asthma or COPD.

People with asthma or COPD valued a collaborative relationship with their HCP which reflects findings in other studies from patients' perspectives (Been-Dahmen et al., 2017; Dwarswaard et al., 2016; Schulman-Green et al., 2016; Sheridan et al., 2017) as well as the value patients attach to partnership (Bastemeijer et al., 2017). Communication, trust and continuity were the foundations of a good relationship which in turn facilitated tailored and person-centred support. People with asthma/COPD particularly valued aspects of communication which have been found to affect self-management in other reviews including: open communication using a mutual language; opportunities to self-evaluate their condition; and opportunities to ask questions (Miles et al., 2017; Russell et al., 2018; Schulman-Green et al., 2016). Participants had a preference for HCPs to enquire about their wellbeing reflecting the value of compassion such that HCPs that are interested in the patient as a person and empathise with them (Bastemeijer et al., 2017). The

concept of epistemic injustice (Fricker, 2011) has been invoked to describe patients' experiences of clinical responses to breathlessness (Hutchinson et al., 2018) and is echoed in these findings. This is where a speaker's testimony is given reduced credibility due to preconceptions or lack of shared understanding which could have a negative psychological impact on the speaker (Fricker, 2011). This may help to explain patients' desires to contribute their experiences in the clinical encounter. This reinforces the message that some aspects of the patient-provider interaction may not only contribute to appropriate clinical actions in managing asthma/COPD but also may have important benefits for the overall wellbeing of the person (Arora et al., 2009; Street et al., 2009).

Some components of SMS which feature strongly in the literature such as action planning and goal setting (Pinnock et al., 2017; Zwerink et al., 2014) were not clearly prioritised as preferences by people with asthma or COPD in this review. Some people with asthma and HCPs have been found to have negative views of actions plans (Miles et al., 2017). While participants did appreciate motivation to continue behaviour changes as in other studies (Been-Dahmen et al., 2017; Dwarswaard et al., 2016; van Hooft et al., 2017), these were often through HCPs commenting on and valuing progress. This supports the idea indicated in previous research (Sheridan et al., 2017; Sutcliffe et al., 2018) that patients may value aspects of interventions which are not often framed as the critical intervention components. While interventions such as action plans and goal setting are key features of SMS in asthma and COPD (Pinnock et al., 2017; Zwerink et al., 2014), further research

to understand patients' preferences may help provide clearer insight into the characteristics of SMS that help patients to engage with these strategies.

4.4.1 Strengths and limitations

This review synthesised qualitative studies on SMS preferences of people with asthma or COPD, which has not been previously conducted to the best of my knowledge. The review and synthesis provide a basis for future research which seeks to understand how to provide SMS for asthma and for COPD which is aligned with patient preferences. However, the studies in this review gathered data primarily from people with COPD while few related to asthma. Additionally, the extent of multimorbidity among these participants was not clear. It would be important to examine if similar preferences hold in the context of multimorbidity given its prevalence and evidence that having multiple chronic conditions creates additional challenges for self-management (Liddy et al., 2014).

Quality appraisal results were not used in the synthesis to prioritise findings. Most studies were of moderate to high quality and a study whose quality was ambiguous was included because it did not alter the synthesis. It should be noted that the synthesis does not draw directly on raw data but on findings which involve the interpretations of the primary researchers. While the appraisal suggested that participant's voices were adequately represented, in some studies there was a lack of reporting of the philosophical perspective or cultural/theoretical location of researcher within the studies. This information should be included in future studies to enable quality appraisal.

4.4.2 Clinical Implications and Future Research

This review provides a basis for understanding the strategies which need to be prioritised from a healthcare delivery perspective to provide SMS which aligns with patients' preferences. However, the varying types of support preferred by different people and contexts highlights that SMS is not a 'one size fits all' approach and that it needs to be tailored to the individual (Dwarswaard et al., 2016; Trappenburg et al., 2013). In relation to preferences for personalised scheduling of support, it will be important to understand how these preferences can be incorporated safely, so that findings of delays in seeking healthcare and associated negative outcomes, reported in this review, can be reduced.

The review reinforces that communication skills are fundamental to developing an interpersonal relationship between the individual and their HCP to facilitate SMS and need to be a focus of training for HCPs. In addition, the preference for seeing the same HCP, identified elsewhere (Been-Dahmen et al., 2017), also supports the need for the healthcare systems to facilitate continuity of care.

Most of the included studies were conducted as part of an evaluation of SMS interventions. Additional information could be gained from open exploratory research on SMS preferences of people with COPD/asthma which is not limited to evaluating interventions. In addition, further studies could help explore research gaps such as whether there are preferences for the type of health professionals patients would like to be supported by (Dwarswaard et al., 2016) as well as other contextual factors which could influence SMS

preferences. Further research also needs to establish the value placed on face-to-face contact and how the preferences for accessibility of SMS and in-person care can be optimally balanced.

4.4.3 Conclusions

This review has identified what is currently known about the SMS preferences of people with asthma or COPD based on qualitative research with these populations. The thematic synthesis identified attributes of SMS that are preferred by people with COPD or asthma as well as gaps in research which can inform future studies. There is a strong preference among this group for increased education provided by competent HCPs. A collaborative relationship between the individual and their HCP was central to SMS and was characterised by communication, trust and continuity over time. Improving access to SMS was also found to be a critical issue from the perspective of participants in the studies, however the optimal means of enhancing accessibility warrants further research. Understanding the preferences of people living with chronic disease holds promise to shed light on the attributes of SMS which may help people to engage with SMS and facilitate better self-management. Further qualitative studies which openly explore self-management preferences of adults with asthma/COPD can help to address current research gaps. To this end, a qualitative study exploring the SMS preferences of people with asthma or COPD in Ireland is reported in Chapter 5.

Chapter 5. The preferences of people with asthma or COPD for self-management support in Ireland: a qualitative descriptive study

This chapter describes a qualitative descriptive study of the support preferences of 20 people with asthma and/or COPD in Ireland. Given that many of the studies reviewed in Chapter 4 were evaluations of interventions, this study contributes to the literature in terms of purposively exploring self-management support (SMS) preferences. Primary qualitative research is also key to identifying the SMS attributes directly relevant to the population as well as the acceptable levels of attributes and terminology in order to enhance the clarity of the discrete choice experiment (Bridges et al., 2011; Coast et al., 2012; Mangham et al., 2009). This chapter will be submitted as a paper for publication to the Journal of Clinical Nursing.

Abstract

Self-management support interventions have had beneficial outcomes for people with asthma and people with COPD, though challenges remain in their implementation. Increased understanding of the support preferences of people with asthma/COPD can help inform the development of future interventions to meet patients' needs. A qualitative descriptive study was conducted to explore preferences using semi-structured focus group and individual interviews with 20 participants who had asthma and/or COPD in Ireland. Qualitative content analysis was used. Three themes were identified: support accessibility – access to routine and unscheduled support from healthcare professionals with specialist knowledge; consultation content – comprehensive and person-

centred content; and the person-provider relationship: patient input and continuity over time. Routine support for people with asthma/COPD needs to be comprehensive in addressing the individual patient's challenges. During exacerbations, access to timely advice is a priority and preferably provided by a healthcare professional who knows the patient's medical history and takes the patient's concerns seriously.

5.1 Introduction

Globally, chronic respiratory diseases such as asthma and COPD adversely affect quality of life and account for a large proportion of hospitalisations and mortalities (Ampon et al., 2005; Miravitlles & Ribera, 2017; World Health Organisation, 2019). Supporting people with chronic disease in the process of self-management is a key strategy to reduce this burden (Bodenheimer et al., 2002). Self-management involves individuals, living with a chronic disease, actively managing physical, social and emotional wellbeing within the context of their daily lives (Miller et al., 2015). Self-management support (SMS) is considered effective as a multi-level approach to facilitating self-management that involves a healthcare professional (HCP) and patient partnership at the clinical level that in turn is supported by resources, training and systems at the organisational level (Kawi, 2012). Interventions to enhance self-management for both asthma and COPD have had positive outcomes including improved health-related quality of life, symptom reduction as well as reduced hospitalisation rates (Lenferink et al., 2017; Pinnock et al., 2017; Zwerink et al., 2014).

However, there remain challenges in delivering SMS as part of routine care which may curtail the benefit of these interventions at the population level (Entwistle, Cribb, & Owens, 2018; Grady & Gough, 2014). The reach of interventions and extent to which patients engage with recommended self-management practices are two such challenges. Interventions such as Pulmonary Rehabilitation programmes, commonly delivered in group sessions to people with COPD, have been found to reach limited numbers of the target population (Bolton et al., 2013; Jones et al., 2014). A qualitative synthesis found that participation in COPD support programmes was influenced by many factors including patient perceptions that the programme would be unhelpful; unsuitable; and challenges in accessing the programme (Sohanpal et al., 2015). There are also SMS interventions in which many participants do not enact the recommended self-management behaviours. For example, a randomised controlled trial of an SMS intervention for people with COPD did not significantly reduce hospital admissions overall. However, an exploratory analysis found that participants who made appropriate treatment changes (42% participants classified as successful self-managers through reviewing diary records) had lower hospital admissions (Bucknall et al., 2012). This research suggests that there are challenges for people with chronic respiratory disease in engaging with recommended self-management behaviours and potential challenges in the acceptability of SMS.

The need for further critical discussion on what constitutes appropriate support for self-management has been raised (Entwistle, Cribb, & Owens, 2018). These researchers encourage a broader focus on understanding what matters to people living with chronic disease rather than on disease control. Eliciting

patients' perspectives and values may help to enhance the acceptability of interventions (Araújo-Soares et al., 2019). Qualitative and quantitative studies have offered insights into the preferences of people with asthma and COPD. Adults with asthma have differed in having preferences for active, collaborative or passive roles in treatment decision-making (Caress et al., 2005; Caress et al., 2002). The communication skills of the HCP and the patient-professional relationship were identified as facilitators of participation (Caress et al., 2005). A qualitative synthesis of studies on SMS from the perspectives of adults with asthma/COPD, as presented in the previous chapter, found that their preferences included having prompt and easy access to a HCP, a collaborative relationship with a HCP over time, and tailored support content including education and psychological support (O'Connell et al., 2019). However, few of the studies reviewed related to asthma and they mainly evaluated participants' experiences of specific interventions rather than purposively exploring preferences for SMS, prior to intervention development (O'Connell et al., 2019).

Further research is needed to explore the SMS components of greatest value from the perspectives of people with asthma/COPD. This may help enhance the acceptability of interventions and thus increase potential for implementation and positive impact. There is some commonality between Asthma and COPD in that management of symptoms needs to address shortness of breath, chest tightness, wheeze and cough (GINA, 2019; GOLD, 2019). However, the clinical profile of these chronic respiratory diseases also differs with COPD having a later onset and more persistent and progressive nature than asthma. While self-management has benefitted from both generic

and disease-specific approaches to research (van Houtum et al., 2015), the study of asthma and COPD together is aimed to shed light on common and disease specific preferences.

The aim of this study was to explore and describe the SMS preferences of people with asthma and/or COPD in Ireland. In addition to the literature reviewed in Chapter 4, this study will be used to inform the design of a discrete choice experiment to measure the relative preferences for features of support from the perspectives of people with asthma or COPD.

5.2 Methods

5.2.1 Study design

A qualitative descriptive design was used for this study (Bradshaw et al., 2017; Sandelowski, 2010). This design provides a rich description of participants' experiences of a phenomena in easily understood language (Bradshaw et al., 2017). Qualitative description takes a naturalistic perspective where reality is seen as the subjective experience of the participants as well as the interpretation of the researcher (Bradshaw et al., 2017). The Standards for Reporting Qualitative Research (SRQR; Appendix 5.A) were used to guide reporting (O'Brien et al., 2014). The study was approved by the Social Research Ethics Committee, University College Cork (Log 2018-195) and participants provided written informed consent prior to participation (see Appendix 5.B for ethical approval and Appendix 5.C and 5.D for focus group and individual interview information/consent sheets).

5.2.2 Participants and recruitment

Twenty adults participated of whom nine had a self-reported diagnosis of asthma, ten had COPD and one had both asthma and COPD. The aim was to recruit a sample with maximum variation who represented people with characteristics that may influence perspectives on SMS such as severity of condition, length of diagnosis and rural/urban living (Patton, 2015). To achieve this, the sample was recruited through two national support organisations for asthma and COPD in Ireland and by using electronic and in-person strategies. Methods of recruitment through the national organisations involved sharing a study advertisement through social and electronic communication media as well as providing information in-person at sessions run by the organisations. Data collection was ceased once data saturation was achieved, that is, when no new codes were identified through additional interviews.

5.2.3 Data collection

Eight individual and two focus group interviews (range of three to nine participants per group) were conducted. Focus group interviews were used to generate discussion on a range of ideas relating to SMS and to understand convergent and divergent preferences across participants while individual interviews explored SMS preferences in greater depth in order to shed light on the individual contexts which shape preferences (Dilshad & Latif, 2013). Use of individual and group interviews also provided greater opportunities for participation for people who may have a preference for one of these methods (Lambert & Loiselle, 2008). As the PhD student, with a background in psychology, I conducted all interviews and was not previously known to participants. For focus groups, I moderated the discussion while a second

researcher took notes on the discussion and non-verbal reactions. Individual interviews lasted 20-40 minutes while focus groups interviews lasted 50-80 minutes. Participants took part in either an individual or a focus group interview.

A topic guide (Table 5.1) with open-ended questions based on the synthesis of international literature (O'Connell et al., 2019) presented in the previous chapter, was used with flexibility to probe further into participants' responses (Interview questions included in Appendix 5.E). The topic guide was piloted with the first two participants whose data were included in the analysis. Data collection and analysis were carried out concurrently. An iterative approach was used akin to previous qualitative research for developing a discrete choice experiment (Coast et al., 2012; Coast & Horrocks, 2007). The final focus group interview and last three individual interviews additionally sought participants' perspectives on features that had arisen from analysis of previous interviews as well as using the topic guide.

Table 5.1. Topic guide for semi-structured interviews and focus groups

Experience of managing chronic illness
Support received in managing illness
Support desired to manage illness
Most important elements of support
Ways in which health service can improve support
Nature of support needs
Nature of support provider
Nature of support relationship
Support accessibility

Demographic information was collected before beginning the interview/focus group for the purpose of describing the sample and examining contextual influences on preferences. Asthma control was measured using the Asthma Control Test™ (ACT) which includes five self-report items on asthma symptoms, impact on everyday functioning and use of rescue medication (Nathan et al., 2004). The ACT has a range of 5-25 with higher scores indicating better control. The COPD Assessment Test™ (CAT) was used to measure the impact of COPD on participants' wellbeing and daily life through eight self-reported items (Jones et al., 2009). The CAT has a range of 0-40 with higher scores indicating high impact of COPD.

5.2.4 Data Analysis

The interviews and focus groups were audio-recorded and transcribed. Participants were anonymised through pseudo names. Data were analysed using qualitative content analysis (Bengtsson, 2016). This process began with data familiarisation followed by inductive coding based on the manifest content of the data. The transcripts were re-read to ensure that data pertaining to the aim had been coded (Bengtsson, 2016). Codes were grouped into categories and subsequently categories were further grouped into themes. Negative or deviant cases were examined, and participant demographics were compared with codes to assess if individual factors influenced preference patterns. Coding was primarily carried out by this PhD researcher (S0C) and a sample of interviews was cross-checked by PhD supervisors (VMC and ES). NVivo software, Version 11 (QSR International Pty Ltd. 2015) was used to support data management and an audit trail was kept of the analysis process. The analysis was written up by this researcher and reviewed by supervisors and

contributing authors. Descriptive statistics regarding the sample characteristics were calculated using SPSS Version 24 (IBM Corp, 2016, Armonk, NY, USA).

5.2.5 Trustworthiness

An audit trail of the research process contributed to dependability which included a record of the analysis on NVivo. The PhD researcher sought to develop rapport with participants through information exchange prior to interview and expressing empathy during the interview (Bradshaw et al., 2017). Reflexivity was developed through checking of coding and review of the findings by supervisors; notes taken by the PhD researcher immediately after individual interview and during analysis; and notes taken by a second researcher during focus group interviews (Probst & Berenson, 2013). The purposeful maximum variation sampling approach and description of the sample aimed to inform the transferability of findings (Bradshaw et al., 2017). Direct quotations strengthen the confirmability of the research (Bradshaw et al., 2017).

5.3 Results

Demographic characteristics of the participants are presented in Table 5.2. Asthma control ranged from poorly controlled through to well-controlled while COPD impact ranged from low to very high.

Three themes were identified from the data: support accessibility, consultation content and person-provider relationship (see Appendix 5.F for sample coding tree). Predominantly, there was congruence in the coding and themes across

participants. However, some differences were identified across asthma and COPD, according to symptom severity and for those with multimorbidity.

Table 5.2 Demographic characteristics of participants, mean (SD) or n (%)

Characteristic	n = 20
Age, years	57.26 (14.81)
Gender,	
Male	2 (10%)
Female	18 (90%)
Living Arrangements	
Living alone	3 (15%)
Living with a partner/family	16 (80%)
Living in shared accommodation	1 (5%)
Length of asthma diagnosis (years)	25.38 (13.31)
Length of COPD diagnosis (years)	9.39 (8.28)
Asthma control, mean ACT score	17.50 (4.88)
Impact of COPD, mean CAT score	22.45 (6.46)
Presence of other chronic illness	12 (60%)

5.3.1 Support accessibility

5.3.1.1 Accessing support in acute phases of illness

Participants' accounts highlighted the importance of quick and timely access to services when experiencing exacerbations of their condition, that is, changes in their symptoms ranging from mild to severe. When experiencing a change in symptoms that required additional support, participants preferred to consult with a HCP within one or two days. As a first point of contact, for many participants, there was a preference for being able to phone a HCP. Some participants were happy with text or email to check with a HCP regarding self-managed treatment changes in response to symptoms. Participant accounts indicated a preference for having a "second opinion"/consultation regarding

what actions to take as well as support in reducing panic experienced in relation to breathing difficulties, often mentioning an ‘asthma nurse’ or ‘respiratory nurse’ for this role:

> No she’ll just ring you and she’ll know. She’ll ask you a few questions, but I think especially if you’re really out of breath, she’ll get you to kind of calm down a bit and she’d be able to work from there and suggest whether you’d need to go into A&E (Maureen).

Some participants described delaying making contact with their HCP due to fears of being hospitalised; perceptions that HCPs are busy; that they would be wasting HCP time; or might be perceived to be dramatic by their HCP and others around them. The cost of accessing services was also a deterrent for participants. However, some noted that they had access to a medical card through which most healthcare services are provided free of charge in Ireland. This encouraged early help-seeking: “I found [medical card] an amazing help because I'm able to go down to my GP before I get ill… and she'll sort me out” (Lucy). Participants suggested that asthma and COPD should be recognised as part of the national long-term illnesses scheme so that medications are subsidised by government.

5.3.1.2 Accessing scheduled support

Participants’ accounts indicated that having regular scheduled reviews was important in order to identify changes and discuss management of their condition. In terms of frequency, participants believed that bi-annual reviews would be more appropriate than an annual review which had been previously experienced. A pattern in the data was that people who experienced greater severity of symptoms had a preference for more frequent reviews such as

every three months. A small number of participants suggested having a scheduled local service that was available close to home in their local town to avoid travelling long journeys and spending full days attending appointments. As commented by one rural-living participant, it would be good “to know that there is a specialist nurse who will meet up locally however many times a year as possible. To know that is there. I don’t have to travel very far” (Eleanor).

5.3.1.3 Accessing HCPs with specialist knowledge

The primary providers of support for participants in this study were general practitioners (GPs), specialist respiratory nurses and consultants. Other providers of support included practice nurses (based in GP surgeries), pharmacists, physiotherapists, peers and family members. While participants appreciated HCPs with both generalist and specialist remits, many participants valued their HCP having specialist knowledge of their condition. This could include HCPs with a formal respiratory specialist role as well as GPs who acquire additional knowledge relating to managing asthma or COPD. It was considered that GPs or practice nurses without specialist knowledge may not take concerns as seriously. Some participants valued support through the combined care of their GP and respiratory specialists. However, other participants noted barriers to accessing specialist HCPs including lack of awareness of services; lack of referral from primary to secondary care; as well as waiting lists for appointments to see specialists.

> So I felt like even if you could meet someone on a bit more of a specialist level, whether it was a nurse specialist or a consultant, someone to just talk about how I was coping and the fact that I wasn’t and what more could I do (Michelle).

5.3.2 Consultation content

5.3.2.1 Comprehensive person-centred support

Participants valued a comprehensive person-centred consultation about their asthma/COPD during which they had sufficient time with their HCP to address a number of important areas of management. Participants acknowledged the importance of reviewing and adjusting prescriptions. However, many participants spoke of their frustration when checking prescriptions was the main focus of the consultation without attention to managing other aspects of their health. The topics of side effects, constraints on daily life and the emotional impact were most commonly identified as important for discussion. Additionally, participants expanded upon individual challenges requiring personalised support such as managing comorbidities and managing their conditions in the context of exercise. Assistance in managing triggers including allergies and seasonal influences was highly valued especially by people with asthma:

> So I felt that he really wanted to get to the actual root of the problem which is important I think rather than just like having prescription on top of prescription and not dealing with the underlying causes and triggers (Patricia).

5.3.2.2 Information provision

Many participants indicated a need for greater information such as an explanation of their condition; potential side effects of treatments; alternative treatments and the potential serious impact of asthma or COPD. One participant described the difficulty of getting information on side effects of treatments: “You know, there’s no give and take where they could sit you down and explain it to you. Or even get the respiratory nurse to go through it with

you" (Angela). Some participants described anxiety associated with looking up information online. Participants valued when HCPs would verbally explain the information rather than relying solely on leaflets.

5.3.2.3 An action plan

Participants also sought further information on: "the possible progression of the condition; what are maybe red flags to watch out for; maybe when you should kind of seek help" (Alice). While few participants used the term "action plan", participants' accounts consistently indicated a preference for being aware of what steps to take when symptoms change or deteriorate. Some participants valued this as being a written document developed with the HCP that helped to identify symptom levels and associated actions. Participants differed in their preferences for having a plan in the case of day-to-day management with many feeling this was not necessary.

> Now my new asthma nurse actually sat down and … she wrote it all out so on my chart so it's impossible not to understand: here is normal let's say and then she said if I get into this territory, here's what I do and if I get into this territory here's what I do (Aileen).

5.3.3 Person-provider relationship

5.3.3.1 Listening to the patient's account

Participants' accounts indicated a preference for having input into consultations with their HCPs and referred to the importance of being listened to. In practical terms, this involved the HCP giving the person time to talk and provide their perspective: "you know, listened, not just cutting in" (Eleanor). Feeling listened to also involved a sense of empathy from HCPs. Many participants valued HCPs who tried to appreciate their situation and concerns,

saying: “I see where you’re coming from” (Mary). While participants acknowledged that they needed assistance in identifying their symptoms and the appropriate response, many participants considered it important that their knowledge of their own body and changes be taken seriously by HCPs. This was particularly the case for people with asthma who described a perception among HCPs and the wider public that “it’s only asthma”:

> The other thing I suppose is that you do know your own body, so I’d like to be kind of believed if you’re coming saying that you’re feeling unwell. That you’d feel that was acknowledged and validated and that person believed you (Michelle).

There were disparities in participants’ accounts of the value of tests and scans conducted by HCPs such as “listening to [the] chest”, peak flow measures, breathing tests, and chest scans. Many participants perceived physical measures of health to be informative in terms of planning treatment, to help alleviate symptoms and provided reassurance to them about their current health. However, some participants believed measures such as peak flow did not fully represent their experience of their illness: “I always seem to score well and yet I'm in bits” (Mandy). Participants’ comments indicated a desire for HCPs to respond to their experience of changes in their body even if physical tests did not indicate problems: “I find the doctor doesn’t always listen to me because I’m not wheezing at that point and my peak flow is fine but I can feel the difference” (Aileen).

5.3.3.2 Continuity and integration

Participants valued continuity in the support received for managing their health. This was predominantly discussed in terms of GPs in primary care

though it was also mentioned in terms of specialist services. They explained that from the patient perspective, it was tiresome to have to go through one's history with a new HCP on each visit and it was believed that seeing the same HCP would save time. Patients valued HCPs knowing about their illness, their individual triggers and treatment regimens as well as knowing them individually as a person. They believed this knowledge assisted in selecting the most beneficial treatment options.

> I've been to different GPs and some of them wouldn't - they wouldn't really tell you anything. Now she's [GP] not an asthmatic herself but she's just - well she knows me. She knows my asthma. She knows, you know, what works for me and what doesn't work for me (Danielle).

Participants sought someone who would oversee their treatment whom they could contact regarding their asthma or COPD: "one point of contact" (Aileen); a "liaison" (Trish); someone who "joins the dots to see how you're doing" (Angela). Participants valued a service in which there was communication between the different HCPs who support them in managing their health, such that HCPs would be made aware of changes in treatment advised by other providers. This was highlighted across primary and specialist settings, particularly for participants who also had other chronic conditions. Participants identified the potential benefit of a shared health record into which various professionals could input, potentially in electronic form, which does not currently exist in Ireland.

5.4 Discussion

This research explored the SMS preferences of people with asthma/COPD, extending the literature regarding the features of support that are most valued

and accepted from the perspective of service users. This exploratory study found that people with asthma/COPD placed emphasis on communication with their HCPs and valued HCPs listening fully to their concerns. Concurring with other studies, people with asthma/COPD reported dissatisfaction with paternalistic relationships with their HCPs characterised by a lack of opportunity to recount their experiences, input into decisions or ask questions (Hannane et al., 2019; Lingner et al., 2017; O'Connell et al., 2019). Being believed was a particular concern for people with asthma in this study who felt they were sometimes not taken seriously when presenting with symptoms they were worried about. While inadequate listening to the patient's experience has the potential to contribute to sub-optimal treatments to manage exacerbations, people with breathlessness also experience injustice when their experiential knowledge is not taken into account (Hutchinson et al., 2018). This finding also resonates with Self-Determination Theory where relatedness, that is feelings of being respected and understood, is considered a basic psychological need which in turn influences the mental health of the individual (Deci & Ryan, 2000; Ng et al., 2012). Furthermore, it is suggested that the HCPs lack of responsiveness may not only affect the management of a specific episode but may also lead to disengaged coping over time and lower help-seeking (Hutchinson et al., 2018). In addition to enhancing communication skills, training initiatives for HCPs should emphasise the importance of communication, particularly listening to and acknowledging the patient's concerns and experiences, through drawing on evidence such as this study.

Timely access to HCP advice when managing exacerbations was a valued support from the perspectives of people with asthma or COPD in this study.

There has been debate regarding the amount of responsibility that is acceptable to people with asthma/COPD in managing symptom changes (Lingner et al., 2017). It is suggested that exacerbation-related self-management may not be possible for all individuals (Korpershoek et al., 2016). Participants in this study valued reassurance from a HCP regarding actions to take when managing symptom changes. The theory of symptom self-management highlights that many variables influence this process including perceived self-efficacy which can be facilitated by direct mastery experiences as well as verbal persuasion (Hoffman, 2013). Scheduled support which builds skills in identifying and managing exacerbations was valued in the current study. However, unscheduled access to support in managing exacerbations was also valued. Interventions including both unscheduled and scheduled support have led to a reduction in severe exacerbations for COPD and asthma (Alí et al., 2019; Jain et al., 2014). Future research should further clarify the effectiveness and cost-effectiveness of unscheduled support in asthma and COPD. It would be useful to examine if unscheduled support enhances perceived self-efficacy such that people become more confident in self-managing symptoms over time. The current study also identified that cost of consultations and medications affects access and could delay help-seeking. This finding underscores the importance of implementing plans for universal healthcare in Ireland, with care free at the point of delivery, as outlined in the Sláintecare policy in order to ensure that cost does not deter timely help-seeking (Burke et al., 2018; Oireachtas Committee on the Future of Healthcare, 2017).

The findings on the importance of continuity of care, in addition to accessibility and provider relationships, reflect the healthcare system facilitators of self-management in the Self and Family Management Framework (Grey et al., 2015; Schulman-Green et al., 2016). The findings here indicated preferences for continuity in providers and having a single point of contact with services akin to the role of a case manager, as reported in previous literature (Schulman-Green et al., 2016). HCPs with specialist knowledge of asthma/COPD such as a respiratory nurse were valued as a point of contact. In addition, support from different providers was acceptable once adequate communication existed between providers such that HCPs had up-to-date information on the patient's health and treatment. A shared health record was suggested to facilitate communication between HCPs similar to another recent study with people with asthma (Hannane et al., 2019). Electronic records which facilitate information exchange across primary and secondary care HCPs have been proposed in the Irish Health Service (Health Service Executive, 2015b). While studies from patient perspectives, including those with COPD, indicate the positive impact of electronic records on information continuity, challenges are reported from the provider perspective (Waibel et al., 2016; Waibel et al., 2015). Further research is needed to enhance the features of these electronic record systems and their implementation.

This study also highlighted the need for SMS to address a range of challenges which can arise for people living with asthma or COPD. There was an emphasis on problem solving challenges which impact on daily life including managing triggers of exacerbations, side effects of treatments, emotional wellbeing and comorbidities which have been indicated in other recent studies

from the perspectives of people with asthma/COPD (Gardener et al., 2018; Hannane et al., 2019; Lingner et al., 2017). This finding aligns with recent critical perspectives which advocate a 'person-centred' approach to SMS and a focus on helping people to live well with their chronic conditions rather than manage their conditions well (Entwistle, Cribb, & Owens, 2018; Morgan et al., 2017). This approach requires an understanding about what matters generally for the population of interest (Entwistle, Cribb, & Owens, 2018) and the current study contributes to this literature in relation to people with asthma/COPD. However, at the practice level, this requires assessing what individuals with chronic illness want to achieve so that SMS can be aligned with their personal goals (Entwistle, Cribb, & Owens, 2018). Studies from the HCP perspective identify many practical and ethical challenges in achieving this such as patient goals being at odds with biomedical targets (Entwistle, Cribb, Watt, et al., 2018; Franklin et al., 2019). Further research is needed to establish consensus on how person-centred approaches can be incorporated in practice and how the quality of SMS should be assessed (Owens et al., 2017).

5.4.1 Strengths and limitations

A limitation of the study is that participants were predominately female so it is not clear if these findings are indicative of the preferences of males with asthma/COPD given previously noted gender differences in perceptions of dyspnoea, asthma control and healthcare seeking (Chhabra & Chhabra, 2011; Thompson et al., 2016). In addition, the perspectives are those of people in Ireland and thus caution is needed in generalising the findings to other countries because of differences in the organisation of health services. While plans for reform are underway (Burke et al., 2018; Oireachtas Committee on

the Future of Healthcare, 2017), the Irish health system has a predominantly hospital centric model of support for chronic disease, out-of-pocket costs for those who do not qualify for a medical/GP visit card and inequities in access to healthcare across public and private systems (Darker et al., 2015).

5.4.2 Practice implications

The many nuanced elements of management identified in this study suggest that individualised approaches to SMS are key and suggest that where group approaches are being taken, there needs to be an emphasis on assisting people with their individual challenges and applying relevant problem-solving or coping skills. This study suggests that training for HCPs who support people with asthma or COPD should place a greater emphasis on communication skills, particularly in active listening to patients concerns and acknowledging these concerns. HCPs should be aware that physical tests/measures are sometimes not felt to represent a patient's health experience (Lingner et al., 2017). From a service delivery perspective, there is a need to ensure access to support and advice when people experience early indicators of acute exacerbation. The findings suggest that contact with a known provider with specialist knowledge of asthma/COPD is preferable. Systems which allow sharing of information across HCPs such as shared electronic records are needed to facilitate continuity across HCPs involved in support.

5.4.3 Conclusions

This study has identified that people with asthma or COPD prefer comprehensive person-centred support, specialist knowledge, continuity with providers, integration between providers and for HCPs to listen and respond

to their concerns during contacts. The study finds that communication between patient and provider was an important element of support, particularly the HCP listening to the patient's experiences. People with asthma and COPD desire ease of access to support for managing asthma and COPD, particularly in the context of managing exacerbations. While this study has identified many preferable features of support, further research is needed to provide information on the relative importance of service features from the patient perspective in order to inform the prioritisation of features for future implementation. A discrete choice experiment to examine the relative importance of support attributes is developed in Chapters 6 and 7.

Chapter 6. Methodological considerations in developing and piloting a discrete choice experiment

This chapter outlines the steps of discrete choice experiment (DCE) development and describes how guidance on good practice was applied in the development and piloting of this DCE. An overview of the DCE steps is provided and literature is used to support the approaches taken in this thesis. The specific contribution of the previous thesis chapters to DCE development is detailed. A rationale is provided for narrowing the focus of the DCE to the context of support for people with asthma during a flare-up.

6.1 Introduction

A DCE is a measure of stated preferences which provides information on consumers preferences relating to goods or services and the trade-offs they make between different attributes of a service (Bridges et al., 2011; Lancsar & Louviere, 2008). The assumption underpinning DCEs is that preferences are formed based on attributes of goods or services (Lancaster, 1966). Preferences are context-dependant and constructed in the process of decision-making and thus are not stable across situations (Warren et al., 2011). They are informed by values which are defined as concepts or beliefs about desirable outcomes which hold across situations (Klose et al., 2016; van der Weide et al., 2010). Preferences involve various degrees of calculation depending on decision maker's motivation to process information relating to the decision, the level of cognitive restraint and the existence of an underlying attitude (Warren et al., 2011). Thus, preferences concern the choice of one

alternative over another in a particular context. It is proposed that the DCE is well equipped to cope with variation in preferences within and across individuals and models have been developed to account for this (Hensher et al., 2015).

Through providing information on the relative importance of service features trade-offs that participants make between service features, DCEs can assist in policy decision-making (Mandeville et al., 2014; Mühlbacher et al., 2016). This method has the potential to understand the relative importance of features of SMS which can complement qualitative data and studies of patient outcomes (Baker & Fatoye, 2019; Dineen-Griffin et al., 2019). Developing a DCE involves a number of steps and there are calls for greater transparency in the reporting of development processes (Janssen et al., 2018; Vass et al., 2017). These steps will be addressed in relation to the development of a DCE to measure the support preferences of people with asthma in this chapter.

While there were similarities in the support features valued by people with asthma or COPD as presented in Chapters 4 and 5, differences between these populations indicated a need to examine them separately. Both people with asthma and COPD valued being listened to. However, the qualitative study presented in Chapter 5 indicated that the need to be taken seriously was a particular concern for people with asthma when experiencing symptoms changes. Asthma was subsequently chosen as the focus for a number of reasons: fewer studies on support preferences of people with asthma compared to COPD as indicated in the review in Chapter 4 (O'Connell et al., 2019); the Irish self-management support (SMS) framework prioritising asthma

as focus for initial implementation of action plans in Ireland (Chronic Conditions Working Group, 2017); as well as the potential for the DCE to be more challenging for people with COPD given the cognitive burden of DCEs and increased cognitive dysfunction in people COPD (Schou et al., 2012; Veldwijk et al., 2016).

6.2 Steps in developing a DCE

Guidance on good practice was used to develop the DCE in this study. Bridges et al. (2011) outlined 10 steps for the development of a DCE which are described in Figure 6.1. Initially, a research question appropriate for the DCE, with policy relevance, must be identified (Bridges et al., 2011). Chapter 2 provided information on the policy context and the wide range of strategies used in SMS. Chapter 3 provided a rationale for research on the preferences of people with chronic disease and specifically, the need for research methods to help identify important features of support which should be prioritised in the context of numerous approaches and strategies for SMS.

Following the identification of a research question, the aim was to identify and select the attributes and levels for the DCE (Bridges et al., 2011). Attributes are the features that describe alternatives in a DCE and attribute levels differ across service alternatives. The thematic synthesis of qualitative studies on preferences of people with asthma or COPD in Chapter 4 provided initial understanding of the features of support that are important from the patient perspective. Chapter 5, which involved a qualitative descriptive study of preferences in Ireland, helped to identify features of relevance to the target population in Ireland and appropriate terminology for the development of the

DCE as recommended (Coast et al., 2012; Vass et al., 2017). Further stages of DCE design are then discussed in light of the recommended guidance: construction of choice tasks, experimental design (Reed Johnson et al., 2013), instrument design and data collection and analysis (Hauber et al., 2016). The final stages of pre-testing and piloting the DCE are reported in Chapter 7.

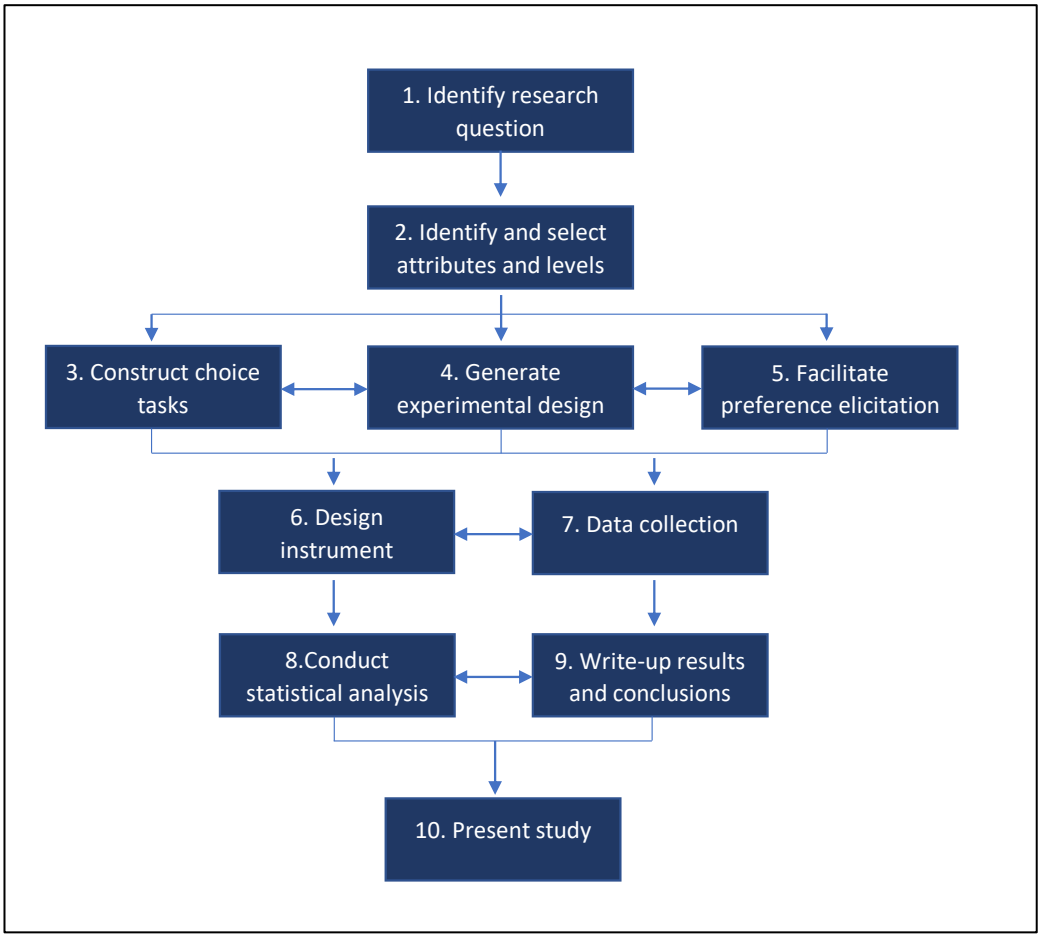


Figure 6.1 Steps to develop the discrete choice experiment

(Adapted from Bridges et al., 2011)

6.2.1 Identification of DCE attributes and levels

6.2.1.1 Attribute identification

Helter and Boehler (2016) described attribute identification and selection as a process involving data collection, data reduction, removal of inappropriate

attributes and appropriate wording of attributes. Attributes should cover areas salient to participants in order to avoid participants making inferences about elements that have been omitted from the decision context (Helter & Boehler, 2016; Lancsar & Louviere, 2008). From the categories in qualitative data reported in Chapters 4 and early data from Chapter 5, 14 potentially relevant attributes were identified (Table 6.1).

Table 6.1 Attribute identification and selection

14 attributes identified	9 attributes selected (broad concept of support)	6 attributes included (support at acute phase)
- Provider of support and advice	Type of healthcare professional	1. Healthcare professional to consult with
- Frequency of Review	How often I have a check-up	
- Access during exacerbation	Getting advice when I have concerns about change(s) in my health	2. Waiting time for response 3. Medium to discuss concerns
- Action Plan	Written plan for managing changes in my symptoms	4. Having a written plan for managing changes
- Cost of services - Cost of medications	Cost of visit	
- Providers knowledge of me pre-consultation - Integration of providers	How well my healthcare professional knows me	5. How well the healthcare professional knows me
- Explanation of information - Impressing seriousness of illness	Explanation of information	
- Acknowledgment of lived experience - Extent of shared responsibility	Listening to my experience	6. Response to my concerns
- Thoroughness of review (tailoring)	Thoroughness of my check-up	
- Linkages to other supports and resources		

6.2.1.2 Attribute selection

However, it is recommended that DCEs have no more than seven attributes in total in order to reduce the cognitive burden on participants (Helter & Boehler, 2016; Reed Johnson et al., 2013). The process of identifying and selecting a limited number of attributes for inclusion in the DCE from rich qualitative data has been acknowledged as challenging (Coast et al., 2012). A number of

considerations were used to aid the selection of attributes. Attributes should be relevant to the research question and to the policy decision context; plausible; capable of being traded; unrelated to each other; not too close to the construct being studied; and not overly dominant on the decision (Bridges et al., 2011; Helter & Boehler, 2016). An iterative process of interviewing facilitated identification of attributes as recommended for qualitative research in the development of a DCE (Coast et al., 2012; Coast & Horrocks, 2007). This was achieved through identifying a potential list of attributes from 5 individual interviews and one focus group interview (Chapter 5). Three subsequent individual interviews and an additional focus group interview followed the topic guide but also probed further into participants perspectives of emerging attributes. This process aided the selection of 9 potential attributes based on the strength of categories in the qualitative data (Table 6.1). These 9 attributes concerned support in acute phases of asthma as well as routine management.

In accordance with recommendations to have no greater than seven attributes (Helter & Boehler, 2016; Reed Johnson et al., 2013), there was a need to further reduce the number of DCE attributes. Informal consultations with four respiratory nurses and four respiratory consultants helped to refine the DCE. This feedback, the available literature and discussions with the supervisory team and an experienced DCE researcher informed the focus of the DCE. The DCE was centred on support during an acute flare-up rather than including attributes relevant to both acute care and ongoing routine support. This was largely due to the emphasis on acute support in the qualitative descriptive study presented in Chapter 5 and supporting evidence that management in

acute phases of disease remain a challenge despite chronic disease management programmes aiming to reduce these (Peytremann-Bridevaux et al., 2015). Using the qualitative data on the perspectives of people with asthma in Ireland, attributes which were more relevant to routine support and less relevant to the context of a flare-up were not included in the DCE. This included frequency of review, thoroughness of review and explanation of information.

As can be seen in Table 6.1, further refinement of attributes included separating ‘access during exacerbation’ into 2 attributes: ‘waiting time’ and ‘medium to discuss concerns’ as it encompassed two features. Lastly, cost of visit was not included. Similarly, cost was not included in a DCE on long-term asthma management which also included attributes relevant to flare-up management such as an action plan and asthma crisis management strategy (Haughney et al., 2007). Cost was identified by a small number of participants in the qualitative study as playing a role but did not feature as prominently as other features in the qualitative data. Furthermore, introducing cost risked reducing the plausibility of the DCE as methods of payment vary in the Irish Health Services. Thus, as recommended by Bridges et al. (2011), this relevant feature was held constant in the DCE with participants asked to imagine that the cost of the services did not differ. This yielded 6 attributes for inclusion in pretesting.

6.2.1.3 Level selection

There is no clear best practice with regard to selecting the levels in a DCE. However, it is suggested that levels should not be greater than 4 in number, should avoid ranges and terms which are open to interpretation such as ‘high’,

'medium' and 'low', and should avoid extreme values (Bridges et al., 2011; Johnston et al., 2017). However, it is also important that there is sufficient range in the levels such that attributes are not ignored because of limited difference in levels (Lancsar & Louviere, 2008). Qualitative data from Chapter 5 was used to identify plausible levels of attributes. Feedback from respiratory nurses and consultants was also used to assess the plausibility of levels of attributes before pretesting. Using these criteria, the attributes and levels in Table 6.2 were selected for pretesting.

Table 6.2 Attributes, attribute description and levels for pretesting

Feature	Description	Levels
Having a written plan for managing changes	Having a written plan made out with a healthcare professional. This would guide you in what symptoms warrant action and the corresponding actions to take.	- Yes - No
Healthcare professional to consult with	The type of healthcare professional you would like to discuss your concerns with. Respiratory nurses and respiratory doctors/consultants have specialist knowledge of asthma.	- General practitioner (GP) - Respiratory nurse - Respiratory doctor/consultant
Medium to discuss concerns	The medium in which you discuss your concerns with your healthcare professional	- Face-to-face meeting - Video/skype call - Phone call - Text/email
Waiting time for response	How long you have to wait to consult with a healthcare professional and get a response to your concerns	- Up to 4 hours - Up to 24 hours - Up to 3 days - Up to 1 week
How well the healthcare professional knows me	How well the healthcare professional knows you from consultations over time. In all cases the healthcare professional would have access to your medical notes.	- Does not know me - Knows me somewhat well - Knows me very well
Response to my concerns	How much your healthcare professional listens to your concerns, takes them seriously and responds	- My concerns are taken seriously - My concerns are taken somewhat seriously

6.2.2 Construction of choice tasks

The design of the DCE involves trade-offs between increasing statistical efficiency and response efficiency. This involves acquiring sufficient data to

model preferences while also ensuring that participants are not overly cognitively burdened by the DCE and answer all choice tasks in line with their preferences (Bridges et al., 2011; Reed Johnson et al., 2013).

6.2.2.1 Full or partial profile choice sets

Full or partial profiles can be utilised in a DCE. Using full profiles means that all attributes will be represented in each choice set. Using partial profiles means that only some of the attributes are presented within each choice set. If there are too many attributes, a full profile can become burdensome for participants (Bridges et al., 2011). On the other hand, full profiles will provide greater information on trade-offs and partial profiles may be confusing for participants (Bridges et al., 2011; Janssen et al., 2018). It is also possible to present all attributes and only allow a certain number of attributes to change across alternatives within a choice set. This is known as 'overlap' and has been found to have positive effects on participation such as reduced drop-out and increased choice consistency (Jonker et al., 2019). Therefore, this research placed restrictions such that a maximum of five out of six attributes could vary across alternatives within the same choice set with all attributes being shown in each choice set.

6.2.2.2 Number and nature of choice alternatives

Many DCEs present two alternatives to participants though it is possible to present more and typically these participants would be presented with less choice sets (Bridges et al., 2011). Alternatives can also be labelled (transport alternatives may be labelled 'train' or 'car' with attributes describing each) or unlabelled (Service A, Service B). Labels have been shown to reduce attention

given to attributes and unlabelled alternatives are suggested to be more suitable where the focus of the DCE is on attribute trade-offs (de Bekker-Grob et al., 2010). For this reason, unlabelled alternatives of ‘Service A’ and ‘Service B’ were used in this DCE.

An additional consideration is having an opt-out alternative in each choice set. The opt-out has been presented in various ways including a 'choose neither' alternative, a ‘no treatment’ alternative or as a status quo such as 'choose my current service'. Broadly, including an opt-out is recommended if the decision context in reality would not involve a forced choice and for the purposes of capturing demand for a service (Bridges et al., 2011; Hensher et al., 2015). Opt-out options have also been found to increase response rate (Watson et al., 2017). For the purposes of the pilot study, it was decided to include an opt-out option. However, including an opt-out alternative does reduce information available to estimate the model and preference structure if people choose the opt-out alternative (Veldwijk et al., 2014). Furthermore, it has been found that people can sometimes choose an opt-out in order to avoid the effort of comparing alternatives (Bridges et al., 2011; Veldwijk et al., 2014). In order to overcome this, some researchers have used a two-staged choice model or dual response format in which participants are first asked to make a forced choice and then provided with the option to opt-out afterward (Muhlbacher & Johnson, 2016). In this pilot study, participants were asked a follow-up question if they would use the service they have chosen (akin to an unforced choice) following a forced choice between two alternatives (Ho et al., 2015). In line with the recommendations of previous research, participants were asked to explain their reasons for not using the services (Veldwijk et al., 2014).

6.2.3 Experimental design

In generating an experimental design, researchers are trying to ensure that parameters of interest can be identified in the final analysis (Hensher et al., 2015; Muhlbacher & Johnson, 2016). If there are a small number of attributes and levels, it may be possible to use a full factorial design, that is, to present all possible combinations of profiles to participants. However, as the number of attributes and levels increase, the number of combinations becomes unfeasible to present to participants even if choice sets are blocked into different survey versions across the sample (Hensher et al., 2015). Thus, fractional factorial designs are often used where a smaller number of choice sets are identified that are capable of providing information on the parameters of interest.

In order to generate an experimental design, it is recommended that information on the attributes, levels and alternatives must be available as well as the intended model for analysis (Reed Johnson et al., 2013). The following are desirable properties of a DCE design: orthogonality: attribute levels vary independently; balance: each level of an attribute appears an equal number of times; and finally efficiency: minimising the variance in order to increase the precision of the final analysis (Bridges et al., 2011). While an efficient design was used for this DCE study, consideration was given to both orthogonal and efficient designs. Both designs are considered in the next subsections justifying why an efficient design was chosen over an orthogonal design.

6.2.3.1 Orthogonal designs

Orthogonal designs are generated satisfying the criteria of orthogonality and attribute level balance. They require a large number of choice sets in order to estimate effects. Thus, blocking is often used where choice sets are blocked into different survey versions for participants. Good practice is to have 8-16 choice sets per participant with the final number being informed by pretesting (Bridges et al., 2011). However, it is argued that in practice, data obtained is rarely orthogonal with this requiring equal distribution of blocked choice sets across the sample and not accounting for individual-level variables which may also be incorporated in analysis (Hensher et al., 2015). In addition, orthogonal designs can include dominant alternatives which do not provide information on parameters of interest. While orthogonal designs are still used, especially in pilot stages, researchers have moved towards efficient designs which seek to overcome the limitations of orthogonal designs (Hensher et al., 2015; Reed Johnson et al., 2013).

6.2.3.2 Efficient designs

The aim of efficient designs is to maximise the information from each choice situation and minimise error associated with the parameter estimates. Efficient designs may not be balanced but removing the criteria of attribute level balance can generate a more efficient design (Hensher et al., 2015). Once there is near balance and orthogonality, designs are well identified (Reed Johnson et al., 2013). Efficient designs also allow greater precision of estimates or estimation of effects with smaller sample sizes (Reed Johnson et al., 2013). However, efficient designs require the researcher to input prior information on the parameters and thus care is needed to ensure that prior

information is accurate (Reed Johnson et al., 2013). Different forms of efficient design also exist. Bayesian-efficient designs can be generated using prior information but rely less on the accuracy of the prior estimates. Thus, a Bayesian efficient design was applied in the pilot phase of this DCE. Prior values were inputted to the design to indicate the hypothesised direction of estimates. The pilot study was used to assess the reasonableness of estimated prior values and to define prior values for the final DCE as recommended (Johnston et al., 2017).

A number of programmes have been used to develop experimental designs which differ in the extent to which they allow researchers to manipulate or control the aspects of design; provide measures to evaluate the design; and the level of expertise required by the researcher (Hensher et al., 2015; Reed Johnson et al., 2013). This DCE study used JMP software, version 14 (SAS Institute, Cary, NC, USA), to achieve the desired properties of experimental design including a Bayesian-efficient design; use of priors; creation of overlap and blocking of surveys.

6.2.4 Preference elicitation and instrument design

6.2.4.1 Appropriate task introduction

The DCE included the following information with the aim of providing an appropriate introduction to the DCE: a clear rationale for the study in order to encourage attention to each of the choice sets (Bridges et al., 2011; Vass & Payne, 2017); a clear description of attributes and levels (Bridges et al., 2011); presentation of the choice context (i.e. potential flare-up) and frequent

repetition of the choice context throughout the questionnaire (Bridges et al., 2011).

6.2.4.2 Respondent information

The instrument also collected respondent information in order to describe the sample as well as factors that may influence preferences (Bridges et al., 2011; Veldwijk et al., 2016). This included length of diagnosis; presence of other chronic illnesses; methods of payment for healthcare as well as current levels of asthma control as measured by the Asthma Control Questionnaire (ACQ)(Juniper et al., 1999).

6.2.4.3 Additional items to assess validity and reliability

Assessing the validity and reliability of DCE data at individual and aggregate levels is another important component of analysis. However, the optimal means of assessment are unclear (Janssen et al., 2018; Janssen et al., 2017; Soekhai et al., 2019). Some tests can be carried out to identify individual 'irrational' responses based on the existing participant data, however, others require the inclusion of additional questions. Without guidance on the most important tests, the decision to include additional questions becomes a trade-off between the additional information sought and ensuring that any additions to the survey do not overly burden to participants such that they might compromise completion of the overall DCE (Janssen et al., 2017). Furthermore, there has been debate over what constitutes an irrational response and thus how these data should be handled (Lancsar & Louviere, 2006). For example, some researchers have assessed stability of choices through repetition of a choice task within a questionnaire (Janssen et al.,

2017). However, it has been shown that different answers to the same choice card presented later may not in fact be irrational and may be due to participants learning throughout completion (Lancsar & Louviere, 2006). This DCE included a measure of choice validity through a dominant alternative test where one choice set contained an alternative where all attribute levels were superior to the levels of the other alternative. Failure to choose the dominant alternative would be considered an irrational response (Ryan et al., 2012).

6.2.4.4 Pre-testing

Pretesting provides qualitative feedback on participants' interpretations of attributes, the readability and the acceptability of the questionnaire (Bridges et al., 2011). Pre-testing also reduces the risk of implausible combinations of attribute levels. If choice tasks contain two attribute levels that do not realistically belong in one service, then this would affect the participant's ability to trade between attributes. A common example is where participants may associate high price with high quality and find a low price/high quality good implausible. While implausible combinations were sought to be reduced through evaluation by the research team, an experienced DCE researcher and clinician feedback, it is highlighted that these should be defined from the perspective of the participant and can be further understood through pretesting (Lancsar & Louviere, 2008). Pre-testing is emphasised as an important component of DCE development as well as pilot testing (Bridges et al., 2011; Johnston et al., 2017).

While pre-tests of DCEs have not often reported in detail, cognitive interviews have been applied for this purpose (Vass et al., 2017). Literature on cognitive

interviewing, recommends sampling participants with a range of characteristics representative of the target population who would be eligible to complete the main DCE (Blair & Conrad, 2011). Use of a framework is suggested to reduce subjectivity in the analysis of cognitive interviews (Drennan, 2013). A pre-test study, reported in Chapter 7, drew upon these recommendations to conduct pre-testing. An iterative process of cognitive interviewing was conducted in rounds where participants consistently identified an issue for amendment which was amended in the subsequent survey until no further issues warranting amendment were identified (Blair & Conrad, 2011).

6.2.5 Data Collection

Many previous DCEs in healthcare have been paper based surveys, delivered face-to-face or by post, whereas now, DCEs are increasingly being conducted online (Soekhai et al., 2019). Online platforms can facilitate more complex pivoting designs and randomisation of survey version, task and attribute order as well as ease of access to large samples with wide geographical coverage (Hensher et al., 2015). Online surveys are often conducted through online panels which may influence the sample and findings of the study (Johnston et al., 2017). Although requiring significant investment of researcher time, surveys administered face-to-face may enhance the quality of data by addressing participant queries in the moment, particularly for participants who may be older or with lower health literacy (Bridges et al., 2011; Veldwijk et al., 2016). A study comparing face-to-face to online methods for a stated preference study found similar results for both methods after they accounted for differences in the composition of the samples for which the face-to-face survey sample included participants who were older and had lower education

(Saloniki et al., 2019). The authors emphasised the need to provide comparable levels of clarification to participants across each survey mode. A self-completed paper based DCE questionnaire was used in this pilot study in a clinic setting with the researcher available to address queries with the aim of representing participants across a range of demographics.

The calculation of sample size for DCEs has been a developing area of research. Traditionally, rules of thumb were used to guide sample sizes (Bridges et al., 2011). However, methods have been developed to calculate minimum sample sizes for DCEs taking parameter estimates into account (Bliemer & Rose, 2010; de Bekker-Grob et al., 2015). The method of de Bekker-Grob et al (2015) takes greater information into account including desired power level and the authors advocate the use of a pilot study to identify parameter values. Within the scope of this thesis, it was aimed to conduct a pilot study to obtain these parameter values in order to develop an efficient design and to calculate the necessary sample size required for an adequately powered DCE study.

6.2.6 Data analysis

Many different statistical models have been used to analyse DCEs. Models differ in how they account for or distribute unobserved variability across the sample (Hensher et al., 2015). The conditional logit model serves as the basis for many discrete choice models (Hensher et al., 2015). The conditional logit model and multinomial logit (MNL) model are the same basic model which use the same methods of calculation and rely on the same underlying assumptions (Lancsar et al., 2017). Sometimes the term ‘conditional logit’ is used to

describe models focusing on relating choices to the components of alternatives while MNL model is used where choices are related to characteristics of participants (Hauber et al., 2016). The MNL model will be the term used henceforth. Random Utility Theory as outlined by McFadden (1974) highlighted that utility comprises of an observed, systematic component and an unobserved, error component. The utility function for any respondent *i* choosing between *j* alternatives is given by the formula:

$$U_{ij} = V_{ij} + \varepsilon_{ij}$$

Where V_{ij} refers the systematic or observed utility captured by observed factors and ε_{ij} refers to the unobserved variation. From this definition, it is possible to see that the characteristics of observed utility (V_{ij}) depend on the specification of ε_{ij}. Therefore, the way in which error is modelled has implications for the estimated utility.

The MNL model assumes the error is identically and independently distributed according to a type 1 extreme-value distribution (Hauber et al., 2016). This assumption is described as independence of irrelevant alternatives (IIA) and often does not hold for choice data. The MNL model, through assuming IIA, does not account for variance across choice questions or groups of respondents (scale heterogeneity) or unobserved systematic differences in responses across participants (preference heterogeneity) and so other models have been developed to better account for this variation (Hauber et al., 2016). The nested logit model, the heteroscedastic error components model and the mixed multinomial logit model (MMNL), relax the IIA assumption in different ways and have been combined in the generalised mixed logit model (Lancsar

et al., 2017). Another type of analysis is latent class analysis where a number of classes are used to describe patterns of preferences. There are many potential models for choice data and it is suggested that it may not be possible to know the most appropriate analysis strategy until analysing the data and comparing the goodness of fit of the models (Bliemer & Rose, 2010).

The models which account for greater heterogeneity may require larger sample sizes due to the inclusion of more parameters (Hauber et al., 2016). For the purposes of the pilot study, the analysis was based on the MNL model. The MNL model can be a reasonable approximation of preferences where alternatives are unlabelled (Lancsar et al., 2017) as is the case in the forced choice component of this study. The MNL was also used for the dual response unforced choice analysis using an alternative specific constant as detailed in Campbell and Erdem (2019).

6.3 Conclusion

This chapter provided a description of the recommended steps in developing a DCE and the actions taken in this DCE to achieve those steps. A wide range of evidence was used to select and provide rationale for actions to meet the recommendations of DCE development. Challenges in this process included selecting a limited number of attributes from rich qualitative data; identifying the optimal presentation of choice sets; and justifying the inclusion of some measures of DCE validity while balancing the number of additional questions with the overall survey burden. The next chapter presents findings from a pilot study of the DCE including as well as pre-testing using cognitive interviews.

Chapter 7. Preferences for support in managing symptoms of asthma flare-up: a pilot study of a discrete choice experiment.

This chapter describes a pilot study of a discrete choice experiment (DCE) to measure adults' preferences for support in managing symptoms of an asthma flare-up. Following on from the steps in developing a DCE outlined in Chapter 6, the resultant features of the DCE are described in this chapter including the refinement of the DCE following cognitive interviews (Section 7.2). A multinomial logit (MNL) model is used to analyse preferences from 38 people with asthma recruited from specialist asthma clinics. Implications for future DCE research and clinical practice are discussed. It is planned to submit the content of this chapter to *The Patient - Patient-Centered Outcomes Research.*

7.1 Background

Asthma is a chronic respiratory illness which affects many people worldwide. Prevalence estimates vary from 1-21% across countries (To et al., 2012). Globally, asthma was the 11th leading cause of years lived with disability in 2015 (Vos et al., 2016). Asthma symptoms include shortness of breath, chest tightness, wheeze and cough and are characteristically variable between individuals and over time (GINA, 2019; Pickles et al., 2018). People with asthma may experience 'flare-ups' or exacerbations which are periods of worsening of symptoms compared to the individual's normal level of symptom severity (GINA, 2019; Jones et al., 2019; Pickles et al., 2018). Flare-ups can result in hospitalisation and or absenteeism/loss of productivity in work which

have social and economic costs to the individual and society (Mukherjee et al., 2016; Nguyen et al., 2017; Pickles et al., 2018; Sullivan et al., 2011).

Maintaining optimal asthma control involves the use of controller medication while additional treatments such as reliever inhalers are taken when necessary (GINA, 2019). The Global Initiative for Asthma recommends that support for asthma includes disease education and medication adherence, teaching and demonstrating proper inhaler technique, providing smoking cessation advice and support, goal-setting, as well as providing assistance in identifying and managing an individual's asthma triggers (GINA, 2019). Many interventions have been designed to improve the management of asthma including self-management support (SMS) to enable individuals to actively engage in managing their condition for optimum disease control and wellbeing (Miller et al., 2015; Pinnock, 2015; Pinnock et al., 2017). Chronic disease management interventions providing SMS for asthma have shown positive effects in terms of quality of life and asthma severity but impact on emergency department (ED) visits or hospital admissions have been small and inconsistent across studies (Peytremann-Bridevaux et al., 2015; Pinnock et al., 2017). Analyses of asthma-related deaths indicate that a large proportion are preventable, approximately 30% of which involve delays in help-seeking (Goeman et al., 2013). These findings suggest a need for continued research on factors affecting the management of acute exacerbations including help-seeking (Lawson et al., 2014).

A number of factors that can influence management of an asthma flare-up and help-seeking in the context of a flare-up have been identified. International

guidelines recommend action plans to aid patients in recognising and responding to worsening of their symptoms (GINA, 2019). These are individually tailored plans made between the patient and the healthcare professional (HCP). They detail actions required to manage different levels of symptoms. While many studies have indicated that action plans have positive effects; overall, the evidence is unclear and of low quality (Gatheral et al., 2017). Furthermore, despite recommendations, action plans are often not implemented as intended (Price et al., 2019). A qualitative study to explore implementation barriers found that patients often do not value action plans and that often HCPs do not prioritise them (Ring et al., 2015). Studies have also explored reasons for emergency department (ED) attendance for acute asthma. While ED presentation may be required due to severity of the exacerbation, in other cases people attend ED due to the inaccessibility of their primary care provider, wating times for an appointment, or due to greater perceived expertise in the ED (Fleetcroft et al., 2016; Lawson et al., 2014). Further information is needed to understand the service features that are most important from the patient perspective in order to identify interventions that are most likely to lead to timely and appropriate help-seeking. To the best of my knowledge, no study has quantitatively examined how people with asthma value and trade-off different attributes of support when managing a flare-up.

A DCE is a technique for eliciting preferences for goods and services. DCEs provide information on the relative importance of different features of a good/service and how people trade-off between features. The DCE is underpinned by Lancaster's model of consumer choice (Lancaster, 1966) which suggests that the utility of a good/service is derived from its attributes.

A DCE is a written instrument which asks participants to hypothetically choose their preferred service alternative from two or more alternatives. These alternatives are described by multiple attributes which are features of the service and levels of attributes vary across the alternatives. For example, waiting time until an appointment is frequently an attribute of primary care and different levels are presented in the hypothetical service alternatives such as waiting one day versus waiting three days for an appointment (Kleij et al., 2017). Participants typically complete a number of choice tasks which are used to derive the utility of attributes.

DCEs have been increasingly employed in healthcare to explore preferences of patients, providers and policy-makers for different characteristics of goods and services (de Bekker-Grob et al., 2012; Kleij et al., 2017; Soekhai et al., 2019). DCEs can be used to explore preferences for service features in order to aid policy decisions regarding the features of a service that should be implemented (Lagarde & Blaauw, 2009; Mandeville et al., 2014). While there is some debate on how well these hypothetical data predict real choice and behaviour (Rakotonarivo et al., 2016), the DCE affords the opportunity to directly compare service features from the perspectives of service users, providing information that cannot be obtained through other sources of data.

A checklist for the application of DCEs in healthcare has identified a number of stages in developing a DCE including identifying and selecting the attributes and levels of attributes; constructing choice tasks; generating an experimental design; designing the DCE instrument for data collection and pretesting the instrument (Bridges et al., 2011). The application of these stages will be further

detailed below in the methods section. A pilot study, is an additional important step to enhance the reliability and validity of the DCE (Janssen et al., 2017; Johnston et al., 2017; Lancsar & Louviere, 2006; Soekhai et al., 2019). A pilot study facilitates an evaluation of the survey administration process (Johnston et al., 2017) and provides quantitative information on preference estimates which can be used as priors for the design of the main DCE along with calculation of sample size (de Bekker-Grob et al., 2015; Vass & Payne, 2017). This chapter reports on a pilot study of a DCE to examine the relative importance of service features for adults managing an asthma flare-up in Ireland.

7.2 Methods

7.2.1 Study Design

A pilot study of a DCE was conducted. It is envisaged that this pilot will be used to inform the main DCE study at a later stage following completion of the PhD research. The International Society for Pharmacoeconomics and Outcomes Research checklist for the application of DCEs in healthcare was used as a guide for DCE development (Bridges et al., 2011). The study was approved by the Clinical Research and Ethics Committee of the Cork Teaching Hospitals (Reference number ECM 4 (w) 10/09/19 & ECM (fff) 10/09/19; Appendix 6.A). The researcher provided an information leaflet and explained the study to participants within the clinic, allowing an opportunity for them to ask questions. Written informed consent was provided prior to participation (See Appendix 6.B and Appendix 6.C for the information/consent sheets for pre-test and pilot studies). The final questionnaire is included in Appendix 6.D

7.2.2 Participants

Participants were recruited through a tertiary-level, specialist outpatient asthma clinic in the South of Ireland. Members of the Respiratory team identified patients who met the inclusion criteria: adults (aged 18 and over) with a diagnosis of asthma longer than one year, who had previously attended the asthma clinic, who could read and write in the English language and provide informed consent. The researcher provided an information leaflet and explained the study to participants within the clinic, allowing an opportunity for them to ask questions.

7.2.3 Designing the Discrete Choice Questionnaire

7.2.3.1 Identifying attributes and levels

As reported in Chapter 6, a review of qualitative literature on preferences of people with asthma (O'Connell et al., 2019), and a qualitative descriptive study of support preferences including people with asthma in Ireland were used to identify attributes and terminology relevant to the population of interest for this study. An iterative process of interviewing outlined in Chapter 5 and 6 facilitated selection of attributes as guided by Coast et al. (2012). Researchers also used criteria for selection of attributes such as plausibility, relevance to the research question, independence from other attributes (Bridges et al., 2011; Helter & Boehler, 2016). Respiratory clinicians also provided feedback on the appropriateness of the attributes selected for the DCE prior to pretesting. Six attributes were identified for inclusion in pretesting, with seven attributes suggested as the maximum number to reduce the cognitive burden on participants (Helter & Boehler, 2016; Reed Johnson et al., 2013). These attributes concerned having a written plan for managing changes; the type of

healthcare professional to consult with (GP, Respiratory nurse, Respiratory doctor); waiting time for response (ranging from 4 hours to 1 week); the medium to discuss concerns (face-to-face, video call, phone call, text/email); how well the HCP knows the person; and the HCPs response to their concerns (extent to which taken seriously). See Table 6.2 for further detail.

7.2.3.2 Constructing choice tasks

Participants were presented with choice sets asking them to make a choice between two unlabelled hypothetical alternatives – Services A and B (Table 7.1). This choice between these two service alternatives is described as a forced choice. Following each choice set, participants were provided with an option to opt-out of choosing the service – akin to an unforced choice – through asking whether they would use the service chosen in reality or not. A dual response format where participants were provided with the option of opting out after making a forced choice was used in order to minimise information loss regarding attributes/levels in the DCE (Veldwijk et al., 2014). An open question at the end of the questionnaire asked for reasons for opting out as recommended (Veldwijk et al., 2014). Overlap between attribute levels was included such that only 5 attributes varied across service alternatives in each choice set. Previous evidence suggests that this reduces cognitive burden (Jonker et al., 2019; Kessels et al., 2011). A utility neutral design was used to generate choice sets for the purposes of pretesting in JMP software, version 14 (SAS Institute, Cary, NC, USA). The pilot experimental design is described in Section 7.2.3.5.

Table 7.1 Example of a DCE choice set in Pilot study

	Service A	Service B
Having a written plan that I can refer to	No	Yes
Healthcare professional to consult with	A respiratory nurse	A general practitioner
Time to consultation	Next day	Same day
Method of communication	Face-to-face meeting	Face-to-face meeting
Healthcare professional's knowledge of my health	Knows me and has my notes	Does not know me but has my notes
Response to my concerns	Appears to listen	Appears not to fully listen

7.2.3.3 Constructing the DCE instrument

The DCE aimed to measure preferences for services in the context of a developing asthma flare-up and thus participants were asked to imagine that they were experiencing a change in their symptoms that may lead to an asthma flare-up. This context was regularly repeated throughout the instrument. Participants completed 13 choice sets in total –12 experimental choice sets preceded by a sample dominant choice task. A dominant choice task contains one alternative which is superior to the other alternative on every attribute and thus is expected to be chosen if participants understand the task and are making choices based on levels of attributes (Janssen et al., 2017). Participants were also asked to rank the attributes in order of preference following completion of the choice cards.

The questionnaire also comprised demographic questions providing information on self-reported age, gender, length of diagnosis, presence of other chronic conditions, education and method of payment for healthcare, taking into account the private/public nature of healthcare delivery in Ireland. Participants also completed the Asthma Control Questionnaire (ACQ)(Juniper

et al., 1999) an internationally recognised, validated measure of asthma control (author permission received) which comprises self-report questions regarding symptoms and a measurement of forced expiratory volume. ACQ scores range from 0-6 with scores ≥ 1.5 deemed indicative of sub-optimal asthma control (Juniper et al., 2006).

7.2.3.4 Pretesting: cognitive interviews

Cognitive interviews were conducted combining a recommended think–aloud approach verbal probing in response to participants' reactions and debriefing question following completion (Veldwijk et al., 2016). The researcher used an interview protocol (Appendix 6.E) guided by a framework denoting key problem areas in instrument development: understanding of terminology, unanswered questions, understanding of response options, order of questions, cognitive burden and any other issues raised by participants (Conrad & Blair, 1996; Drennan, 2013). Cognitive interviews were conducted in three rounds where amendments were made to the survey based on issues identified in a previous round until no further amendments were indicated (Blair & Conrad, 2011). Interviews were conducted one-to-one in a quiet location in the clinical setting, lasting between 11 and 36 minutes and were audio-recorded.

Sixteen participants (9 male, 7 female) meeting the inclusion criteria completed cognitive interviews. Participants were broadly representative of the target population of the DCE (Blair & Conrad, 2011). Demographic characteristics are presented alongside pilot study participants in Table 7.4. Six participants (37.5%) had at least one other chronic illness and participants had varied

levels of education and methods of payment for healthcare. Eight participants (50%) were classified as having controlled asthma as defined by ACQ < 1.5.

Comments from cognitive interviews were categorised according to the framework that guided questioning (Conrad & Blair, 1996; Drennan, 2013). Problems identified and associated resolutions are detailed in Table 7.2 and key amendments are discussed. In terms of lexical issues, the choice context was re-worded using the term ‘flare-up’ instead of ‘attack’ in order to capture subacute changes in symptoms and gradual rather than abrupt worsening of symptoms. Within temporal/response option issues, attribute levels were amended where perceived to be too extreme e.g. waiting greater than 2 days for consultation. The attribute regarding response to concerns was altered to focus on listening to reduce ambiguity. All attributes were perceived as important by participants though having a written plan was mentioned as being of less importance. However, this attribute was included due to its policy relevance. Participants suggested that HCPs having access to records was important and this was incorporated as a level in the attribute regarding the HCP’s knowledge of patient’s health.

Table 7.2 Key refinements to the DCE instrument resulting from pretesting

Item	Problem	Resolution
Lexical: words	- Original scenario wording including 'acute attack' not capturing milder flare-up and gradual worsening of symptoms	- Wording altered to 'a change in your symptoms and you are concerned this may develop into a flare-up of your asthma' based on literature in this area
	- 'Medium to discuss concerns' was not clear to participants at recollection	- Wording altered to 'method of communication'
	- 'Having a written plan for managing changes' not adequately understood, questioned if generic or kept by HCP or patient	- Labelled as 'Having a written plan that I can refer to' simplified description, included that it is personalized and kept by participant
Inclusion/ exclusion:	- Question if necessary to fill in answers in description	- Made clearer that first section is introduction through text and font
Temporal: response options	- Text/email and skype methods of communication perceived as less suitable by participants	- Removed level of 'text/email' and 'skype/video call'
	- Up to 1 week and up to 3 days perceived as unacceptable	- Reduced time range and altered the 'time to consultation' from hour durations to consultation within the 'same day', the 'next day' or 'two days later'.
	- Assuming access to notes is not realistic and participants have difficulty with idea of HCP knowing them well	- Included notes and revised wording to: • Neither knows nor has my notes • Does not know me but has my notes • Knows me and has my notes
	- Response to my concerns - expression 'taken seriously' problematic and hard to conceptualise	- Focus changed to 'listens' given the regular use of this expression and its prominence in previous research and DCE methods
	- Low value often given to written plan Additional attributes of importance identified by participants such as a shared health record system.	- Considered removal of written plan but it was retained given policy implication. Some additional suggestions considered outside of scope of DCE. Idea of HCPs having access to notes was included within existing attribute HCP knowledge of patient's health.
	- 'Choose neither' option somewhat confusing and sometimes not completed	- Revised opt-out to a question requiring a response with phrasing: if you were experiencing a change in your symptoms, would you use the service chosen if it were available to you? Yes, No
Logical: Order	- Participants unsure if questions are linked, perceive questions to be repetitive and some unsure if scenario applies to second choice question presented	- Added to introduction that scenario remains the same but each question stands alone; features of service will always be the same but that the details within these (levels) will differ; every question helps to get a greater understanding of peoples' preference; and context will remain the same and will be printed at the top of each page
Computational: mental operations	- Originally some perception that 6 attributes may be too many	- 6 attributes were perceived as acceptable in later rounds.
	- Some participants considering their own experience or trying to represent other people's preferences	- Emphasis placed on answering from personal perspective and to focus on their preferences rather than experience
	- Participants not using all numbers from 1-6 on rating question	- Refine instruction to convey need to use all numbers from 1-6

Clarifications to wording were also included in the task introduction to enhance task understanding and overcome logic issues. In terms of computational issues, there was a concern in the initial round that six attributes may be too many. However, six attributes were perceived as acceptable in later rounds following clarifications. The final attributes for inclusion in the pilot study are detailed in Table 7.3.

Table 7.3. Attributes, description and levels as presented to participants in pilot study*

Feature	Description	Levels in choice card	Sign
1. Having a written plan that I can refer to	A personalised plan made out with a healthcare professional which you keep so that you can refer to it. It identifies the actions you need to take in response to your symptoms.	• Yes • No	+ sign - sign
2. Healthcare professional to consult with	The healthcare professional may have general medical knowledge (e.g. a GP) or may have specialist knowledge of asthma (e.g. respiratory nurses or respiratory doctors/consultants).	• A general practitioner • A respiratory nurse • A respiratory doctor/consultant	- sign + sign
3. Time to consultation	The time you have to wait to consult with a healthcare professional from the time you make the initial contact	• Same day • Next day • Two days later	+ sign - sign
4. Method of communication	The medium through which you discuss your concerns with your healthcare professional	• Face-to-face meeting • Phone call	No a priori sign
5. Healthcare professional's knowledge of my health	The healthcare professional may have access to a record/notes of your medical history and/or knows you from previous consultations	• Neither knows me nor has my notes • Does not know me but has my notes • Knows me and has my notes	- sign + sign
6. Response to my concerns	How much your healthcare professional listens to your concerns and addresses them	• Appears to listen • Appears not to fully listen	+ sign - sign

*Sign column was not presented to participants and indicates hypothesised direction of preferences used to inform the efficient design.

7.2.3.5 Experimental design

A Bayesian D-optimal design was generated using JMP software, version 14 (SAS Institute, Cary, NC, USA) for estimation of main effects within a multinomial logit (MNL) model which was the planned model for analysis. This design approach reduces dominant alternatives and minimises the error around co-efficients, facilitating parameter identification using smaller sample sizes than orthogonal designs (Bridges et al., 2011). Prior values were used to provide direction for attribute levels according to logic (See Table 7.3). For example, with regard to time to consultation, it was expected that consultation on the same day would be preferred (+ sign) to the next day which would in turn be preferred to two days later (- sign). The design contained 24 choice-sets divided into two blocks of 12 in order to reduce cognitive burden. The presentation of choice sets was reverse-ordered in half of the surveys to reduce potential bias from the order of answering choice cards (Bridges et al., 2011).

7.2.4 Questionnaire administration

In the pilot study, participants completed the paper-based survey individually with the opportunity to seek clarity from the researcher if they were experiencing challenges with completion. Surveys took approximately 15 minutes to complete. Participants had the option of returning the survey on the day or taking it home and returning it in a stamped addressed envelope.

7.2.5 Statistical Analysis

Effects coding was used as this is advantageous over dummy coding where there is risk of misinterpretation through conflation of the constant term and

the utility of the reference level in the regression model (Bech & Gyrd-Hansen, 2005). The attribute levels of the opt-out alternative were set to zero. Choice data were analysed using a MNL model which has the same economic properties as the conditional logit model outlined by McFadden (1974) where utility involves an observable component (systematic utility) and unobservable (error) component. The MNL model assumes that error is identically and independently distributed according to a type 1 extreme-value distribution and thus can be used to identify patterns of choice across individuals and choice sets (Hauber et al., 2016). The MNL model was also applied to the unforced choice data using an alternative specific constant (Campbell & Erdem, 2019). Stata 16 (StataCorp. 2019) was used for analysis.

Attribute dominance was assessed which was where a participant always chose the predicted better level of one attribute across choice sets with this suggesting that the participant may not be trading across DCE attributes (Janssen et al., 2017). Task non-attendance, that is the extent to which participants payed attention to tasks, was also assessed through examining if any participant consistently selected the alternative in the same spatial position across choice sets (Janssen et al., 2017).

7.3 Results

Of the 56 people invited to participate in the study, 42 (75%) returned surveys. Of the 12 surveys taken home, seven (58.33%) were returned by post. Four participants were removed from analysis because they completed 4 or less choice sets. Therefore, thirty-eight participants were included in the analysis

and equal numbers (n=19) completed each survey block. This yielded 456 choice set observations.

7.3.1 Participant characteristics

The demographic characteristics of the 38 participants (17 male, 21 female) are provided in Table 7.4. The mean age was 52.29 (*SD* = 14.67) while the mean length of time since diagnosis was 23.48 (*SD* = 16.39). The sample included participants with varying healthcare payment methods. Fourteen participants (38%) had another chronic illness. Close to 60% (n=22) of participants had third level education while 5.3% (*n*=2) had no formal education. Just under three quarters of participants (*n*=27, 71.1%) had uncontrolled asthma.

Table 7.4 Characteristics of participants. Data presented as mean (SD) or n (%)

Characteristic	Pre-test (n= 16)	Pilot (n = 38)
Age (years)	52.38 (11.10)	52.29 (14.67)
Gender		
Male	9 (56.3%)	17 (44.7%)
Female	7 (43.8%)	21 (55.3%)
Education		
No formal education	2 (12.5%)	2 (5.3%)
Junior/intermediate cert. (or equivalent)	3 (18.8%)	7 (18.4%)
Leaving certificate (or level 5 equivalent)	2 (12.5%)	7 (18.4%)
Third level qualification	9 (56.3%)	22 (57.9%)
Length of asthma diagnosis (years)	22.97 (18.18)	23.48 (16.39)
Asthma control score (mean)	1.5 (1.2)	2.1 (1.3)
Payment for healthcare		
Payment with own money	7 (43.8%)	12 (32.4%)
Health insurance	8 (50%)	12 (32.4%)
GP visit card	0	3 (8.1%)
Medical card	5 (31.3%)	20 (54.1%)
Presence of other chronic illness	6 (37.5%)	14 (37.8%)

7.3.2 Choice Card Completion

All participants selected the superior alternative in the dominant choice task. Task non-attendance was not found, that is, no participant consistently selected the alternative in the same spatial position across choice sets. Eleven participants (28.9%) exhibited attribute dominance, always choosing the preferred level of their desired attribute. The attributes which were dominant were response to concerns (*n*=5), time to consultation (*n*=3), type of HCP (*n*=2) and method of communication (*n*=1).

7.3.3 Discrete Choice Modelling Analysis

Within the context of a forced choice between two service alternatives – Service A and Service B (see Table 7.1), the direction of preferences aligned with a priori expectations as detailed in Table 7.3. With effects coding, coefficients from the model are preference weights and *P* values indicate if the estimated coefficient is statistically different from the mean effect of the attribute (Hauber et al., 2016). The coefficients (preference weights) and 95% confidence intervals are reported in Table 7.5 and are presented graphically in Figure 7.1. The differences in preference weights between the most preferred level and the least preferred provide an estimate of the relative importance of an attribute and are included in Table 7.5. McFadden's pseudo R^2 was 0.273 which indicated good model fit (Hauber et al., 2016).

Among the six attributes, 'Response to Concerns' was the highest valued in terms of support to address an asthma flare-up with the highest relative importance value. Participants had a strong positive preference for a HCP who appears to listen while a HCP who appears not to fully listen was negatively

valued. The second most important attribute was ‘Time to consultation’. There was a positive preference for having a consultation on the same day as participants sought help compared to a negative preference for a consultation two days later. There was a trend of increasing preference across levels of ‘Time to consultation’ with a consultation the next day preferred to two days later.

The third most important attribute was ‘Knowledge of Patient’s Health’. Participants showed a preference for a HCP who knows them and has their medical notes relative to a HCP who neither knows them nor has their notes. The data also indicated a preference for a HCP with medical notes but no previous contact with the patient relative to a HCP with neither previous contact nor the patient’s notes. The fourth attribute in terms of relative importance was the ‘Healthcare Professional’ to consult with. Participants showed a strong preference for consulting with a respiratory doctor/consultant. They indicated a small but non-significant increase in preference to consult with a respiratory nurse compared to a GP, but both of these were significantly less valued compared to the respiratory doctor/consultant.

‘Method of Communication’ was fifth in order of importance among the six attributes. There was a preference for communicating face-to-face instead of over the phone. The least important attribute was ‘Having a written plan’ which was not significantly valued in the context of the other attributes and had a low relative importance value.

Table 7.5. Result of MNL models for forced choice

	Co-efficient	Standard Error	95% Confidence Interval		Relative Importance
			LB	UB	
Having a written plan					0.213
No	-0.106 (omitted level)	0.098	-0.298	0.085	
Yes	0.106	0.098	-0.085	0.298	
Healthcare professional					0.927
A general practitioner	-0.337** (omitted level)	0.124	-0.581	-0.094	
A respiratory nurse	-0.252*	0.119	-0.486	-0.019	
A respiratory doctor/consultant	0.590**	0.123	0.349	0.830	
Time to consultation					1.207
Two days later	-0.675** (omitted level)	0.139	-0.947	-0.402	
Next day	0.142	0.126	-0.106	0.390	
Same day	0.533**	0.127	0.283	0.782	
Method of communication					0.669
Phone call	-0.335** (omitted level)	0.077	-0.485	-0.184	
Face-to-face meeting	0.335**	0.077	0.184	0.485	
Knowledge of patient's health					1.036
No notes/previous contact	-0.573** (omitted level)	0.122	-0.812	-0.334	
Notes but no previous contact	0.110	0.119	-0.123	0.342	
Notes and previous contact	0.463**	0.152	0.166	0.761	
Response to concerns					1.533
Appears not to fully listen	-0.767** (omitted level)	0.086	-0.936	-0.597	
Appears to listen	0.767**	0.086	0.597	0.936	
ASC *service b*	0.037	0.131	-0.220	0.294	
AIC	479.397				
BIC	520.622				

** significant at p <0.01 *significant at p <0.05,. ASC = alternative specific constant. Values for the omitted level were calculated as the negative sum of the calculated levels and standard error was calculated as outlined in Hauber et al. (2016). Relative importance calculated as difference between most preferred and least preferred level.

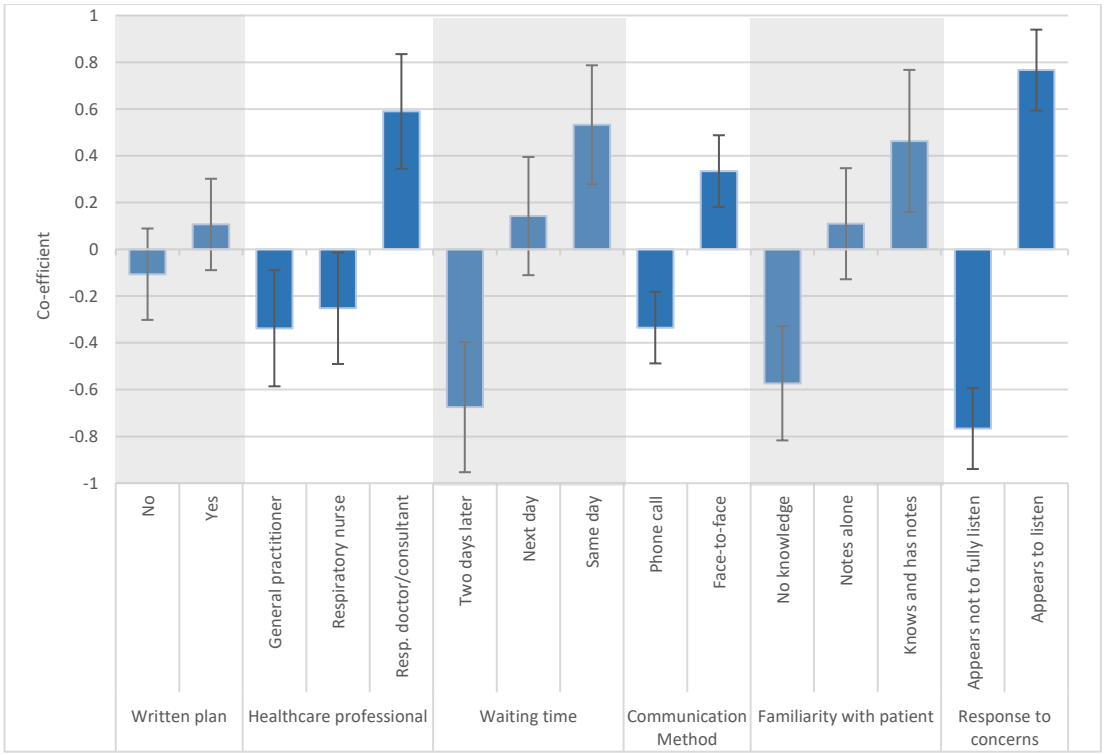


Figure 7.1 Coefficients for attribute levels and 95% confidence intervals in the forced choice MNL model.

The results for the dual response/unforced choice context are reported in Appendix 6.F. The same patterns of attribute preferences were found as in the forced MNL model. In the unforced choice context, participants said they would not use the preferred Service A or B in 69 instances (15.13%). This alternative had a significant negative co-efficient in the MNL (-1.036, *SE* = 0.158) indicating that participants had a strong preference to engage with the service alternatives rather than to opt-out of using these. Sixteen participants (42.1%) chose this option. Reasons for not using the service were due to unacceptable levels of attributes; most commonly the provider not fully listening, waiting until the next day or two days later for consultation, HCP not having notes/knowledge of patient and communication over phone.

7.3.4 Attribute ranking

Data from 28 participants who ranked attributes from 1-6 following completion of the choice cards, found that attributes ranked as most important (number 1) were time to consultation (25%, *n* = 7), response to concerns/listening (21.4%, *n* = 6), type of HCP (21.4%, *n* = 6), HCP's knowledge of patient's health (14.3%, *n* = 4), having a written plan (14.3%, *n* = 4) and method of communication (3.6%, *n* = 1). Having a written plan was most commonly ranked of least importance (50%, *n* = 14) followed by method of communication (32.1%, *n* = 9).

7.4 Discussion

To the best of this researcher's knowledge, this is the first DCE to measure the preferences of adults with asthma in the context of support for managing flare-ups. Using information on the preferences of people with asthma may help to enhance support for the management of asthma flare-ups and ultimately address issues of delayed access to healthcare services prior to ED usage and asthma-related deaths (Goeman et al., 2013; Langer et al., 2013). Primarily, this pilot study contributes to the refinement of the DCE instrument. However, findings of the pilot study have implications for health services though these findings should be interpreted with caution given the small sample size. The analysis indicates that having a provider that appears to listen was the most preferred attribute in this study. Recent qualitative studies have indicated that being listened to and feeling understood are important from the perspectives of people with asthma (Hannane et al., 2019; Lingner et al., 2017). This pilot DCE complements and extends these previous qualitative

findings, providing quantitative support that this feature is valued more than other features of support studied here.

Listening may be important from the patient perspective because it facilitates the identification and use of appropriate treatment. Shared-decision making regarding treatments has been found to increase adherence to treatments (Wilson et al., 2010). However, a lack of listening and responsiveness to breathlessness from the HCP may also contribute to a sense of injustice and negative affect from the patient's perspective, conflicting with human needs to be understood and respected (Hutchinson et al., 2018; Ng et al., 2012). The high value placed on listening in this DCE suggests that listening skills should be a key focus of training for HCPs dealing with people experiencing asthma flare-ups.

While consulting with a provider who knows the patient and has their notes was most valued, a provider with access to notes was also valued compared to having no notes or history on the patient. In the literature, continuity of care has been conceptualised with reference to the availability and use of past information about the patient's health (continuity of information); the ongoing relationship with providers (relational continuity); and the patient's experience of services as responsive to their needs, that is, continuity of clinical management (Haggerty et al., 2003). There has been debate regarding the extent to which continuity of information is sufficient to provide support for self-management in the absence of relational continuity such as when support is provided through non-localised telephone services, in this case for COPD (Hurst et al., 2010; Kielmann, Powell, et al., 2010). The data in this study

suggest that support from an unknown HCP may be acceptable provided the HCP has access to patient history/notes and that features such as timely access and listening are also present in the service. This finding supports the development of information sharing systems such as electronic health records in Ireland (Health Service Executive, 2015b). Further research with the DCE can help clarify the extent to which participants trade-off these attributes.

This study indicated a preference for providers having specialist knowledge with the respiratory doctor/consultant being most preferred. Specialist knowledge and medical expertise in managing changing symptoms have been valued in qualitative studies, as indicated in Chapters 4 and 5 of the thesis (Lawson et al., 2014; O'Connell et al., 2019). A review undertaken to inform integrated care in Ireland, recommends that specialist support be delivered through a specialist nurse working across a cluster of primary care practices (Savage et al., 2016). However, in this DCE study, participants expressed a strong preference for consulting with a respiratory doctor and no significant difference in preferences between the respiratory nurse and the GP in the context of support for an asthma flare-up. This finding requires further validation with a larger sample and exploration of the underlying factors influencing preferences. These HCPs may be perceived to contribute different elements of support with the respiratory nurse having specialist knowledge but the GP facilitating access to necessary medications to manage a flare-up. In Ireland, nurse prescribing is a developing area of practice to which participants may not be accustomed (Nursing and Midwifery Board of Ireland, 2018).

The method of communication, face-to-face being preferred to telephone, was important though somewhat less than the previously discussed attributes. While previous qualitative research in Chapter 4 suggested that face-to-face interaction is valued because care is felt to be more personal (O'Connell et al., 2019), cognitive interviews used to develop this DCE suggest that people valued physical assessments by the HCP. The cognitive interviews suggested that a video consultation was not valued and thus it was not included in the DCE. However, as a result of the COVID-19 pandemic, there has been an increase in the use of online/video consultations and increased guidance developed to achieve this as well as strategies to achieve elements of physical examination (Greenhalgh et al., 2020; Shaw et al., 2020). This may influence preferences for remote communication and it may be pertinent to include video consultations as a method of communication in future DCEs in this area.

Having a written plan was not valued as highly as other attributes in the pilot study. While written action plans are a recommended component of asthma support in Ireland and internationally (Chronic Conditions Working Group, 2017; GINA, 2019), they have not been shown to be effective in improving outcomes above standard care across studies (Gatheral et al., 2017; Villa-Roel et al., 2018). This study provides quantitative data to support the qualitative finding that patients do not value action plans highly compared to other aspects of support (Ring et al., 2015). However, a written plan may be important for a subgroup of people with asthma such as those who are newly diagnosed, as suggested in a qualitative study with people with asthma and HCPs (Ring et al., 2015). In this study, four participants (14%) ranked a written plan as most important suggesting a plan may be important for a subgroup of participants.

Alternatively, an action plan may be less valued because participants may have a lack of experience of using action plans as part of self-management. A DCE examining preferences for an SMS programme for epilepsy found that those who had experienced a similar intervention were more favourable of it in the DCE (Atkinson-Clark et al., 2018). Multi-level barriers exist to the usage of action plans in practice (Ring et al., 2015). Given recommendations to use asthma action plans both internationally and nationally in Ireland, further research is needed to understand how barriers can be addressed, incorporating the patient's perspective.

In terms of the future use of the DCE, this pilot study has provided parameter values which can be used to develop an efficient design for the full-scale DCE and to calculate the sample size required (de Bekker-Grob et al., 2015; Johnston et al., 2017). Furthermore, the pilot study suggests that unforced choice format may benefit from revision. Participants selected that they would not use their chosen service in a small proportion of DCE choice sets. Participants' reasons for choosing to opt-out in this study were concerned with unacceptable features of a service rather than having 'no treatment'. Thus, consideration should be given to narrowing the opt-out to a status quo option, that is a service alternative that they currently experience (Determann et al., 2019). Alternatively, it may be pertinent to remove the follow-up opt-out question in the interest of reducing cognitive burden.

7.4.1 Strengths and limitations

A strength of this study is the systematic approach to developing the DCE in accordance with good practice guidance and reporting of the development

process including the qualitative research that informed the DCE. A lack of transparency has been noted in DCE studies of preferences of people with chronic diseases (von Arx & Kjær, 2014) and there have been calls for greater reporting of the qualitative research methods used to develop DCEs (Coast et al., 2012; Vass et al., 2017). The design and administration of this DCE sought to counter potential biases on DCE data. Cognitive interviews informed the appropriate range of levels for attributes which helped avoid ceiling and floor effects, that is failing to detect the impact of an attribute because participants choose only levels at the upper or lower limit, respectively (Bridges et al., 2011). The order of choice sets was varied to reduce ordering effects and the use of overlap should have helped counter a tendency to answer based on the order of presentation of attributes (Kessels, 2016).

The sample size is a limitation of the pilot study with data collection curtailed due to the COVID-19 pandemic and access to clinics for research purposed prohibited from March 2020. A future study with the required sample size is needed to investigate if these trends hold. Nevertheless, the sample size in this study is within the acceptable range for a pilot study to inform sample size calculation for the main study (de Bekker-Grob et al., 2015). A calculation taking account of the DCE hypotheses has estimated that a minimum of 266 participants (forced choice MNL model, $\alpha = 0.05$, 80% power) would be necessary to estimate significant co-efficients for all attributes in the current design (de Bekker-Grob et al., 2015). The MNL model used in this analysis is limited in the way that it accounts for error and more advanced models have the capacity to capture correlation across multiple responses from the same participant and heterogeneity across participants which provide a more

accurate analysis of preferences (Hauber et al., 2016). With a larger sample size, subgroup analyses and models which account for heterogeneity such as the mixed multinomial logit model (MMNL) should be used (Lancsar et al., 2017).

The sample in this study were recruited through a specialist clinic and primarily includes people with moderate to severe asthma, of whom 71% had uncontrolled asthma. Therefore, these preferences may not hold for people with mild asthma or those with controlled asthma who may be predominantly supported by their GP. In addition, having access to a specialist clinic including specialist asthma doctors and nurses, may have influenced the preference for type of HCP in this study given that participants all had a previous visit to a specialist clinic. However, the sample provided good representation across a range of participant characteristics, including those with more than one chronic condition, and evidenced a relatively high response rate. The clinic also includes people living in rural and urban areas.

7.4.2 Conclusions

Information provided through the DCE on the relative importance of features of support during an asthma flare-up from the patient perspective can inform the features of services which should be implemented so that they are aligned with patients' preferences. The pilot study suggested that having a provider who fully listens to a person's concerns is the most important attribute of support in the context of an asthma flare-up which suggests a continued need for emphasis on communication skills in HCP training. Having a written plan was valued less than other attributes presented in the DCE which suggests a

need for further research to understand how implementation of action plans can be facilitated. Additional research is needed to explore differences in preferences across groups and individuals in order to understand how support can be tailored to meet the needs of people with asthma.

Chapter 8. Discussion

The overarching aim of the research presented in this thesis was to develop and pilot a discrete choice experiment (DCE) to measure the support preferences of people with chronic respiratory diseases in Ireland, ultimately focusing on individuals with asthma. In this final chapter of the thesis, an overall summary of the main findings is presented followed by a discussion on the research conducted and included in the previous chapters. While the studies presented in each of the chapters thus far have been discussed within the context of relevant literature, the discussion in this chapter aims to draw together the main findings with consideration to the broader literature. The discussion of findings in this chapter focuses on three areas: preferences for support in managing a flare-up; the importance of flexible unscheduled care pathways; and advancing SMS within the health system. The strengths and limitations of the research in this thesis are noted. Finally, recommendations for research, policy and practice are detailed before providing an overall conclusion to the thesis.

8.1 Summary of main thesis findings

Self-management support (SMS) frameworks have been developed for implementation in a number of countries, including Ireland, as part of healthcare policy reforms aimed at addressing the burden of chronic disease. Prior to this research, little was known about the processes which underpinned SMS framework development, implementation and evaluation plans. A document analysis of eight SMS frameworks highlighted gaps in patient

involvement with only two frameworks clearly reporting the involvement of people with chronic disease in framework development (Chapter 2). The findings of a qualitative study with leads of SMS framework implementation highlighted the limitation of inadequate patient input into policy development and implementation. The need to include the priorities and preferences of people with chronic conditions as key stakeholders in relation to SMS was identified from this qualitative study (Chapter 3).

To this end, steps were taken to design a DCE to elicit the preferences of people with chronic respiratory diseases. The findings from the thematic synthesis of qualitative studies on the SMS preferences of people with asthma and/or COPD (Chapter 4) revealed that their preferences centred around the 'types of support' with reference to education and psychological support, 'the support relationship' which highlighted the importance of communication and continuity with providers, and 'accessibility', that is, the need for timely access to support in a manner preferred by the individual. A notable research gap identified from this meta-synthesis was that only two of the fifteen studies had sampled people with asthma. The findings from a content analysis of the qualitative descriptive study with people with asthma or COPD (Chapter 5) concurred with the themes identified from the meta-synthesis of published qualitative research. However, regarding 'the support relationship', there was greater emphasis on the importance of the healthcare provider listening to the patient compared to other aspects of communication. In particular, 'being taken seriously' was highlighted as important for participants with asthma. In terms of accessibility, participants valued timely access to healthcare when

experiencing changes in symptoms indicative of an asthma flare-up. Some participants also identified cost as a barrier to accessing help.

Given the specific preferences of people with asthma and the paucity of recent research on support for asthma it was decided to focus the DCE on support in the context of experiencing an asthma flare-up (Chapter 7). The most valued attribute was 'response to concerns' where people with asthma appreciated a provider who appears to listen. Other preferences identified in the findings included shorter time to consultation (within same or next day); consulting with a provider who knows them/has their medical notes; consulting with a respiratory doctor/consultant; consulting face-to-face rather than over the phone. This pilot DCE study therefore makes a new contribution to knowledge on patient preferences for SMS regarding asthma flare-ups. This is an area that has received little research attention in the past and apart from a small number of qualitative studies, no studies have attempted to glean patient preferences using a DCE. The end-product of the thesis is a systematically developed DCE instrument which can be used to shed further light on asthma support preferences in future research. The findings gleaned from the research presented in this thesis have implications for healthcare policy and delivery in supporting patients to manage their asthma.

8.2 Discussion of main thesis findings

8.2.1 Preferences for support in managing an asthma flare-up

This research contributes to new knowledge having examined the relative importance of features of support in managing a flare-up in the DCE pilot study. The findings indicated that preferences included listening to the patient's

account, timely access, continuity of care, type of healthcare professional and method of communication.

The pilot DCE study indicated that having a provider that listens to the patient's account was the strongest preference for people with asthma experiencing symptoms of a flare-up. This finding adds to a growing body of qualitative research, highlighting that communication and listening to patients' concerns are important components of support, particularly in responding to breathlessness (Hannane et al., 2019; Hutchinson et al., 2018; Lingner et al., 2017). The person-provider relationship was found to be a key focus of support preferences in both the qualitative synthesis and qualitative descriptive study in the current research. The qualitative descriptive study particularly highlighted the importance of communication and listening to the patient's experience.

The emphasis placed on aspects of the relationship by people with asthma/COPD, and particularly the emphasis on being listened to, lends support to the hypothesis that a sense of relatedness is a basic psychological need that contributes to people's wellbeing, as indicated in Self-Determination Theory (Deci & Ryan, 2000; Ng et al., 2012). According to this theory, relatedness, as a basic psychological need means "feeling understood and cared for by others" (Ng et al., 2012). The need for HCPs to understand patients' concerns and take them seriously is inherent to advancing 'person-centred' SMS (Entwistle, Cribb, & Owens, 2018). These researchers highlighted the need to balance biomedical professional goals with responsiveness to patients' agendas thereby considering issues of importance

to people with chronic illness (Entwistle, Cribb, & Owens, 2018). The current research reinforces that listening skills need to be part of ongoing HCP training. Evidence-based training strategies such as experiential methods and lay trainers may be helpful to HCPs that are responding to people with asthma/COPD to build communication and empathy skills (Davies et al., 2018).

The findings of the pilot DCE provide an understanding of the relative importance of features of support in managing a flare-up of asthma. The attribute on HCP knowledge of the patient was the third most important attribute following time to consultation. This finding draws attention to discussions in the literature regarding the important aspects of continuity of care in managing chronic conditions (Meiqari et al., 2019). In response to a study that highlighted the importance of patient medical history/notes within the context of supporting continuous access to care in 'high risk' patients with COPD (Hurst et al., 2010), researchers have cautioned against losing sight of relational continuity between the patient and HCP over informational continuity (Kielmann, Powell, et al., 2010). Likewise, Hudson et al. (2019) argued that while informational continuity is important, this cannot be in isolation of relational continuity in order to achieve optimal patient outcomes and experiences. These researchers addressed continuity within the context of palliative care.

However, the literature has offered little differentiation between the value of relational and informational continuity of care in different contexts (Hudson et al., 2019; Kielmann, Powell, et al., 2010; Meiqari et al., 2019). The pilot DCE

study in this thesis contributes to new knowledge in this regard in the context of managing a flare-up of asthma. Being known by the HCP from previous interactions in addition to the HCP having notes was highest valued. However, a HCP having a patient's medical records (information continuity) was preferred to no records where there was no previous relationship between patient and provider. This finding highlights that it may be acceptable to ensure informational continuity in absence of relational continuity for support in assisting people in manging asthma flare-ups. Caution is warranted regarding the pilot DCE results. Future research with the DCE could clarify the extent to which people with asthma trade-off relational continuity with other features of support by incorporating more advanced techniques with a larger participant sample.

The value of the HCP having access to the patient's history in the pilot DCE, coupled with the suggestion of a shared electronic record in the qualitative study in Chapter 5, supports recommendations for the implementation of shared electronic records between primary and secondary care (Chronic Conditions Working Group, 2017). A health service strategy for electronic health records in Ireland aims to enhance HCP access to patient information and to facilitate the delivery of remote care (Health Service Executive, 2015b). However, progress in the implementation of electronic health records has been slow in Ireland compared to other countries. Elsewhere, for example in the Catalan health system, patients including those with COPD have reported benefits of electronic records in terms of information continuity across health settings (Waibel et al., 2016; Waibel et al., 2015). However, there have been challenges from the provider perspective and in a particular region where

information was only partially shared across health system levels (Waibel et al., 2016; Waibel et al., 2015). This highlights the importance of studying the implementation and optimal features of electronic medical records going forward. A further consideration is how information can be shared with out-of-hours service providers given that lack of prior patient information has been noted as a challenge in this component of unscheduled care (Drinkwater et al., 2013).

The relatively low value associated with an asthma action plan was an important finding of the DCE pilot study given that written asthma action plans have been recommended both internationally and nationally (Chronic Conditions Working Group, 2017; GINA, 2019). One hypothesis proposed in Chapter 7 is that asthma action plans may be more useful for specific groups such as those who are newly diagnosed, as suggested elsewhere (Ring et al., 2015). While it was not possible for this DCE pilot study to examine group differences, it was found that a number of participants rated the written plan highest despite the low importance of the attribute overall. Thus, a written plan may be important for a subgroup of people with asthma. Future research with the DCE could examine if individual variables are associated with preferences.

A second explanation for the lower value of the written action plan may be that preferences were influenced by a lack of participant experience of action plans. Research suggests that action plans are not well implemented in practice (Price et al., 2019). A study of preferences for an SMS programme for epilepsy found that those who had experience of group-based SMS interventions were more favourable towards their use (Atkinson-Clark et al., 2018). These

researchers suggested that people may need to be encouraged to use services that are new to them and ultimately may feel they are beneficial and valuable (Atkinson-Clark et al., 2018). The lower value of written action plans evidenced in the DCE indicates a need to examine how they can be better implemented including a focus on enhancing buy-in for patients and providers. Research on patient and provider perspectives of action plans suggests a cyclical relationship where the patient's perception of the usefulness of action plans may be influenced by the provider's perceptions and vice versa (Ring et al., 2015).

Lastly, the lower value associated with an action plan in the pilot DCE study may also be explained by challenges in the use of action plans by people with asthma when needed. People with asthma/COPD may find it challenging to enact their knowledge and skills for managing a flare-up due to anxiety and panic experienced when changes in their breathing occur (Korpershoek et al., 2016; Schmid-Mohler et al., 2019). Action plans are frequently not followed in accordance with guidelines by people with asthma (Cross et al., 2014). Furthermore, in previous research, where preferences for having an action plan were found, these preferences were related to having a brief written note on how to take medications (Haughney et al., 2007). A full action plan with information on identifying symptoms and associated management actions was least preferred. Thus, it may be that action plans, in their current format, are perceived as challenging to use in practice. As will be discussed in the following section, support at the time of experiencing flare-ups may aid people to use action plans and facilitate timely management of an exacerbation in the home/outpatient environment.

8.2.2 The importance of flexible unscheduled care pathways

The systematic review and thematic synthesis of qualitative studies reviewed in this thesis and the findings of the primary qualitative study in Chapters 4 and 5 respectively, highlighted a number of preferences relating to self-management support including the accessibility of support. In particular, the importance of access to services during potential flare-ups of asthma/COPD was highlighted. This involved prompt access to advice when experiencing changes in symptoms where people were unsure how to manage or sought reassurance regarding their actions. Within the synthesis, many studies involved frequent support and monitoring of symptoms as part of the interventions, though some participants found frequent scheduled appointments burdensome and preferred flexibility in contacting a HCP as needed. Participants in the qualitative study reported in Chapter 5 sought timely access to support if concerns about their health decline and its management arose. Of note, none of the participants included in the qualitative study reported involvement in interventions with regular weekly/daily support. The emphasis on access to unscheduled support raises questions about the extent to which routine and scheduled support programmes and education can help people manage exacerbations by themselves, as has been previously discussed in relation to asthma and COPD (Korpershoek et al., 2016; Lingner et al., 2017).

Flexible unscheduled support pathways, as indicated by the preferences of people with asthma/COPD in this research, may contribute to a reduction in exacerbations and unscheduled emergency department admissions (Alí et al., 2019; Jain et al., 2014). A panel of respiratory care providers in a Delphi study

agreed that self-management of exacerbations may be appropriate for some COPD patients but not all (Korpershoek et al., 2017). Two retrospective cohort studies on integrated care programmes incorporating unscheduled support found reductions in severe exacerbations of asthma and COPD, though the researchers noted that further randomised and controlled studies are needed (Alí et al., 2019; Jain et al., 2014). Both interventions involved education on regular management such as inhaler technique while the study by Jain et al. (2014) also incorporated an action plan. However, these studies provided unscheduled care pathways through 'on-demand' visits or 'priority consultations' with outpatient providers and 24/7 telephone support. The researchers in one study suggested that the latter components contributed to the reduction in severe exacerbations, with a greater number of outpatient appointments reported in the integrated care group compared to a comparison group (Alí et al., 2019).

Unscheduled support may also aid the use of action plans. In one of the integrated care interventions described above, it was reported that 24/7 telephone support aided patients to initiate and successfully use an action plan (Jain et al., 2014). Recent reviews indicate that there is insufficient evidence on the role of ongoing or unscheduled support in the use of action plans (Gatheral et al., 2017; Howcroft et al., 2016). However, the results of the current research, from the patient perspective, combined with relevant literature suggest that it may be pertinent to develop flexible unscheduled care pathways. These flexible services could support people in the timely management of an exacerbation or flare up of their condition, including the use of an action plan.

The findings of the current research suggest a need to reconsider the meaning of ‘unscheduled’ care for people with asthma/COPD. This would involve a shift from hospital care to flexible unscheduled care pathways which would involve services such as out-of-hours clinics and telephone support lines that could support people in deciding on appropriate courses of action to manage a flare-up (Drinkwater et al., 2013; Turnbull et al., 2019). Researchers have previously cautioned against reduced healthcare utilisation as a target of self-management support initiatives (Gately et al., 2007; Nolte & Knai, 2015; Van de Velde et al., 2019). From a HCP perspective, unscheduled care use is seen as a necessary component of support for people with chronic diseases when experiencing exacerbations (Drinkwater et al., 2013). As a result, HCPs report tensions when working with policies that seek to reduce unscheduled care use (Drinkwater et al., 2013). The current findings provide a case for creating flexible care pathways where people can access timely advice on managing symptoms which may increase some forms of unscheduled healthcare utilisation but ultimately reduce ED admissions and hospitalisation. However, it may be that flexible support services should be targeted at high-risk individuals as part of a risk stratification approach (Bourbeau & Echevarria, 2020; Savage et al., 2016). The qualitative research presented in Chapters 4 and 5 indicated that preferences for support may differ according to severity of the condition. Furthermore, the above studies reporting on interventions involving unscheduled care focused on severe asthma and COPD (Alí et al., 2019; Jain et al., 2014).

8.2.3 Advancing self-management support within the health system

This thesis began by examining national/state level frameworks on SMS for chronic disease. The frameworks endorsed systems approaches to SMS including actions at the level of patients, HCPs, organisations, the health system and wider environment. The study of preferences of people with asthma/COPD in this thesis highlight preferences relevant to broader systems elements such as continuity of care and timely access to services. Addressing patients' preferences in this regard would reflect a broader health systems approach to SMS, thereby going beyond the predominant patient education approach to SMS in healthcare which is viewed as limited without consideration for whole system approaches (Kennedy, Rogers, Chew-Graham, et al., 2014; Mills et al., 2017). In addition to the previously discussed system elements of continuity of care and timely access, the findings from the qualitative descriptive study highlight the importance of addressing healthcare costs for people with asthma/COPD. The studies presented in Chapter 2 and Chapter 3 identified strategies including greater stakeholder involvement which may help advance the implementation of support for self-management across a health system through SMS frameworks.

The qualitative study reported in Chapter 5 identified the need to address healthcare costs incurred by patients to provide an equitable system of SMS. Cost was identified as a reason to delay help-seeking by some people with asthma/COPD, though further research is needed to understand the impact of out-of-pocket spending for this group in Ireland. Across OECD countries, out-of-pocket costs have contributed to unmet healthcare need with an average of 10% of consultations skipped due to cost (OECD, 2017). Close to 60% of

Ireland's population incur out-of-pocket costs for healthcare, without access to the GP visit card scheme or a medical card which facilitates access to most healthcare for free (Department of Health, 2019a). Furthermore, neither asthma nor COPD are currently included within the long-term illnesses scheme in Ireland and thus, the costs of medications are not covered for many people with these conditions (Health Service Executive, 2019).

The finding regarding the challenge of costs for patients reported in Chapter 5 provides support for the implementation of Sláintecare – Ireland's planned universal system of healthcare (Oireachtas Committee on the Future of Healthcare, 2017). However, there has been doubt as to whether political support can be sustained to facilitate its 10-year implementation given early delays in achieving milestones (Burke et al., 2018; Connolly & Wren, 2019). Furthermore, its implementation may not resolve out-of-pocket healthcare costs for patients. Callander et al. (2019) cautioned against assuming that a universal health system results in financial risk protection for patients. They recommended ongoing assessment of out-of-pocket healthcare expenditure incurred by patients in countries with universal healthcare.

This thesis focused on understanding preferences of people with asthma/COPD with the aim of adapting healthcare so that it can be better aligned with their preferences. In the qualitative study with SMS framework leads from various countries, presented in Chapter 3 of this thesis, the involvement of people with chronic conditions was recognised as contributing to the success of SMS implementation. Framework leads acknowledged that more was needed to ensure that the 'patient voice' was reflected in SMS

frameworks. However, the need for multiple stakeholder perspectives when considering strategies for healthcare delivery was also noted. In terms of health system reform for SMS, processes of public and patient involvement (PPI) and co-design may help negotiate viable solutions across stakeholders (Araújo-Soares et al., 2019). However, it is imperative to ensure that the patient involvement is meaningful in the context of other stakeholders (Greenhalgh et al., 2019). In the cross-country analysis of SMS frameworks reported in Chapter 2, where there was patient involvement, the number of patients was often limited in comparison to other stakeholders. This may result in the marginalisation of the patient perspective.

In addition to stakeholder input and consultation, additional strategies for advancing the implementation of system-wide SMS frameworks were identified in Chapters 2 and 3 of the thesis. Advancing this policy level is seen as key to reducing the gap between research and practice (Nilsen et al., 2013). While inter-country consensus processes have been used to identify strategic directions for SMS (Mills et al., 2017), the research reported in this thesis extends this literature through providing an account of the implementation of SMS frameworks at national/state-levels. Facilitators of implementation identified in Chapter 3 reflect the strategic actions arising from the inter-country consensus process, as reported by Mills et al. (2017). This includes ensuring SMS is a strategic priority across policy levels; developing policies in conjunction with a range of stakeholders; the creation of champions; and communication platforms to facilitate collaboration across stakeholders (Mills et al., 2017). The qualitative study in Chapter 3 identified further detail pertaining to useful courses of action. For example, sustained mechanisms of

communication across different stakeholders including senior leaders were suggested to keep SMS on the agenda. In addition, it was highlighted how platforms which facilitate champions to communicate with other stakeholders were necessary to enable champions to build support. The importance of tailoring messages for different stakeholder groups was also highlighted, with this acknowledged as key factor in communicating with policymakers (Cairney & Kwiatkowski, 2017). Lessons identified in Chapter 3 contribute to early research on the implementation of national and state-level frameworks on SMS for chronic disease. Policy recommendations are further outlined in Section 8.4.2.

8.3 Strengths and limitations

8.3.1 Strengths

Research on the policy processes relating to SMS frameworks was guided by relevant frameworks and theory. A framework is valuable when making cross-country comparisons to enhance reliability (Cacace et al., 2013). Firstly, in terms of the document analysis, the health policy triangle (Walt & Gilson, 1994) provided a means to comprehensively explore elements of health policy (Buse et al., 2012). When qualitatively exploring the process of SMS framework implementation, the Multiple Streams Approach to Policy Implementation (Howlett, 2018) provided a useful lens which provided strength to the findings of the study while also allowing novel findings to emerge.

Social desirability had the potential to influence studies in this thesis, though its effects may have been reduced by research factors. In the interviews with qualitative key informants reported in Chapter 3, there was potential for

participants to represent their work in a favourable light. However, participants provided open accounts of implementation. The bias was likely reduced through anonymity, with frameworks not being named in the analysis. Furthermore, across all the primary studies in the thesis, the researcher was not previously known to participants and in the case of people with asthma/COPD, the researcher had not been involved in their care. This may have contributed to participants providing honest accounts of their experience and preferences.

Furthermore, efforts were made to enhance the trustworthiness of qualitative studies in this thesis with regard to credibility, confirmability, dependability and transferability (Lincoln & Guba, 1985). Strategies to enhance credibility included systematic in-depth field work and conscientious analysis of data, developing rapport with participants, prolonged engagement with the data, searching for alternative patterns and reporting complexities as well as reflexively considering the impact of the researcher and research design on the findings (Bradshaw et al., 2017; Patton, 2015; Probst & Berenson, 2013). Additional actions to strengthen trustworthiness involved purposeful sampling and rich participant description (transferability) and use of direct quotations and an audit trail enhance confirmability and dependability (Patton, 2015).

Representativeness of the sample of people with chronic respiratory disease may be considered a strength across the research overall, though there were limitations within each study. Overall 74 people living with chronic respiratory disease (63 with a diagnosis of asthma, 10 with COPD and one with both asthma and COPD) participated across qualitative interviews, cognitive

interviews and the DCE pilot study. Participants in the qualitative descriptive study were predominantly female which may have influenced the findings given some potential gender differences related to perceptions of breathlessness and help-seeking (Chhabra & Chhabra, 2011; Thompson et al., 2016). Furthermore, recruitment through support organisations may have identified people who were more active in the management of their asthma/COPD. However, samples for the DCE, including cognitive interviews and the pilot study, provided balanced gender representation and represented many relevant demographic groups including those with multimorbidity. The samples recruited in specialist asthma clinics represented people with moderate to severe asthma and frequently uncontrolled asthma may not be representative of preferences of the broader asthma population who are managed through primary care.

The systematic process of developing the DCE in line with good practice guidance was a methodological contribution of this thesis (Bridges et al., 2011; Coast et al., 2012). This included systematic approaches to synthesizing secondary literature, collecting primary qualitative data and cognitive interviews, with the lattermost being rarely reported in the literature (Vass et al., 2017). Cognitive interviews provided information to refine the instrument in a number of ways including the acceptable number of attributes and appropriate range of levels. This helped avoid ceiling and floor effects where extreme attribute levels could have unduly influenced the DCE results (Bridges et al., 2011). In addition, recent literature was used to inform decisions regarding elements of the design with the aim of balancing response and statistical efficiency. This included the use of overlap in choice sets (having no

more than five attributes vary across alternatives in the same choice set) (Jonker et al., 2019); a dual response format to provide an opt-out (Muhlbacher & Johnson, 2016), and use of an efficient design which was blocked to reduce cognitive burden.

8.3.2 Limitations

Lack of detail on the health system context of SMS frameworks is a limitation of the research reported in Chapter 3 of this thesis where the contributing health systems were not identified. Previous analyses have highlighted how courses of action for chronic disease management are influenced by the underlying organisation of health systems (Nolte & Knai, 2015; Nolte et al., 2008) and information on context aids comparison of policies (Cacace et al., 2013; Marmor et al., 2005).

The focus on the concept of SMS may also have precluded the identification of literature which could provide insights relevant to the aims of this thesis. The study of SMS frameworks was limited to frameworks with a specific focus on self-management. However, countries may include components of self-management within chronic disease management policies and may not use this terminology. These broader policies were not represented in this research and may have useful contributions. A similar limitation applies to literature on preferences of people with asthma/COPD. While variations on the term ‘self-management’ were included in the search, it may have failed to detect some documents which were relevant but using alternative terminology such as symptom management and empowerment.

There were also limitations relating to the DCE. In terms of design, JMP software was useful for achieving many desired aspects of the design and while it seeks to maximise efficiency, it did not provide a measure of d-efficiency in order to evaluate the statistical efficiency of the design (Hensher et al., 2015). The small sample size in the DCE pilot study suggests caution in the findings and precluded more advanced analysis which could help understand potential heterogeneity in support preferences. COVID-19 impacted data collection such that the pilot study sample was smaller than anticipated. Lastly, PPI was not used in the development of this DCE. There is recent evidence showing that researchers are beginning to use PPI to develop DCEs (Shields et al., 2020). While challenges are acknowledged such as management of PPI member expectations and appropriate training on DCE methods to facilitate member contribution, it is suggested that PPI has the potential to further enhance the design, survey materials and recruitment for the DCE (Shields et al., 2020).

8.4 Implications and recommendations

A number of recommendations for future research, policy and practice are drawn from the thesis findings. These are summarised in Table 8.1.

Table 8.1. Implications for research, policy and practice

Research recommendations • Conduct final DCE adequately powered (*n* > 266) to examine if trends hold when accounting for heterogeneity and explore the influence of individual characteristics on preferences. • Compare qualitative processes of attribute development for DCEs with a view to developing clearer guidance on optimal approaches. • Engage in research and co-design with diverse stakeholders, including HCPs and people with chronic disease, in order to identify suitable features of interventions.
Policy Implications • SMS frameworks should include detail on actors involved and detail on processes of involvement as well as contexts of health systems that influence policy to enable other countries to learn and consider applicability to their health system. • To implement an SMS framework, communication mechanisms across stakeholders are needed. These would need to facilitate contact with senior leaders as well as contact between healthcare professionals. • Engage in patient involvement in policy development and implementation of SMS frameworks. • Conduct and report on evaluations of the implementation of SMS against key performance indicators • Address barriers to support for people with asthma/COPD at system level through providing universal healthcare and shared electronic records.
Practice Implications • HCPs should aim to tailor SMS to the individual, both the content and the delivery approach • HCP training should continually focus on communication and use evidence of its importance from the patient perspective to motivate HCPs • Facilitate timely access to advice when experiencing an asthma flare-up from a provider who is skilled in responding to concerns and knows the patient's history

8.4.1 Future research recommendations

The DCE should be conducted with a powered sample (*n* > 266), as per de Bekker-Grob et al. (2015), to confirm preferences and provide strengthened evidence to policy makers and planners regarding the type of support valued in managing an asthma flare-up. Future analyses should take account of preference and scale heterogeneity (Hauber et al., 2016). Further analyses are also needed to compare preferences across groups such as a comparison of preferences for an action plan between those who are newly diagnosed compared to those with longer diagnoses. Further research is also needed to

examine the impact of multimorbidity given challenges associated with managing multiple chronic conditions (Liddy et al., 2014). This is important given the prevalence of multimorbidity in asthma and COPD, which was noted as 60% in a study conducted within the Irish primary context (O'Kelly et al., 2011). Future studies using a DCE design could compare differences in preferences between those who do and do not experience multimorbidity.

While the process of DCE development aimed to be systematic, it was challenging to identify and select attributes from the qualitative data. An iterative qualitative process was used as outlined by Coast and colleagues (Coast et al., 2012; Coast & Horrocks, 2007). Structured approaches such as the nominal group technique or ranking exercises may be helpful in identifying and selecting attributes (Hiligsmann et al., 2013; Ratcliffe et al., 2017). There is no clear guidance on which of these approaches to attribute development is optimal and there is limited data comparing the effectiveness of different approaches (Coast et al., 2012; Hiligsmann et al., 2013). Furthermore, recent literature has highlighted the benefits of PPI in DCE development including the process of attribute development (Shields et al., 2020). Future comparisons are needed to advance guidance on attribute development.

This thesis aimed to identify the preferences of people with asthma/COPD in order to inform healthcare so that it can be further aligned with their needs and values. In this way, it contributes to the ethos of person-centred support for self-management that advocates reduced focus on the biomedical model and supporting people with chronic illness in what matters to them (Entwistle, Cribb, & Owens, 2018). However, continued research is required in order to

understand how to balance ethical and practical considerations of person-centred approaches to SMS (Entwistle, Cribb, Watt, et al., 2018; Franklin et al., 2019). The perspectives of other stakeholders including HCPs and the use of co-design approaches may help identify how SMS can be implemented throughout a health system.

8.4.2 Policy Implications

The commonalities in the overall processes of SMS implementation highlight the potential for health systems to learn from each other in terms of implementing SMS. However, the document analysis in Chapter 2 highlighted areas of information that were not detailed in framework documents. It is recommended that detail on the actors and how they were involved is included in future policy documents relating to SMS. Furthermore, greater detail on the contexts of the health system which influence choice of SMS strategies may be helpful in understanding if approaches in one health system are appropriate for application in another (Cacace et al., 2013; Marmor et al., 2005).

Lessons for implementing SMS frameworks were drawn up based on the accounts of key informants in Chapter 3 (detailed in Table 3.4). Suggestions for communication across stakeholders included sustained mechanisms of communication with senior leaders and a communication platform across HCPs. This aimed to provide an opportunity for champions to build support for SMS strategies as well as facilitating a collaborative approach to implementation where HCPs views could be taken into account and plans adapted accordingly. Further research is needed to clarify optimal systems of communication across stakeholders.

Further lessons for implementing SMS policy included embedding SMS frameworks in policy and healthcare reform standards at various levels of the health system; adapting plans to fit with other policy drivers; identifying and measuring performance indicators across a health system while allowing for local flexibility in approaches. Measuring performance involves greater clarity on the deliverables of SMS while another component is embedding deliverables in evaluation systems so that it becomes easier to glean pertinent data. Lastly, greater involvement of people with lived experience of chronic illness is needed. While the value of patient involvement is accepted, the appropriate means to achieve this are less clear. As discussed earlier, it is imperative to ensure that involvement is done in a meaningful way and is not tokenistic. The process needs to be tailored to the population and aims of the project (Greenhalgh et al., 2019).

The implementation of other interlinked health policies and strategies is needed to facilitate SMS. Given that cost was a potential deterrent to help-seeking during flare-ups in the Irish population, it is recommended to advance the implementation of Sláintecare in Ireland with care free at the point of delivery (Burke et al., 2018; Oireachtas Committee on the Future of Healthcare, 2017). Plans for the implementation of electronic health records should also receive attention in light of the value placed on HCPs having up-to-date patient history (Health Service Executive, 2015b).

8.4.3 Practice implications

The qualitative studies of support for managing asthma/COPD found that people desired support with many elements that contribute to their wellbeing

ranging from medical support, education, psychological support and instrumental support. The range of supports desired as part of SMS highlight how SMS needs to be tailored to the individual (Dwarswaard et al., 2016; Trappenburg et al., 2013). Where group-based approaches are taken, it is important to facilitate the identification and management of individual challenges. Evidence of variation in preferences for some forms of support such as peer support, highlight that practitioners should consult with the patient to understand their preferences in order to collaboratively decide how support should be provided.

Communication was a key focus of the studies which suggests the need for emphasis on communication as part of HCP training. HCPs should be aware of the value patients place on listening to their experience and the finding that some people feel that measures such as peak flow are sometimes not representative of their experience. A clear rational for changing practices (Lawn & Schoo, 2010) along with evidence based training approaches are needed to enhance interpersonal skills (Davies et al., 2018). However, the broader system factors such as time for consultation also need to be addressed in order to facilitate the application of these skills (Davies et al., 2018).

The findings of the DCE provide evidence for models of care which include unscheduled support pathways for managing asthma-flare-ups. Interventions providing unscheduled support that have reduced severe exacerbations and hospitalisation have been noted to be cost-effective due to saved hospital resources (Jain et al., 2014). In Ireland, a quality improvement initiative for

COPD known as the COPD Collaborative has enhanced management of acute exacerbations through consultant-led, multidisciplinary teams (MacDonnell et al., 2019). The initial work of the collaborative focused on the hospital setting and results have shown promise including reduced wait time to see a specialist and reduction in length of stay (MacDonnell et al., 2019). However, the future aim of the collaborative is to shift the model to primary care with the objective of reducing hospitalisation (MacDonnell et al., 2019). It was also suggested that the model may be applicable to other chronic diseases. The findings of this thesis support such an approach for people with asthma as well as people with COPD. The findings from the pilot DCE study suggest quick access (same day or next day) to a provider who listens and knows the patient from previous contact or at least has their medical notes would be valued features of the service for people with asthma.

8.5 Overall conclusion

SMS has the potential to reduce the burden of chronic disease with people playing an active role in manging their wellbeing. However, implementation of SMS initiatives is challenging. This thesis made a number of contributions to understanding how implementation of SMS could be enhanced.

A study of national/state-level SMS frameworks and accounts of their implementation by framework leads identified the challenge of sustaining support for the implementation of SMS frameworks. Lessons to facilitate long-term support were identified. Enhanced communication across stakeholders was important in both securing high-level buy-in but also to adapt and ensure the suitability of plans from HCP and patient perspectives. The input of people

with chronic disease was particularly important in guiding the implementation of SMS frameworks with the potential to enhance services so that they better meet patient need.

This thesis subsequently examined the preferences of people with asthma and COPD in qualitative studies. Preferences were identified in relation to the patient-provider relationship which involved empathetic communication and continuity over time. Preferences for support with many different aspects of management suggested a need to tailor support to what matters to the person with chronic disease. Access to support when managing an exacerbation or flare-up was important in addition to routine support and suggests that even with long-term SMS, people would benefit from systems of acute access which could enhance early management of flare-ups/exacerbations.

Lastly, the relative importance of support features for managing an asthma flare-up were examined quantitatively through a DCE pilot study to provide information to assist prioritising features of support for implementation. A HCP who fully listens to their concerns was the most important of these, followed by timely access and continuity of care. These preferences can be used to inform unscheduled support interventions. The DCE has been systematically developed, pre-tested and piloted and can be further used to explore preferences for support in managing an asthma flare-up.

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