‘It’s a nice thing to do but…’: Exploring the methods and impact of Patient and Public Involvement (PPI) in trials.

A thesis submitted to the National University of Ireland, Cork for the degree of Doctor of Philosophy in the School of Public Health

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About the title

The title of this thesis comes from the countless conversations that I have had with other researchers over the course of this PhD research. During these conversations, when I would explain that my research is focused on PPI, I was always met with a similar reaction- 'Oh yeah, I mean, I think PPI is a really nice thing to do but…'. The 'but' would then be followed with various reasons why they hadn't incorporated any PPI into their research to date – '…I don't understand why research funders are so set on it' or '…do we really know that it makes a difference to our research' or '…nobody seems to know how to actually do it properly'.

Over the last three years, I haven't encountered anyone that didn't think PPI was a nice thing to do. But at the same time, I have only met a handful of people that were confident that they could do it properly and that it would definitively lead to better quality research. Now that research funders, ethics committees and academic journals require PPI, I believe we need to lessen the distance between the semi-skeptics and the fully fledged PPI-ers. To do this, we must address their concerns. We need evidence on the methods and impact of PPI.

And that in essence is what this research is all about!

This is for my Granny, Maureen Whelan (née O'Keefe), the eternal teacher who has inspired us all to pursue lifelong learning x

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List of Abbreviations

APEASE	Affordability, Practicability, Effectiveness and cost-effectiveness, Acceptability, Side effects and safety, Equity
BCT	Behaviour Change Technique
BCTv1	Behaviour Change Taxonomy version1
BMJ	British Medical Journal
CAB	Community Advisory Board
CINAHL	Cumulative Index to Nursing and Allied Health Literature
COREQ	Consolidated criteria for reporting qualitative research
CREC	Clinical Research Ethics Committee
CTU	Clinical Trial Unit
DCU	Dublin City University
DNS	Diabetes Nurse Specialist
DRS	Diabetic Retinopathy Screening
ENTREQ	Enhancing transparency in reporting the synthesis of qualitative research
GP	General Practitioner
GRAMMS	Good Reporting of a Mixed Methods Study
GRIPP	Guidance for Reporting Involvement of Patients and Public
HCP	Healthcare professional
HRB	Health Research Board
HRB TMRN	Health Research Board Trial Methodology Research Network
IDEAs	Improving Diabetes Eye-screening Attendance
IRC	Irish Research Council

LIVE	LIstening to the Voice of Experience
NHS	National Health Service
NIHR	National Institute for Health Research
NUIG	National University of Ireland Galway
PCORI	Patient-Centered Outcomes Research Institute
PICO	Population, Intervention, Comparison, Outcome
PICOS	Population, Intervention, Comparison, Outcome, Study type
PPI	Patient and Public Involvement
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RCSI	Royal College of Surgeons Ireland
RCT	Randomised Controlled Trial
SAB	Study Advisory Board
SMS	Short Message Service
SPIDER	Sample, Phenomenon of Interest, Design, Evaluation, Research type
SPSS	Statistics Package for the Social Sciences
SREC	Social Research Ethics Committee
SWAT	Study Within A Trial
TCD	Trinity College Dublin
TRUST	Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism Trial
UCC	University College Cork
UCD	University College Dublin
UK	United Kingdom
UL	University of Limerick

Declaration

I declare that this thesis 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:

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Date:

_____28.09.20_____

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To Ber- 'Where would we be without Ber?' You would not believe how many times I have said that over the last three years. Thanks so much for all your help and thanks for always checking in at exactly the right time! Thank you!

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To Martin. You have been my biggest supporter right from the beginning of this journey. I know that we have had to put certain aspects of our lives on hold so that I could finish this out. Thank you for accepting this PhD as part of both of our lives, for never putting

any pressure on me and for endlessly never entertaining any of my self-doubts- ‘Shur, you’re well able’! I am so grateful for everything you do for us. Thank you!

Thesis Abstract

Background and Aims

Patient and Public Involvement (PPI), defined as research carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them, is increasingly recognized as an essential component of health research. The rationale for PPI is based on a moral argument where the people whose lives are most affected by research should have a say in what is researched and how it is carried out, and a pragmatic argument that PPI can improve research quality. Although PPI is now required by many research funders, academic journals, and ethics committees, progress to achieve greater involvement has been patchy and slow. There is a lack of clarity on how to conduct 'strong' PPI and on why PPI should be used. Research is needed on suitable PPI methodologies and on the impact of PPI if we are to develop a shared understanding of what works, when, how and why. Therefore, the overarching aim of this thesis was to contribute to the evidence on the methods and impact of PPI by exploring PPI contributors' experiences and contributions at the design, conduct and dissemination stages of trials.

Methods

At the design stage, two Studies Within A Trial (SWAT) were conducted within the intervention development phase of the Improving Diabetes Eye-screening Attendance (IDEAs) pilot trial. The first used a mixed methods convergent design to compare people with diabetes and healthcare professionals' experiences of taking part in three different

types of consensus meetings to inform intervention development and assess whether their experiences differed according to group composition. The second used a qualitative design to compare people with diabetes and healthcare professionals' contributions to the intervention content and assess whether their contributions differed according to group composition. At the conduct stage, a systematic review and narrative synthesis was conducted on trial researchers' perceptions of the impact of PPI on trial retention. At the dissemination stage, a mixed methods SWAT, including an embedded randomised trial, was conducted within the Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism (TRUST) trial to identify, develop, and evaluate a patient preferred method of receiving trial results.

Results

Involving PPI contributors simultaneously with other stakeholders led to a perceived lack of common ground where both stakeholders felt reluctant to fully express their opinions. It also led to conflicting opinions which were difficult to incorporate into the intervention being developed. Researchers perceived PPI to have a positive impact on trial retention as it helped trial researchers to foster a trusting relationship and improve communication with trial participants. PPI was also perceived to improve trial retention by ensuring the trial location was suitable and accessible and enabling researchers to establish cultural appropriateness by ensuring that community customs, norms and social activities were considered in the research design. Although, PPI contributors were involved in the development of the trial result letter, the results of the embedded randomised trial

suggested that PPI did not make a difference to participants' understanding of trial results.

Conclusions

This research shows that although there are a wide variety of methods used to involve PPI contributors, the method used can have an important influence on the impact of involvement. The results suggest that it may be more suitable and useful to involve PPI contributors separately rather than simultaneously with other stakeholder groups. This finding may assist researchers and PPI contributors in designing and conducting more meaningful and effective involvement activities. This research found that PPI can influence the research process by creating and fostering trust between researchers and participants and PPI contributors can help researchers to communicate more effectively with research participants. Although, the results suggest that PPI did not make a difference to participants' understanding of results, suggestions for how researchers should approach future evaluations of the methods and impact of PPI have been put forward. This research paves the way forward for building an evidence base for PPI to ensure that a shared understanding of what works, when, how and why is developed among researchers, patients, members of the public and research funders.

1. Introduction

1.1 Introduction

Patient and Public Involvement (PPI) is increasingly recognized as an essential component of health research. In the UK, INVOLVE, the national advisory group supporting active public involvement in health services, public health and social care research defines PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to' 'about' or 'for' them' (1).

The rationale for PPI is based on two lines of argument. Firstly, a moral and ethical argument of 'nothing about us without us' where the people whose lives are most affected by research should have a say in what is researched and how it is done (2-4). Secondly, a pragmatic argument that PPI can improve the quality, relevance, and uptake of research (2, 5-8). It has been suggested that this can happen through: influencing research priorities; helping solve ethical dilemmas; helping with recruitment strategies; influencing how data is collected, analysed and interpreted to ensure a patient and public perspective; and ensuring communication and dissemination of outputs is in a language and format that is accessible to patients and the public (9).

In recent years, several countries have been working to embed PPI principles and practices in health and social care research, and PPI is now required by many research funders, academic journals and ethics committees (3, 10). Despite this changing environment of PPI in health research and the potential for PPI to have a positive impact, progress to achieve greater involvement is 'patchy and slow' (11).

PPI is a complex activity; there are a wide variety of involvement tasks and activities, as well as a wide range of methods used to involve PPI contributors. Evidence on what works, when, how and why is lacking (9). Although it is acknowledged that different PPI methodologies have an important role in shaping the impact of PPI (12), current reports on suitable and effective PPI methodologies are insufficient with many researchers arguing that PPI is too complex to be evaluated as 'it depends' on too many different factors (13-15).

Similarly, current reports on the impact of PPI are 'piecemeal and inconclusive' and are largely based on researchers' reflections on the impact of working with PPI contributors (9, 16-18). Although these ad hoc and anecdotal reports allow us to develop an initial understanding of the potential impact of PPI, they can often conflate the aims of PPI with its achievements and seldom report any negative impacts (19, 20). Research funders that are contingent on having 'strong' PPI without clarity on how to conduct 'strong' PPI or on why PPI should be used can tempt researchers to exaggerate the scope of PPI in grant applications in order to obtain funding leading to tokenistic involvement. Research is needed to develop a credible and robust evidence base on the methods and impact of PPI.

1.2 Aim

This thesis aims to contribute to the evidence on the methods and impact of PPI by exploring PPI contributors' experiences and contributions at the design, conduct and dissemination stages of trials.

1.3 Objectives

1. To identify a suitable and effective way to involve multiple stakeholders in research.
    - Compare people with diabetes and healthcare professionals' (HCPs) experiences of taking part in different types of consensus meetings to inform intervention development (Chapter 3).
    - Identify and compare people with diabetes and HCPs' contributions during different types of consensus meetings to inform intervention development (Chapter 4).
2. To explore the impact of PPI on the research process.
    - Identify and compare people with diabetes and HCPs' contributions during different types of consensus meetings to inform intervention development (Chapter 4).

- Systematically review trial researchers' perceptions of the impact of PPI on participant retention in randomised controlled trials (RCTs) (Chapter 5).

- Investigate methods of disseminating trial findings to participants by using a PPI approach to identify, develop and evaluate a patient based approach to receiving trial results (Chapter 6).

1.4 Thesis outline

This thesis contains seven chapters. This first chapter provides an overview of the structure of the thesis. Chapter 2 draws on a review of the literature to describe the current role of PPI in health research and presents the rationale for generating evidence on the methods and impact of PPI. Chapters 3-6 correspond to the aim and objectives outlined above (see **Figure 1.1**). Chapter 7 provides an overall discussion of the main findings, the strengths and limitations of the thesis, the implications and suggestions for future research. This chapter also includes a reflection on PPI during the COVID-19 outbreak and a reflection on the INVOLVE definition of involvement.

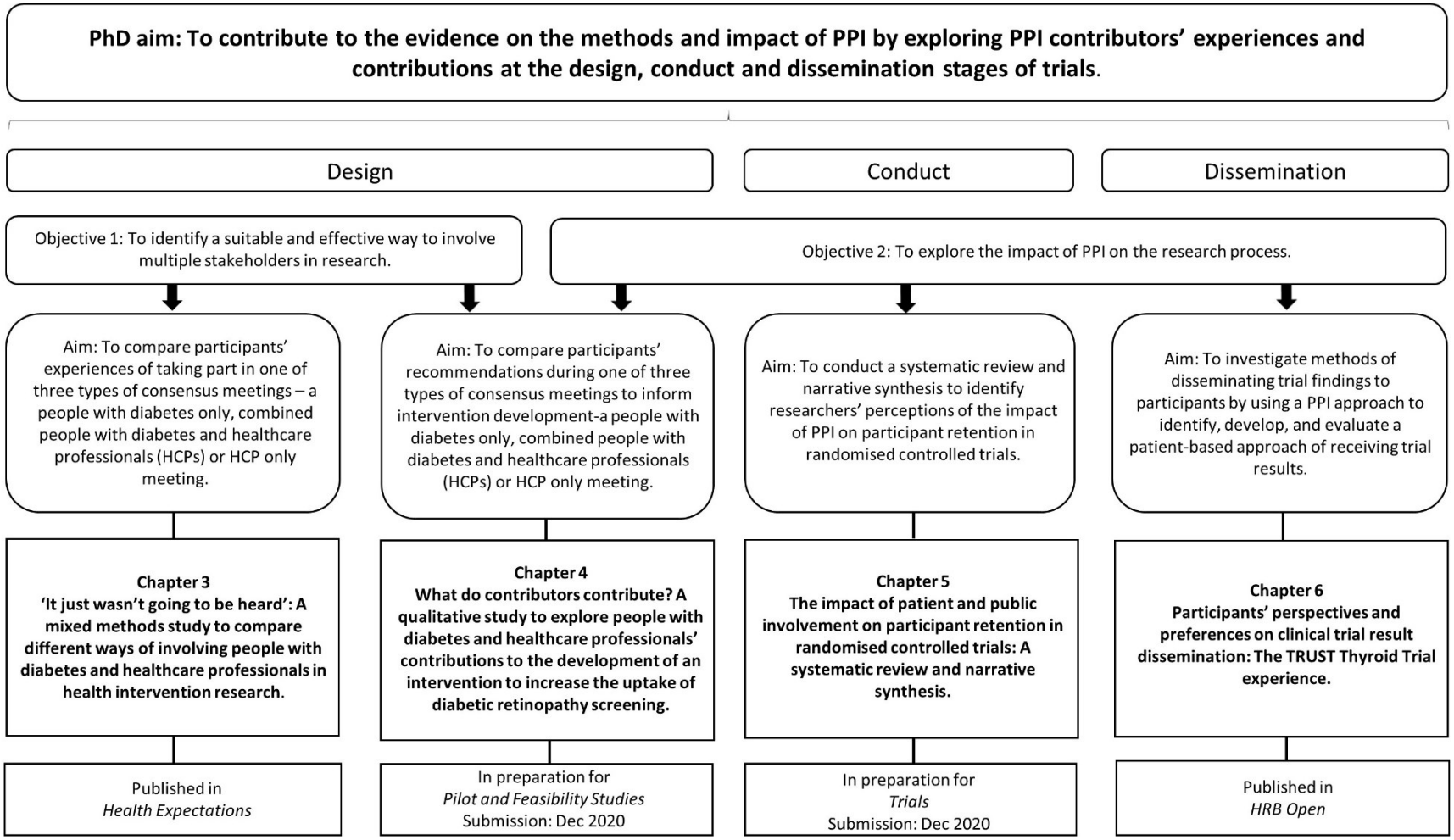

Figure 1.1: Overview of thesis including aim and objectives

2. Background

2.1 Overview

This chapter presents a brief overview of the current role of PPI in health research. First, PPI is defined and the rationale for PPI is described as well as the wide range of PPI approaches and methods. Second, the changing environment for PPI in health research is discussed. Third, the potential positive impacts of PPI are outlined along with the numerous and persistent challenges to PPI. Finally, the need for robust evidence on the methods and impact of PPI is presented.

2.2. Patient and Public Involvement (PPI)

2.2.1 Definition of PPI

For this thesis, I have adopted the INVOLVE definition of PPI. INVOLVE is the national advisory group supporting active public involvement in health services, public health, and social care research in the UK. INVOLVE define PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them (1). In working with this definition, I have taken the most widely accepted and inclusive definitions of 'patient' and 'public'. The term 'patient' is used to refer specifically to those who have experience of disease or illness (21). The term 'public' encompasses all those associated with the use of health care including patients and potential patients, people who use health and social services; informal carers; parents/guardians; disabled people; members of the public who are potential recipients of health promotion programmes, public health programmes and social service interventions; and organisations that represent people who use services [6]. The terms 'patient' and public' are being used to

denote roles in a specific situation, not categories of people, and are not mutually exclusive, as many individuals fulfil many roles, often at the same time (22).

The term 'involvement' is intended to mean the *active* involvement of patients and members of the public in health research projects and in research organisations. Patients and members of the public can be actively involved by contributing to and/or making decisions about what research is conducted, how the research is carried out and how it is disseminated.

PPI is distinct from patient and public participation in research which relates to the *passive* involvement of patients and members of the public, where they are recruited by researchers to become study participants or subjects and their data is collected, analyzed and published as study results. PPI is also distinct from patient and/or public engagement which similarly reflects a *passive* involvement of patients and members of the public, where researchers aim to raise awareness of research amongst patients and members of the public by disseminating research results, sharing knowledge, or engaging in one-way communication.

Values and principles of involvement

INVOLVE highlights six values that should be carefully considered and implemented when conducting PPI: respect, support, transparency, responsiveness, diversity and accountability (23). These values and principles along with principles in practice have been summarised in **Table 2.1** below.

Table 2.1: Summary of INVOLVE's values and principles for public involvement in research

Values	Summary principles	Principles in Practice
Respect	Researchers, research organisations and the public respect one another's roles and perspectives	• Public members' skills, knowledge and experience are respected • The knowledge and experience of researchers and others involved in administering or managing research skills are respected • Public members are included as key partners of research • Public members are involved from the outset • Public members' contributions to the research are recognised
Support	Researchers, research organisations and the public have access to practical and organisational support to involve and be involved	• Public members have access to learning and development to support their involvement in research • Researchers and others have access to learning and development to support public involvement in research • There is flexibility to support public involvement -public members' expenses are covered, and they are informed in advance if payment will be offered for their time • Infrastructure within research organisations enables and supports public involvement in research
Transparency	Researchers, research organisations and the public are clear and open about the aims for and scope of the involvement in research	• Researchers and others involved in the research openly discuss with public members the purpose, scope, and expectations in advance of their involvement in the research • Researchers provide clear information to public members about their role and their input • Public members are open about their ability to contribute

Responsiveness	Researchers and research organisations actively respond to the input of public members involved in research	• Public members, researchers and others contribute to collaborative decision-making • Researchers and research organisations are committed to public involvement and are willing to act on the input of the public • Public members commit to their involvement in research and are willing to contribute to the research
Fairness of opportunity	Researchers and research organisations ensure that public involvement in research is open to individuals and communities without discrimination	• Public members, researchers and others understand and sign up to the principles of equality, diversity and inclusion as defined in the Equalities Act 2010 • Researchers and research organisations ensure that public involvement opportunities are accessible to all • Information is presented in accessible and alternative formats and written in plain English
Accountability	Researchers, research organisations and the public are accountable for their involvement in research and to people affected by the research	• Researchers and research organisations have policies in place for the governance of public involvement in research and public accountability • Researchers and research organisations are accountable to public members involved in the research • Public members are accountable to researchers, research organisations and others for their involvement • Researchers, research organisations and public members assess the impact of public involvement in the research

2.2.2 Rationale for PPI

In the literature and policy discourse, PPI is justified by two general lines of argument. The first of these is an ethical or moral argument (2, 3). This argument incorporates ideas concerning democracy and rights, citizenship, power distribution, accountability, and empowerment. As part of this, a commonly cited argument is that as citizens and taxpayers, members of the public have the right to influence research that is being funded through public monies and that might have an impact on their health status (3, 4). This includes how research is designed and undertaken and how research findings are disseminated and implemented once a study is complete. The slogan 'nothing about us without us', which is believed to be over five centuries old, encapsulates this argument (24). This rights-based argument is also seen as a means of empowering minority and disadvantaged groups in society (7, 25), and so, many authors have argued that careful consideration should be given to the appropriateness of methodologies used to involve these 'seldom heard' groups (26-28).

The second line of argument is a pragmatic or consequentialist argument (2, 14, 29), where PPI, by bringing a real-world and lived-experience perspective, has the potential to improve the quality, relevance and impact of health research (2) via a number of mechanisms: increasing its relevance to patients; improving recruitment and retention rates of research participants; extending the range of people represented in research studies; and improving dissemination of findings beyond academic audiences (10). Previous authors have provided anecdotal accounts of how PPI can positively contribute to research by suggesting relevant research questions and outcomes, ensuring that

consent forms and information sheets are user-friendly, and assisting with the recruitment of participants, data collection, data interpretation, and dissemination (5-8).

2.2.3 PPI approaches and methods

PPI is a complex activity and there are a wide variety of approaches to involvement. These approaches vary depending on the theoretical model of involvement employed, the role of PPI contributors, the duration of involvement, the activities in which PPI contributors are involved, the specific methods used to involve and the relevant stage of the research process.

2.2.3.1 Theoretical models of involvement

PPI is a continuum from research with no involvement through to research that is initiated, undertaken and controlled by patients and members of the public (25). The number of components, levels or categories within this continuum varies depending on the theoretical model employed.

In the UK and Ireland, the theoretical model of PPI most frequently employed is the 'levels of involvement' model. This model was originally put forward by Boote, Telford and Cooper (7) and describes three levels of PPI: consultation, collaboration and user control. *Consultation* includes types of involvement that allow the researcher to obtain representatives' views. At this level, what the representatives say can be influential, but they have no power to ensure the researcher acts on their views (7). Consultation is

largely focused on feedback, for example, asking representatives to review research protocols, participant information sheets and drafts of published papers. Consultation can be implemented at all stages of the research process using a variety of methods and on a range of scales; for example, drawing on the views of a small group of representatives through a focus group or a large group through administration of a questionnaire (7, 30-32).

Collaboration involves an ongoing partnership between researchers and PPI contributors where contributors have more ownership of the research and can, at least in theory, contribute more directly to the direction of the research (7). Contributors may, for example, sit on a steering group for a research project, and help guide the project from its early planning stages through to dissemination. However, the exact nature of the collaboration differs between research projects.

User controlled (also referred to as consumer-controlled) is research that is actively controlled, directed and managed by service users and their service user organisations (7). Service users decide on the issues and questions to be looked at, as well as the way the research is designed, undertaken and disseminated (30). Researchers become involved at the request of the service users themselves; or consumer organisations commission research into a topic of interest to them (7). This type of research requires a strong commitment on the part of service users and is the least common of the three 'levels of involvement' (32).

The ‘levels of involvement’ model is a condensed version of Arnsteins ‘ladder of participation’ which has been a touchstone for policy makers and practitioners promoting user involvement for over 40 years (**see Figure 2.1**). Each rung on Arnsteins’ ladder represents increasing degrees of participation: from non-participation of manipulation and therapy; through to the tokenism of informing, consulting, placating, to citizen power through partnership, delegated power and citizen control (33). Arnsteins’ model frames citizen participation as an overt struggle for power between public sector managers and public activists and community members. This struggle continues to have some resonance but fails to engage with the complexity and nuances of PPI. These complexities have given rise to reinterpretations of the model including Wilcox’s five-rung ladder (34), Burn’s ladder of citizen empowerment which attempts to incorporate degrees of participation and quality of engagement (35) and Choguills’ adaptation for use in developing countries (36). Despite different interpretations, refinements and revisions to Arnstein’s model over the years, they all retain an important common feature, a ‘hierarchal approach’ with ‘citizen control’ portrayed as the ideal form of involvement.

While these different theoretical models include different conceptualizations of involvement, they notably all fail to capture the complexities of involvement such as the diversity of actors, the importance of process as well as outcome, and the integration of a systematic approach to engagement and feedback (37). Since its inception, the ‘levels of involvement’ model has been widely adopted and promoted by INVOLVE. In more recent times, however, INVOLVE have encouraged researchers to view involvement in

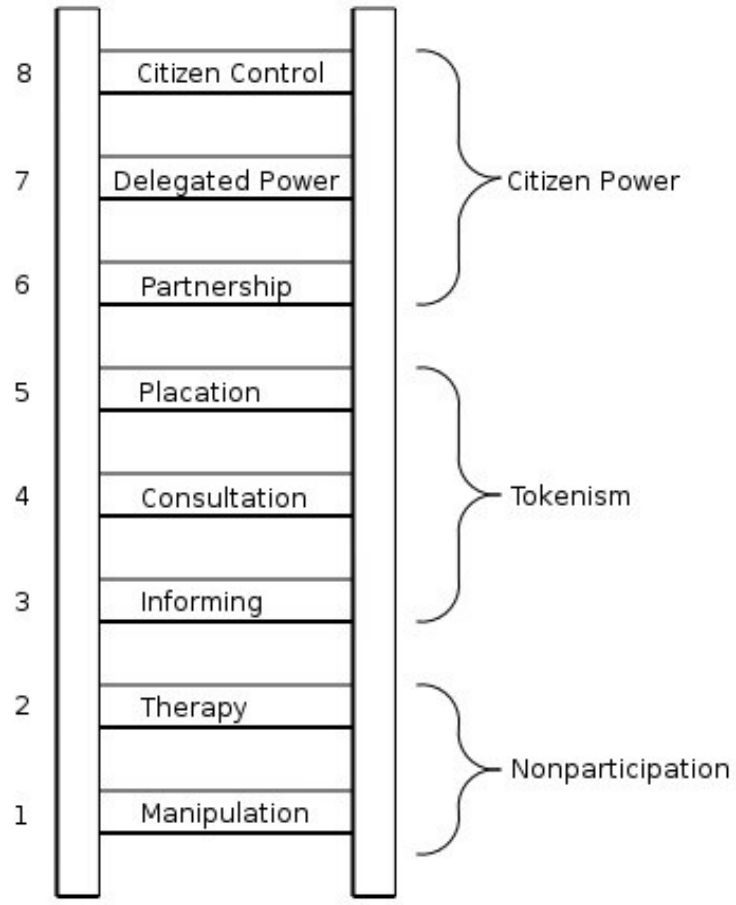

Figure 2.1: Arnsteins' ladder of citizen engagement

terms of 'approaches' to involvement rather than 'levels'. This shift in thinking encourages researchers and PPI contributors to recognize the complexities of involvement. It also encourages researchers to understand that the boundaries between categories are not so clear cut and research projects may combine two or three levels of involvement (32). For the purpose of this thesis, I have chosen to adopt a combination of consultation and collaboration approaches where wider groups of PPI contributors

will be consulted on specific aspects of each study and individual PPI contributors will be collaborators throughout the research.

2.2.3.2 The role of PPI contributors

PPI contributors' roles vary from managerial roles (involvement in the set-up and day-to-day running of the project), oversight roles (involvement in determining the direction of the research), and responsive roles (involvement guided by researchers) (9, 38, 39). Crocker et al. (2017) identified a range of distinct roles that may be played by individual PPI contributors at different stages in a research study (40):

- The expert in lived experience- able to consider the acceptability and feasibility of proposals for the target population, having lived through the experience under study
- The creative outsider- able to think 'outside the box' by bringing a fresh perspective
- The free challenger- able to challenge researchers without fear of consequences
- The bridger- able to make research more relevant and accessible by bridging the gap between researchers and the public, including patients
- The motivator- helping to highlight the importance of a piece of research as a motivation for engagement
- The passive presence- where just the presence of a PPI contributor has an influence on how researchers think

2.2.3.3 The duration of the involvement

The duration, frequency and regularity of patient and public involvement varies across research projects and programmes (9, 38, 41). Involvement may be ad hoc (drawing on PPI at intervals as required), or long-term (spanning the duration of the project), (9, 18, 38, 42). Although, long-term involvement across the research cycle has been rarely reported on in the literature (41, 43).

2.2.3.4 Involvement tasks and activities

There are a wide range of tasks and activities in which PPI has been reported, these often vary depending on the stage of the research. Ball et al, provide an overview of the wide range of PPI tasks and activities that are evident in the literature (9). Some examples of tasks and activities during the research preparation and design phase include identifying, generating and prioritising research topics or questions, providing input into funding decisions, contributing to the development of research proposals, advising on the development of surveys and interview guides, scope and search strategy for reviews, feasibility of conducting research in real-world settings, cultural issues that may need to be considered, sampling, ethical issues and patient information materials (39, 44-47).

Examples during the study conduct stage include advising on recruitment and retention issues, actively engaging in participant recruitment, contributing to the conduct of literature reviews, collecting data from participants by conducting interviews, administering surveys and facilitating focus groups, contributing to data analysis tasks and helping researchers to identify key findings (39, 44, 47-49).

Examples at the dissemination and translation stage include contributing to drafting journal articles, reports, summaries (including lay summaries) and press releases, participation in the release of results and publications and determining avenues to share findings (38, 46, 48, 50, 51).

2.2.3.5 The specific methods used to involve

PPI contributors can be involved through diverse methods of involvement. Some examples highlighted in the literature include advisory group meetings, PPI group meetings (both face-to-face and virtual), expert workshops, working collaboratively with the research team, surveys, interviews, focus groups, consensus meetings, discussion forums, patient panels, use of facilitation tools (e.g. World Café and Dotmocracy) social media, online discussion forums, structured priority-setting exercises, ad-hoc advice via PPI panels, sitting on funding panels and grant review committees and corresponding and reviewing documents via email (9). The number of individuals involved can vary greatly, for example an advisory board usually has between one and five service users, whereas priority setting exercises can involve hundreds or even thousands (41).

2.2.3.6 Stages of the research process

PPI can be conducted at any stage of the research process from priority setting and drafting study protocols right through to conducting the study, interpreting the results and communicating and disseminating research findings (See **Figure 2.2** below) (41, 46-48, 50, 52-55). Shippee et al. (2015) conducted a systematic review on 202 articles relating to PPI in biomedical and health services research and identified that PPI was

conducted in three key phases of the research cycle: the preparatory phase, the execution phase and the translational phase. According to their proposed framework, each phase comprises several distinct stages. The preparatory phase involves patients and/ or the public in addressing the question of what to research through two stages: agenda setting and contributions to preparing or reviewing funding applications. The study execution phase includes PPI in four stages: study design and procedures, recruitment and participation, data collection and data analysis. The translational phase consists of post-analysis activities in three stages: dissemination, implementation and evaluation (53). Some frameworks cover similar stages and phases to those outlined by Shippee et al., while others focus on specific parts of the research cycle or organise stages where contributions can take place through an alternative lens (9). For example, Ray and Miller (2017) categorise PPI according to: what the scope of the research where involvement takes place is (e.g. for defining and prioritising a topic of research questions and hypotheses, defining an intervention, specifying outcomes to be measured); project methods (i.e. whether PPI contributors are involved in research design, implementing research methods, recruitment); and interpretation (analysis, making sense of the findings, synthesis, anticipating alternative interpretation or controversy)(56).

There are variations in the degree to which PPI is conducted and reported across the different stages of the research cycle (9). For example, PPI is more frequently reported in the set-up and conduct stages of the research process than in the data collection, data analysis, dissemination and translation phases (39, 48, 53, 57) and there is a lack of PPI in funding decisions and in the evaluation of research (9, 53, 57).

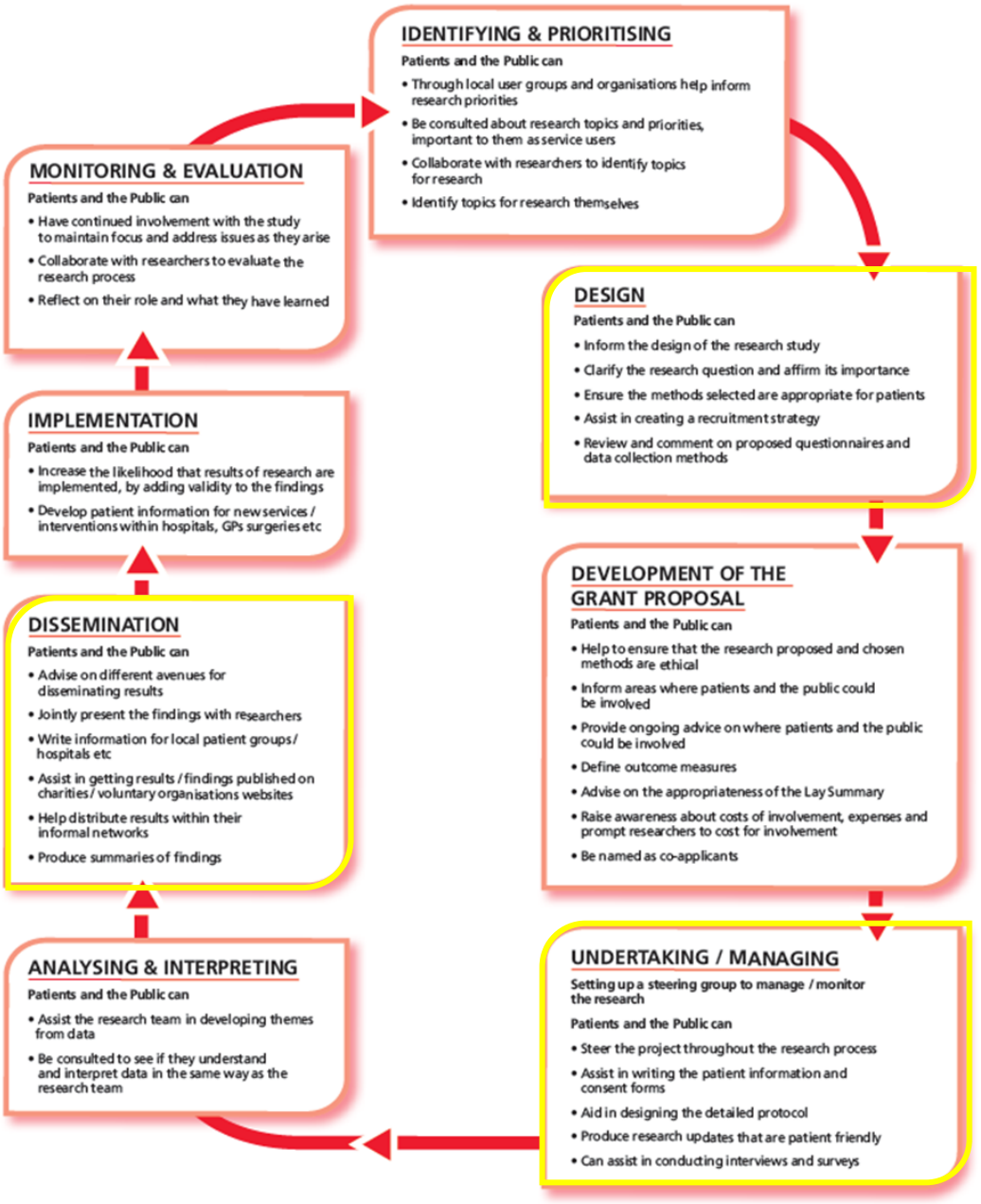

Figure 2.2: PPI at different stages of the research process *(58).*

This thesis explores PPI contributors' experiences and contributions at three distinct stages of the trial process: trial design, conduct and dissemination. The corresponding stages of the research process have been highlighted in yellow in **Figure 2.2.** However,

each of the studies presented in this thesis has had PPI involvement at different stages. This ranges from the study design stage right through to dissemination. Further details on PPI involvement in the different stages are presented throughout the following chapters in this thesis.

2.2.4 The changing environment for PPI in health research

In recent years, several countries have been working to embed PPI principles and practices in health and social care research, and PPI is now required by many research funders, academic journals and ethics committees (3, 10).

In the UK, this movement has been led by INVOLVE, which was originally set up in 1996 and is now integrated with the National Institute for Health Research (NIHR). In the US, this work is being carried out by the Patient-Centered Outcomes Research Institute (PCORI) and in Canada, the Strategy for Patient-Oriented Research (SPOR) has embedded PPI within the Canadian Institutes for Health Research funding calls.

In January 2016, Irelands' primary health research funder, the Health Research Board (HRB) launched its 2016-2020 strategy which referred to PPI as a core principle and contained the commitment to strengthen and develop PPI within the HRB and in HRB supported projects and programs (59). It was the first state funding body to formally launch an implementation plan for PPI. The implementation plan included asking all researchers to provide details in their research funding applications of any public involvement in the design, conduct or dissemination of their study, introducing public reviews of some its research funding applications and partnering with the Irish Research

Council (IRC) to launch a funding call which specifically aimed to support and promote PPI within Higher Education Institutions in Ireland-PPI Ignite (60). Five institutions were successful in the PPI Ignite call including: Trinity College Dublin (TCD), National University of Ireland Galway (NUIG), University of Limerick (UL), University College Dublin (UCD), and Dublin City University (DCU). The overarching aim of PPI Ignite was to build capacity in institutions that would provide researchers with the support they need to involve patients and the public in their research and convey this in their grant applications (61). In May 2020, the HRB and IRC opened a call for the establishment of a formal National PPI network. The overarching aim of the National PPI Network is to support and further build capacity for high quality PPI in health research throughout Ireland through a network that serves its members and benefits the wider community (62). The five PPI IGNITE institutions and two new partner institutions: University College Cork (UCC) and the Royal College of Surgeons Ireland (RCSI) prepared and submitted a joint application. If successful, the national PPI network will be established in March 2021.

The Irish Research Council (IRC) have also made significant efforts to embed PPI within Irish research. Since 2007, it has supported 'Campus Engage', a national platform funded by the Higher Education Authority which aims to promote civic engagement activities in Irish higher education (63). Although its focus is not primarily on PPI, its widespread promotion of engaged research and active citizenship has created a supportive environment for the advancement of PPI. As well as co-funding the PPI IGNITE and National PPI Network calls with the HRB, the IRC is also in the process of developing protocols to include PPI as a requirement in its research funding applications and this is likely to be rolled out shortly.

Academic journals have also begun to embed PPI in health research. For example, in 2014, the British Medical Journal (BMJ) launched its patient partnership strategy, seeking 'to promote patient partnership by walking the talk' (64, 65). The partnership strategy was informed by an international patient advisory panel and launched several innovative editorial practices, including patient peer review and patient co-production of educational articles. In 2015, as part of the patient partnership strategy, journals in the BMJ's portfolio began requiring authors to include a PPI statement in their academic publications. If patients were not involved in the research, authors must clearly state their reasons for the omission.

Research ethics committees also stipulate that members of the public be involved in research design and study conduct (64). However, this practice varies by organisation and country. In the UK, most ethical review boards now require PPI to be considered in the development of ethical approval applications (64). While research ethics committees in Ireland have not formally adopted this approach, one of the key objectives proposed in the National PPI network application which was recently submitted to the HRB/IRC is to embed PPI in institutional policies and structures including University Ethics committees (62).

2.2.5 The potential positive impacts of PPI

Current reports on the impact of PPI in health research have been described as 'piecemeal and inconclusive' and are largely based on researchers' reflections on the impact of working with PPI contributors (16-19). These accounts are ad hoc and

anecdotal and can conflate the aims of PPI with its achievements, and seldom report any negative impacts (19, 20). Some studies have conducted more robust processes of assessing impact. For example, gathering views of PPI contributors and academic researchers via interviews, pre and post involvement questionnaires and focus groups (16, 66). However, most of these studies have asked simple questions about whether involvement makes a difference and do not specifically evaluate the impact of PPI on particular aspects of the research process, or contain any evidence of impact (67). For example, researchers may report that involving PPI contributors helped to ensure that study materials were more understandable and accessible to members of the public but will not provide any details of the improvements that were made or contain any evidence of impact. Nevertheless, these reports have allowed us to develop an initial understanding of the potential impact of PPI. These impacts can be classified into three main categories: perceived impacts on the research process, impacts on researchers and impacts on PPI contributors.

2.2.5.1 Perceived impact on the research process

According to qualitative studies on the perceived impact of PPI in trials, researchers and PPI contributors believe that PPI can help to improve the relevance of research by ensuring that research funds are appropriately prioritised and that the evidence that research produces is of interest to patients and members of the public (44). It has also been suggested that PPI can improve the acceptability and accessibility of research by improving the clarity of participant information, removing jargon and making it more salient to potential participants (26, 68-70). A paper presenting researchers' reflections

of participatory action research with young injecting drug users highlighted that PPI has the potential to improve recruitment rates and facilitate more representative sampling as it can help researchers to access 'hard to reach' populations through PPI networks (71). The same paper also suggested that PPI can shorten the timeframe of research by improving the design of study protocols and expediting ethical approval (71). PPI has also been suggested to improve research dissemination. For example, a multi-method evaluation of the impact of consumer involvement in the London Primary Care Studies Programme reported that PPI increased the impact of research by broadening the opportunities for dissemination (72).

2.2.5.2 Impact on researchers

A qualitative case study of researchers' experiences of user involvement reported that PPI helped them to understand the views and experiences of research participants and helped them to connect to the 'real world' (73). Researchers have also reflected on how PPI allowed that to understand participants' cultures which gave them greater respect and helped them to develop a good rapport with the community (71, 74, 75). Researchers have also described gaining new insights into their research areas which helped to challenge their assumptions (75). A questionnaire study of researchers' and PPI contributors' perceptions of PPI impact during one randomised controlled trial highlighted that researchers' felt PPI had enabled them to develop new research ideas and focus on issues that were important to the community they were researching (76). Researchers have also reported that PPI helped them to develop new skills and find new

ways of working. Some researchers reported gaining facilitation, communication and conflict resolution skills and provided support and advice to PPI contributors (71, 75).

2.2.5.3 Impact on PPI contributors

A systematic review of the conceptualisation, measurement, impact and outcomes of PPI in health and social care research classified the impacts on PPI contributors into three main areas: personal benefits, impact on their level of knowledge and impact on their level of skill (77).

Papers reporting PPI contributors' reflections report personal benefits including feeling empowered (78, 79), listened to and valued (69, 80), more positive (81, 82), more confident (69, 81, 82) and feeling a sense of fulfillment and satisfaction (83-85). Service users reflections on being involved in a research advisory group experienced a sense of mutual support from being part of a team (83). Another study reporting PPI contributors' reflections reported that they appreciated the social interaction with others (84). Contributors have also felt they had given something back and had made a difference (69, 86, 87). Impacts on their level of knowledge include reports of gaining access to better information about their condition, and having the opportunity to exchange and compare this information with others (69, 83). This, in turn, allowed them to better manage their condition and solve related problems (88, 89). PPI contributors have also reported gaining a greater understanding of the research process (4, 69, 81) which led to increased levels of trust in the research process (90, 91). Impact on their level of skills

include gaining skills in research methodology (4, 92), public speaking (69, 83) and listening to other people's perspectives (81, 93).

2.2.6 Challenges to PPI

Despite the changing environment of PPI in health research and the potential for PPI to have a positive impact, progress to achieve greater involvement is 'patchy and slow' (11).

In 2018, Price et al. conducted a review of PPI statements in BMJ journals before and after the PPI reporting requirement was introduced (64). In the year before the PPI reporting requirement, 0.5% of research articles reported PPI activity. In the year following the requirement, 11% of research articles reported PPI activity. Although the new requirement was associated with an increase in reporting PPI, the numbers are much lower than the journal's target (65). The review also found that PPI statements varied greatly in quality and content, with some articles demonstrating a lack of awareness and understanding of the concept of PPI (64).

The challenges to effective involvement are numerous and persistent. Ball et al. have categorized these challenges into four types (9). These include systemic challenges in the research system, challenges related to the capacity of individuals to engage, administrative and management challenges, and challenges related to culture values and attitudes. An overview of the different types of challenges and examples of each type are presented in **Table 2.2** below.

Table 2.2: Challenges to involving patients and the public in research as summarized by Ball et al. (9)

	Type of challenge	**Examples**
1	Systemic challenges in the research system, related to the governance of PPI in research and to knowledge management.	• inappropriate financial resourcing of PPI activities • poor reporting on PPI processes and limited monitoring and evaluation • insufficient coordination and shared learning between different PPI bodies • limited patient and public awareness about engagement needs and opportunities
2	Challenges related to the capacity of individuals to engage	• lack of experience, knowledge, skills or confidence • lack of access to training • health and wellbeing related challenges such as inability to travel to research meetings.
3	Administrative and management challenges	• limited administrative support for implementing PPI processes such as organising meetings and timely payment of contributors • lack of in-built mechanisms for giving feedback to PPI contributors
4	Challenges related to culture, values and attitudes	• tokenism • dismissive attitudes of some researchers • challenges to managing expectations of PPI contributors about the nature and scale of engagement • managing power dynamics in teams

2.3 The need for evidence on the methods and impact of PPI

> 'The scope and scale of patient and public involvement in research is expanding but we lack a shared understanding of what works, when, how and why' (9)

Many of the deeply rooted challenges outlined above call for change in research cultures primarily in the knowledge, attitudes and expectations of researchers and patients/ members of the public (13). Within the health research community, opinion about the value of PPI appears divided, with some researchers proactively embracing and implementing PPI and others arguing that it represents a threat to the quality or robustness of research design and data collection (94). Those that are currently proactively implementing PPI mainly do so based on the moral and ethical argument. They inherently believe that PPI is of intrinsic value, and, as such needs no further justification (13). As Arnstein noted almost 50 years ago- 'The idea of citizen participation is a little like eating spinach; no one is against it in principle because it is good for you' (33). However, current increasing demands for PPI from research funders, journals and ethics committees require PPI to be universally adopted within the health research community, not just by those that believe it 'is a good thing to do'. Funding applications that are contingent on having 'strong' PPI without clarity on how to conduct 'strong' PPI or on why PPI should be used can tempt researchers to exaggerate the scope of PPI in grant applications in order to obtain funding (55). This often results in superficial engagement and inefficient use of resources, also known as tokenistic involvement (13, 55, 95-98). It is thought that this type of 'tick-box' involvement is neither meaningful nor effective and does not allow PPI to reach its full potential (11, 75, 94, 96, 97). Lack of

public awareness and understanding of research and PPI has also been identified as a significant barrier to meaningful involvement which can result in researchers finding it difficult to recruit PPI contributors and ensure diversity amongst PPI contributors (13, 99).

2.3.1 Why focus on PPI in trials?

Randomised controlled trials (RCTs) are widely accepted as the 'gold standard' for measuring the effectiveness of interventions (100). In an RCT, trial participants are randomly assigned to one of two groups; one (the experimental group) receiving the intervention that is being tested, and the other (the comparison or control group) receiving an alternative (conventional) treatment. The two groups are then followed up to see if there are any differences between them in outcome. The results and subsequent analysis of the trial are used to assess the effectiveness of the intervention, which is the extent to which the treatment, procedure, or service does patients more good than harm (101). RCTs can test the effectiveness of clinical or behavioural interventions. Clinical interventions include new medicines, therapies, devices, diagnostic techniques and surgical procedures, as well as optimising existing products and procedures to promote better health and welfare (102, 103). Behavioural interventions are studies in which the primary purpose is to evaluate attempts to influence behaviour or the consequences of any resultant behaviour change. Behavioural interventions are becoming increasingly important to public health as lifestyle behavioural risk factors contribute strongly to a wide range of health problems (104). For the past half century, RCTs have reshaped medical knowledge and practice as they are viewed as the most stringent way of

determining whether a cause-effect relation exists between an intervention and an outcome (100, 105).

There are two main reasons why this research focuses specifically on PPI in trials rather than in health research more broadly. The first of these reasons is practical. Given the increasing international focus on trial methodology research as a way to address trial methodological issues and inefficiencies, over the last number of years the area has seen increasing investment from research funders (106). In Ireland, the Health Research Board Trial Methodology Research Network (HRB TMRN) was established to strengthen trial methodology and reporting on the island of Ireland so that they become 'more relevant, accessible and influential for patients and other service users, practitioners, policy makers and the public' (107). Since 2016, the HRB TMRN have run an annual Study Within A Trial (SWAT) funding call which funds researchers to conduct self-contained research studies that are embedded within a host trial to evaluate or explore alternative ways of delivering or organising a specific aspect of the trial process (108). Over the past three years of this doctoral research, I have learnt that most researchers have to 'bend' or 'adapt' their research to fit with funding calls that are available to them. Laudel has named this process 'the art of getting funded' (109). And, so, although my primary research interest is PPI, I adapted this to fit with the SWAT funding calls which provided me with the much-needed resources to conduct and evaluate PPI. Three chapters in this PhD thesis are based on SWATs which were funded by the HRB TMRN SWAT programme.

Secondly, trials have a number of features that lend themselves particularly well to the evaluation of PPI. The enduring history of public activism in trials existed long before the

phrase or concept of 'Patient and Public Involvement' was coined. Examples include HIV/AIDS activism and Breast Cancer Activism from the 1970s, both of which led to a multitude of changes in how trials are designed and conducted (110, 111). Furthermore, given the patient-focused and patient-facing nature of trials, they are regarded as particularly likely to benefit from PPI (44, 67). Although PPI is increasingly being required in all types of research and not just patient-facing research, as we are in the early stages of evaluating the methods and impact of PPI, it makes sense to do this in the context of research that is accessible and visible to patients and the public. This has been particularly evident during the COVID 19 pandemic in recent months where trial protocols and ethical dilemmas have been part of public consciousness and everyday conversations.

2.3.2 PPI methods- moving beyond the 'it depends' argument

> 'We must move forward pragmatically, to ensure that evaluation efforts are not paralysed by the misguided perception that PPI is too controversial or complex to be studied.'
>
> Dr Antoine Boivin, *British Medical Journal,* 2018 (14)

In the PPI literature, PPI approaches and methods are often referred to as the 'context and process' of involvement (12, 77). Although it is acknowledged that the context and process of involvement have an important role in shaping the impact of PPI (12), some researchers have argued that these features are too complex to be evaluated as 'it depends' on too many different factors (13-15) .

It is now time to move beyond the 'it depends' argument. In 2017 a priority setting exercise, the METHODICAL study, identified sixteen critically important research priorities for PPI in trials. The number one priority identified was 'Developing strong and productive relationships between researchers and PPI contributors' (112). The top five research priorities are presented in **Table 2.3** below. The methods we use to involve PPI contributors may play an important role in developing these strong and productive relationships. For example, whether we involve groups of PPI contributors or mixed groups of PPI contributors with other stakeholders, may lead to different experiences and productivity. Generating evidence on suitable PPI methodologies and how different methodologies can shape the impact of PPI is essential if we are to develop 'a shared understanding of what works, when, how and why' (9).

Table 2.3: *Top 10 Methodological priorities for PPI in clinical trials defined by Kearney et al. (112)*

Ranking	**Topic Title**
1.	Developing strong and productive working relationships between researchers and PPI contributors
1.	PPI practices in selecting trial outcomes of importance to patients
1.	A systematic review of PPI activity in improving the accessibility and usefulness of trial leaflets and information sheets for clinical trial participants
4.	Adapting PPI to the particular needs of individual clinical trials
4.	The resources needed for PPI activity including time and money.
4.	PPI practices to address the challenges of recruiting and retaining participants (e.g. patients) in clinical trials

2.3.3 PPI impact- who will the evidence benefit?

Current evidence on the impact of PPI in trials mirrors the limited evidence on PPI in health research more broadly (75). Current reports are mostly based on perceived impact rather than on any evidence of impact (38, 44, 113). PPI costs time and money and therefore pragmatic claims need scrutiny and evaluation (114, 115). In 2018, Boivin writes 'a vast amount of public money and human capital is invested in health research. Since PPI is increasingly seen as pivotal to improving the value and relevance of research, we need to get serious about how it is done and equally serious about how it is evaluated' (14).

Evidence on the impact of PPI is needed so that those critical of PPI can understand the benefits, costs, and risks before they undertake anything more than a tokenistic approach to obtaining grants (13, 14, 42, 116). For researchers already engaging with patients and the public, this evidence is necessary to understand how best to do PPI and fully reap the benefits of working together and avoid any harmful consequences (14, 94, 116). This evidence will benefit research funders and grant reviewers as they would be better equipped to judge the appropriateness as well as the quality of researchers' plans for PPI in grant proposals (16). And finally, this evidence will benefit members of the public as they can learn if, and how, their contributions can make a difference (12).

2.4 Chapter summary

The scope and scale of PPI is expanding but we lack a shared understanding of what works, when, how and why (9). The overarching aim of this thesis is to contribute to the

evidence on the methods and impact of PPI by exploring PPI contributors' experiences and contributions at the design, conduct, and dissemination stages of trials.

Two chapters of this thesis are based on SWATs conducted within the Improving Diabetes Eye-screening Attendance pilot trial (IDEAs) (117). Chapter 3 compares people with diabetes and healthcare professionals' experiences of taking part in three different types of consensus meetings to inform intervention development and assesses whether their experiences differ according to group composition. Chapter 4 compares the contributions of people with diabetes and healthcare professionals during the three meetings and assesses whether their contributions differ according to group composition.

'PPI practices to address the challenges of recruiting and retaining trial participants' has been identified as one of the top five priority research topics for PPI in trials (118). Chapter 5 presents a systematic review and narrative synthesis on researchers' perceived impact of PPI on participant retention in RCTs.

The results of clinical trials have not traditionally been shared with clinical trial participants and uncertainty persists around what information should be shared, how results should be shared, and who should be responsible for sharing the results (119, 120). Chapter 6 presents a SWAT that was conducted within the Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism Trial (TRUST), a trial of thyroxine versus placebo in people aged 65 years and older (121). The SWAT uses a mixed methods

approach, including an embedded randomised controlled trial, to explore the impact of PPI on participants' understanding of clinical trial results.

Chapter 7 provides an overall discussion of the main findings, the strengths and limitations of the thesis, the implications and suggestions for future research. This chapter also includes a reflection on PPI during the COVID-19 outbreak.

3. ‘It just wasn’t going to be heard’: A mixed methods study to compare different ways of involving people with diabetes and healthcare professionals in health intervention research.

Emmy Racine

Fiona Riordan

Eunice Phillip

Grainne Flynn

Sheena McHugh

Patricia M. Kearney

This paper was published in *Health Expectations* in 2019 (122) (Appendix 10).

3.1 Abstract

Background

Guidelines recommend involving intervention users in the intervention development process. However, there is limited guidance on how to involve users in a meaningful and effective way.

Objective

The aim of this Study Within A Trial was to compare participants' experiences of taking part in one of three types of consensus meetings – a people with diabetes only, combined people with diabetes and healthcare professionals (HCPs) or HCP only meeting.

Design

The study used a mixed methods convergent design. Quantitative (questionnaire) and qualitative (observation notes and semi-structured telephone interviews) data were collected to explore participants' experiences. A triangulation protocol was used to compare quantitative and qualitative findings.

Participants

People with diabetes (recruited via multiple strategies) were randomly assigned to attend the people with diabetes or combined meeting. HCPs (recruited through

professional networks) attended the HCP or combined meeting based on their availability.

Results

16 people with diabetes and 15 HCPs attended meetings, of whom 18 participated in a telephone interview. Participants' questionnaire responses suggested similar positive experiences across the three meetings. Observation and semi-structured interviews highlighted differences experienced by participants in the combined meeting relating to: perceived lack of common ground; feeling empowered versus undervalued; needing to feel safe; and going off task to fill the void.

Conclusions

The qualitative theme 'needing to feel safe' may explain the dissonance (disagreement) between quantitative and qualitative data. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately.

3.2 Introduction

For interventions to be successfully implemented in practice, they need to be acceptable, engaging and feasible to implement (123). Intervention development guidelines recommend involving all appropriate intervention users to maximise the chances of successful implementation (124). User involvement is a broad term that includes (but is not limited to) those receiving e.g. patients and members of the public and delivering the intervention e.g. healthcare professionals (HCPs).

Consensus methods are a way of involving multiple users simultaneously in the intervention development process (125-127). Different users may have different priorities and preferences when making decisions about the content and delivery of an intervention (128, 129). For example, patients and members of the public may be concerned about how an intervention will be received by the target population, whereas HCPs may be more concerned about the cost involved (both time and money) (129). Group dynamics are complex, and some user groups may find it more difficult to voice their priorities and perspectives compared to others (130). Despite increasing emphasis on user involvement, limited guidance exists on how to involve users in a meaningful and effective way. To our knowledge, no research has been conducted on patients and HCPs experiences of being involved in consensus methods and whether their experiences differ according to group composition.

The aim of this Study Within A Trial was to compare participants' experiences of taking part in one of three types of consensus meetings – a people with diabetes only, combined people with diabetes and HCPs or HCP only meeting.

3.3 Methods

This Study Within A Trial (SWAT) was conducted within the ongoing Improving Diabetes Eye-screening Attendance (IDEAs) study. IDEAs is a feasibility study of a multifaceted intervention in general practice targeting HCPs and people with diabetes to improve the uptake of retinopathy screening. As part of the development phase of IDEAs, three separate consensus meetings were held to discuss the acceptability and feasibility of the proposed intervention content and suitable modes of delivery. Recommendations from each meeting were used to refine intervention components that could be delivered in general practice. The first consensus meeting consisted of people with diabetes only; the second meeting consisted of a combination of people with diabetes and HCPs; the third meeting consisted of HCPs only.

Study design

The SWAT used a mixed methods convergent design to understand and compare participants' experiences of taking part in the consensus meetings (Figure 3.1). A one-phase design was used, where quantitative (experience survey) and qualitative (observation notes and semi-structured interviews) methods were used during the same timeframe and were given equal weight in the analysis (131).

Quantitative and qualitative data were collected and analysed separately. Results were merged during interpretation (mixed methods phase). A triangulation protocol was used in this phase to compare key concepts identified in each dataset that related to participants' experiences of taking part in the meetings (131, 132). The Good Reporting of A Mixed Methods Study (GRAMMS) framework and the Consolidated Criteria for Reporting Qualitative Studies (COREQ) were used to guide reporting of the findings (133, 134).

Recruitment of participants

People with diabetes were recruited using an information flyer developed by the research team and a graphic designer (Appendix 1.1). The flyer was distributed using a range of recruitment strategies previously identified by Vat et al. (99) (Appendix 1.2).

All individuals who contacted the study team about involvement were sent a 26-item demographic survey (Appendix 1.3 for survey questions and results). The individuals who returned a demographic survey were randomly assigned (using an online random number generator) to the meeting for people with diabetes only or the combined meeting.

HCPs were recruited through professional networks known to the SWAT and IDEAs study teams. HCPs were initially sent an email or letter inviting them to take part in the consensus meeting. This was followed by a phone call to confirm their attendance. HCPs were either allocated to the HCP only or combined meeting based on their availability to attend.

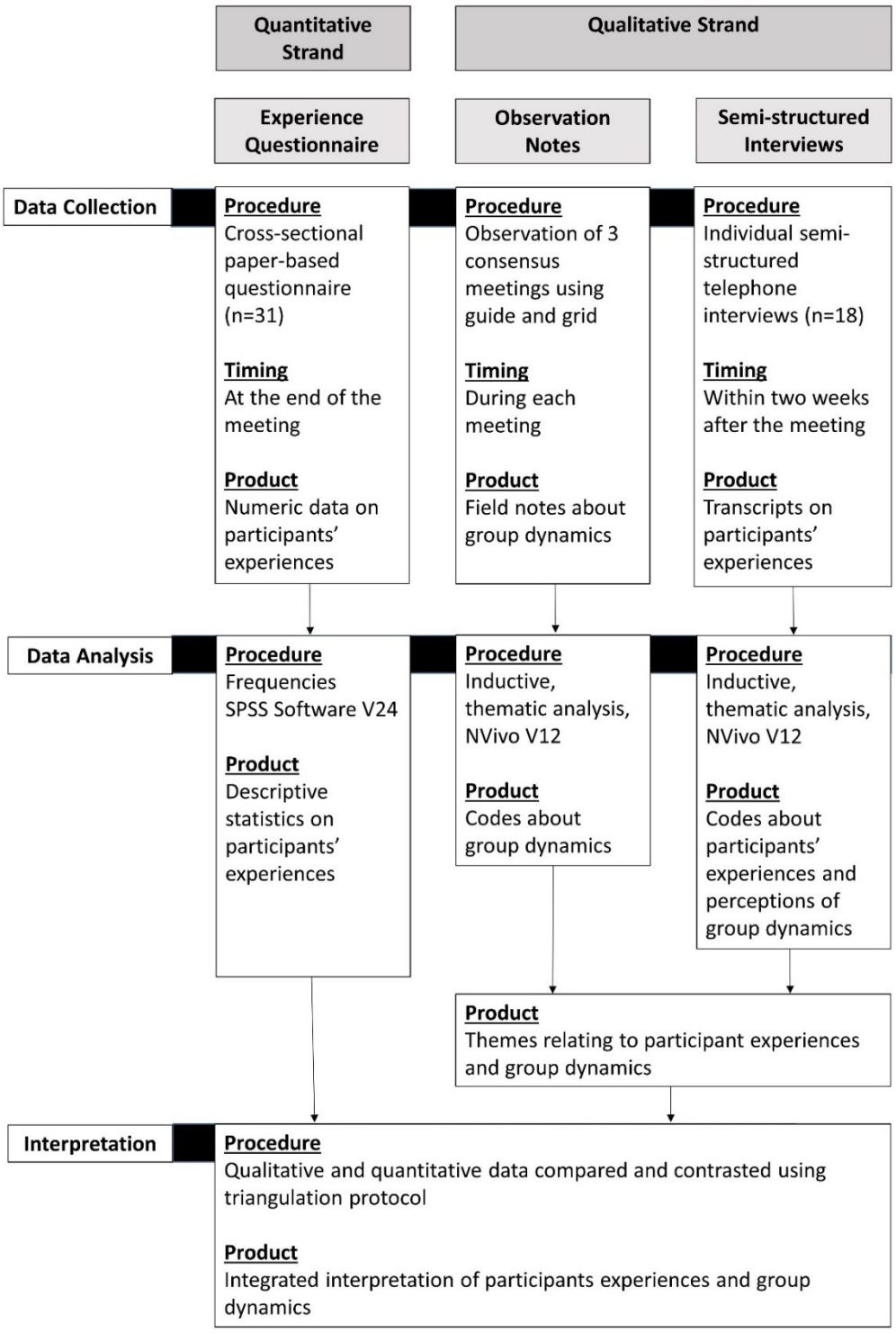

Figure 3.1: Procedural diagram of the convergent study design

Semi-structured consensus meetings

Before the meetings, the IDEAs study team (FR, SMH) developed 1) a short summary of existing evidence on barriers to and enablers of attendance at diabetic retinopathy screening, and interventions to address non-attendance and 2) a survey asking participants to rate intervention components according to acceptability (like it, think it makes sense) and feasibility (think it can be done). The survey was based on measures developed by Weiner et al (135). Materials were reviewed by adult literacy experts (Irish National Adult Literacy Agency) and a Patient and Public Involvement (PPI) group from another research project and revised based on their feedback. Before the meeting, the evidence summary and survey were sent to all meeting participants in electronic or paper format depending on participants' preferences. Survey responses were collated and analysed descriptively by a member of the IDEAs study team (FR) and a summary of the results was prepared to be presented at each meeting.

Each consensus meeting was held from 6.30-8.30pm in University College Cork. Before each meeting (at 6pm), the lead SWAT researcher (ER) held an informal briefing for people with diabetes on key medical and research terms, the aim of the meeting and their role as patient contributors. Each meeting was facilitated by an experienced facilitator (male). During the meetings, a summary of the survey results was presented to participants, followed by a series of small group discussions facilitated by FR, SMH, and EP. Participants were asked how each intervention component would work in practice and which mode of delivery would work best. Each small group was asked to

nominate a lead to feed back their discussion to the larger group. Each group discussion was audio recorded.

Quantitative strand

Experience questionnaire

At the end of each meeting, all participants were asked to complete a questionnaire about their experience of the meeting. The objective of the questionnaire was to understand individual experiences of taking part in the meeting, asking them to rate how they felt about their participation and the participation of other group members; how decisions were made by the group; and the potential impact of the decisions that were made. We were unable to find a suitable validated instrument that was appropriate for our questionnaire objective and context (one-off participatory research process). Therefore, we developed our own questionnaire based on sample items from a non-validated survey instrument published by Schulz et al. (136). For additional information on the questionnaire development, please see Appendix 1.4. The original phrasing of the sample items was maintained, with the exception of some questions that were changed to statements to fit with a Likert Scale format. Agreement with each statement was measured on a five-point Likert scale ranging from 'strongly disagree' to 'strongly agree'. The questionnaire also contained an open-ended comment box for any other comments or suggestions. At the bottom of the questionnaire, participants were invited to 'opt in' if they were interested in participating in a follow-up interview on their experiences of taking part in the meeting.

Quantitative data analysis

Questionnaire responses were entered into SPSS software (version 24) and analysed using descriptive statistics. The five response categories were collapsed into three categories – 'Agree', 'Neither agree nor disagree' and 'Disagree'.

Qualitative strand

Observation notes

The SWAT lead researcher (ER) observed each consensus meeting and took comprehensive field notes. The objective of the observation was to understand how members participated and interacted with other meeting members and how they made decisions for the development of the intervention (group dynamics and decision-making processes). An observation guide and grid were used to guide note-taking and as a reminder of the events and issues of most importance (Appendix 1.5) (137). The observation guide contained two overarching questions: 'How is the group working overall?' and 'How is the group making decisions?'. The observation grid contained six constructs informed by group dynamics and decision-making processes literature (138-141). These constructs were: participation/non-participation, dominance/submissiveness, in-groups/out-groups[1], body language and facial

[1] An in-group is a social group to which a person psychologically identifies as being a member. An out-group is a social group with which a person does not identify.

expressions, gaze, and effect of expert/lay knowledge. After each meeting, the researcher met with the group facilitators to discuss and document their experiences and perspectives as supplementary information.

Semi-structured interviews

Within two weeks of the consensus meetings, semi-structured telephone interviews were conducted with the consensus meeting participants who agreed to take part in an interview in the experience questionnaire. The objective of the interviews was to gain insights into individual experiences of taking part in the meeting in terms of: how comfortable they felt in the meeting; how they felt members of the group interacted with each other; and how they felt they worked together to make decisions (i.e. whether there was agreement, conflict, synergy). Interviews were audio-recorded (see Appendix 1.6 for Interview Topic Guide). Interviews were conducted by ER, a young female PhD candidate. All participants were familiar with ER as she facilitated the briefing session prior to the consensus meetings. At the beginning of each interview, the SWAT lead researcher (ER) stressed to participants that she was independent to the trial study team that were running the consensus meetings and therefore would not be offended if they described negative experiences.

Qualitative data analysis

Field notes were collated, and audio recordings were transcribed verbatim. All qualitative data were managed using NVivo software (version 12). Thematic analysis was carried out following Braun and Clarke guidelines (142). Firstly, an extensive familiarisation process

was conducted by two researchers (ER, EP), where notes and transcripts were read and re-read multiple times. ER open coded all the observation notes and transcripts (using semantic and latent codes) and developed three separate sets of codes- one set for each meeting. The pattern and meanings of codes were then examined across the three meetings to identify one set of candidate or potential themes relating to participants' experiences and group dynamics. Themes were developed using a conventional or 'bottom-up' approach, whereby themes were developed directly from the data (142). ER discussed each theme with EP to revise, refine and define themes.

Mixed methods phase

After separate analysis of quantitative and qualitative data (as described above), the data were compared using a triangulation protocol. Triangulation provides a visual and tabular representation of the findings from qualitative and quantitative data, allowing for a clearer comparison and broader interpretation (143). The steps taken to create the triangulation protocol are outlined in **Table 3.1** below.

Table 3.1: Steps taken to create triangulation protocol

	Step	**Activity**
1.	Collate key findings from each dataset	This was done by examining the original data, interpretation and reports of analysis. For quantitative data, each questionnaire item was deemed as a separate key finding. For qualitative data, multiple key findings were identified within each theme, as themes were too broad in their descriptions to compare directly to quantitative findings.

2.	Group key findings into concepts	Key quantitative and qualitative findings were grouped together into concepts according to how they related to participants' experiences and group dynamics (e.g. freedom of expression, balance of participation).
3.	Create table for triangulation protocol	A table was created with each column representing the data source (questionnaire, observation and interview) and each row representing a key concept.
4.	Map key findings to table	Key findings were then mapped to the table to examine where findings from each method agreed (convergence), offered complementary information on the same issue (complementarity), appeared to contradict each other (dissonance) or appeared in one method and not the other (silence) (144).
5.	Explore inter-method discrepancies	This was done by examining the methodological rigour of each method and re-examining the data in light of the discrepancy (145).

Patient and Public Involvement (PPI) component

A PPI partner (GF) was involved in the SWAT from the outset. The PPI partner is a person with diabetes, previously known to the lead author (ER). She contributed to the initial discussions about the study which ultimately informed the SWAT grant application, reviewed the application and made changes to its content. GF was also involved in the development of materials used to recruit PPI contributors and assisted the research team with recruitment by posting recruitment flyers online via social media networks. In

addition, she contributed to and reviewed each draft of this manuscript and is a co-author on this publication.

Ethics

The study received ethical approval from the Social Research Ethics Committee (SREC) at University College Cork. Written informed consent was obtained from all participants prior to taking part in the consensus meetings and completing the questionnaire. Telephone consent was obtained from participants prior to taking part in the interviews.

3.4 Results

Participants

A total of 36 people contacted the research team expressing an interest in the SWAT. Of these, twenty completed the recruitment survey (see Appendix 1.3 for recruitment survey results). These twenty people were randomly assigned to either the people with diabetes only meeting (4 with type 1 diabetes and 6 with type 2 diabetes) or the combined meeting (6 with type 1 diabetes, 3 with type 2 diabetes and 1 carer). All 10 people attended the people with diabetes only meeting (attendance rate 100%) and 6 people with diabetes attended the combined meeting (attendance rate 60%). An invitation to attend was sent out to 50 HCPs (practice nurses, diabetes nurse specialists, general practitioners and specialist physicians), of which 8 attended the combined

meeting and 7 attended the HCP only meeting (attendance rate 30%). Further details on the recruitment and response rates for each stage of the data collection are shown in Figure 3.2 below.

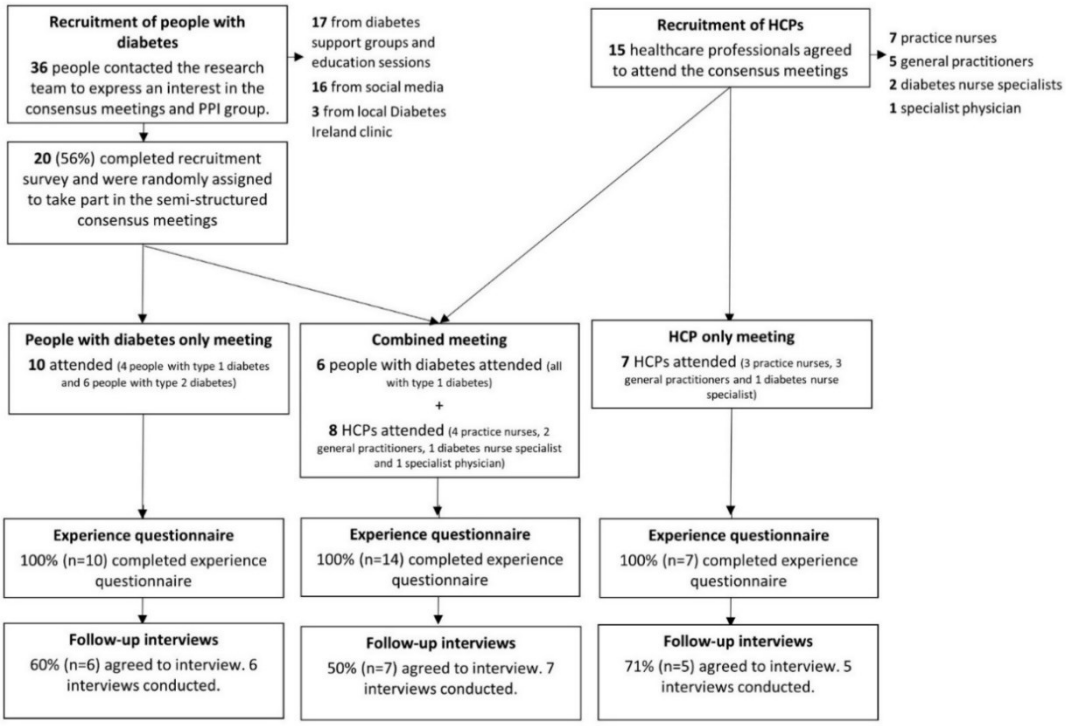

Figure 3.2: Flow diagram of recruitment and response rates

Quantitative results

All consensus meeting participants (n=31) completed the experience questionnaire (response rate 100%). **Table 3.2** shows the results of the questionnaire stratified by meeting type (people with diabetes only, combined and HCP only). The descriptive statistics presented in **Table 3.2** demonstrate that there were no differences in participants' self-reported experiences of the three meetings. All participants across the

3 groups agreed with the statements '*I felt comfortable expressing my opinion in the group'*, *'I felt my opinions were listened to and considered by other group members'* and ' *I did not feel pressured to go along with the decisions of the group even though they did not agree*'. Some participants agreed with the statements that '*I thought that certain individuals spoke more than others in the group'* and '*I felt that certain individuals had more influence over the decision-making process than others'*. A number of participants expressed doubt that they could influence the decisions made during the meeting.

Table 3.2: Results of the participant experience questionnaires stratified by meeting type

Item	Meeting	Agree N (%)	Disagree N (%)	Neither agree nor disagree N (%)
I felt comfortable expressing my opinion in the group	**People with diabetes**	10 (100)	-	-
	Combined	14 (100)	-	-
	HCP	7 (100)	-	-
I felt my opinions were listened to and considered by other group members	**People with diabetes**	10 (100)	-	-
	Combined	14 (100)		-
	HCP	7 (100)	-	-
I felt part of the group (like I belonged to the group)	**People with diabetes**	10 (100)	-	-
	Combined*	12 (92.3)	-	1 (7.7)
	HCP	7 (100)	-	-
I felt pressured to go along with the decisions of the group even though I did not agree	**People with diabetes**	-	10 (100)	-
	Combined	-	14 (100)	-
	HCP	-	7 (100)	-
I felt a sense of trust and openness between group members	**People with diabetes**	10 (100)	-	-
	Combined	13 (92.9)	-	1 (7.1)
	HCP	7 (100)	-	-
	People with diabetes only	3 (30)	6 (60)	1 (10)

I thought that certain individuals spoke more than others in the group	**Combined**	4 (28.6)	6 (42.8)	4 (28.6)
	HCP	3 (42.9)	3 (42.9)	1 (14.2)
I felt that I could influence the decisions made by the group	**People with diabetes**	7 ((70)	-	3 (30)
	Combined	8 (57.1)	1 (7.1)	5 (35.7)
	HCP*	4 (66.7)	-	2 (33.3)
I felt that certain individuals had more influence over the decision-making process than others	**People with diabetes**	3 (30)	6 (60)	1 (10)
	Combined	2 (14.3)	9 (64.3)	3 (21.4)
	HCP	1 (14.3)	3 (42.9)	3 (42.9)
I have increased my knowledge about important topics since participating in this group	**People with diabetes***	8 (88.9)	-	1 (11.1)
	Combined	10 (71.4)	1 (7.1)	3 (21.4)
	HCP	6 (85.7)	-	1 (14.3)
By working together, we can influence decisions that affect the research process	**People with diabetes only**	10 (100)	-	-
	Combined	13 (92.9)	-	1 (7.1)
	HCP	7 (100)	-	-
By working together, we can influence decisions that affect people with diabetes	**People with diabetes**	10 (100)	-	-
	Combined	14 (100)	-	-
	HCP	7 (100)	-	-

*missing data

Qualitative results

In total, 18 questionnaire respondents agreed to be contacted for a follow-up interview. Interviews were conducted with participants from the people with diabetes only (n=6), combined (n=7) and HCP only (n=5) meetings. Interviews were, on average, 34 min in duration (range 18–56 min).

Four themes were developed from the qualitative data relating to participants' experiences and group dynamics: perceived lack of common ground; feeling empowered versus undervalued; needing to feel safe; and going off task to fill the void.

Perceived lack of common ground

In the people with diabetes only meeting, there were differences between participants in terms of diabetes type, length of diagnosis and education level. In the HCP only meeting, differences included profession (e.g. medical doctor, practice nurse, diabetes nurse specialist), experience of working with people with diabetes, and size, location and nature of their practices. During the interviews, participants from these two meetings described these demographic, geographical and clinical differences as '*small*' differences, which they welcomed as they felt it allowed them to bring different perspectives to the topics they were discussing. They focused on the common ground they shared with other meeting participants and identified with one another based on the shared experience of living with diabetes or caring for people with diabetes. They felt that they were all '*singing from the same hymn sheet*' (P3, person with diabetes, people with diabetes only meeting) and described being able to come together to make decisions that incorporated different perspectives:

> "It was interesting to hear their views. We were all on the same page, but we were coming from different angles and we used that then; we came together and made the decisions together." (P2, person with diabetes, person with diabetes only meeting)

In contrast, a lack of common ground was reported by participants in the combined meeting. This created a division in the group, a 'them' and 'us' attitude, which was evident in the interview and observation data. In the interview data, people with diabetes stated that there was *a 'complete clash of perspectives'* (P9, person with diabetes, combined meeting) between people who lived with the condition and HCPs who cared for people with diabetes. HCPs reported that people with diabetes and HCPs were '*two different sides of the divide'* (P11, HCP, combined meeting). The observation data also suggested a division between people with diabetes and HCPs in the combined meeting. At the beginning of the meeting, people with diabetes and HCPs sat on opposite sides of each small table. During the small group discussions, participants expressed their opinions as collective opinions of their stakeholder group. Rather than expressing individual opinions (e.g. 'I think that…'or 'My experience is…'), people with diabetes spoke on behalf of all the people with diabetes in the group, and HCPs spoke on behalf of all HCPs in the group (e.g. 'We feel that… don't we?' and 'As people with diabetes, we think that…'). Moreover, during the small group discussions, each stakeholder group focused their gaze on the other stakeholder group, resulting in people with diabetes and HCPs talking at each other, at opposite sides of each table. This was in contrast to the people with diabetes only and HCP only meeting, where members focused their gaze on all members around the table.

Participants' lacking a sense of shared experience was accompanied by differences in perceptions around the balance of participation. During all three meetings, it was observed that some participants spoke more frequently and for longer than others. In the interviews, participants from the people with diabetes only and HCP only meetings

perceived this unbalanced participation as a natural consequence of any group dynamic. They mainly attributed it to different personalities. In contrast, HCPs from the combined meeting attributed the unbalanced participation to people putting too much emphasis on their own personal experiences:

> "It was very much centred around them [people with diabetes] and a lot of the offerings that I had in terms of experience were nothing in comparison to what they felt as people that have the problem. Which is fine. But that wasn't really the point. The point is that I don't have diabetes, that is not my personal experience. But I am still the one left in the room everyday trying to deal with patients… But I just couldn't come out with it on the night. I just didn't. It wasn't going to be heard." (P12, HCP, combined meeting)

Feeling empowered versus undervalued

In the interviews, participants from the people with diabetes and HCP only meetings reported learning from other meeting members and feeling empowered by the event. In the people with diabetes only meeting, participants stated that they learned from one another about how they can better manage their condition and about the difference between type 1 and type 2 diabetes. Those who had been diagnosed with diabetes for a long time described gaining a renewed compassion for those who were newly diagnosed. Participants from the HCP only meeting reported learning about the importance of encouraging their patients to attend screening, about the roles of different HCPs, and

about the cultural difficulties and language barriers that some practices face due to a high number of non-English speaking patients.

There were also some reports of learning in the combined meeting. People with diabetes said they gained a new insight into the work practices of HCPs – in particular, the increased workload experienced by HCPs. The HCPs reported gaining an insight into the struggles of having to live with a medical condition:

> "I put in a couple of thousand eye drops a year, it doesn't mean anything to me like. But it obviously means something for patients who are having to go through this – and you know the awkwardness of getting appointments and driving to and from appointments and getting a lift and all that side of things." (P14, HCP, combined meeting)

However, participants from the combined meeting reported feeling undervalued by the other stakeholder group. People with diabetes felt that HCPs did not understand how it feels to live with a chronic illness, describing '*a complete clash of the reality of living with diabetes versus a medical professional's perspective'* (P7, person with diabetes, combined meeting). Among some of the HCPs, there was a sense that any contributions they made during the meeting were not valued by people with diabetes because the experience of living with diabetes was deemed more important than the experience of caring for people with diabetes:

> "I've worked in four different GP practices at this stage and all very different. And yet I felt like as if any value that I had to add to the conversation was kind of

almost either misheard or not really heard, or almost felt not quite as relevant because of their personal experiences. Which is fair enough. But that was not what the meeting was about." (P13, HCP, combined meeting)

Needing to feel safe to express honest opinions

In the interviews, participants from the people with diabetes only and HCP only meetings reported an open, honest, relaxed and non-judgemental environment, where everyone had a voice and was heard. This environment made participants feel safe and comfortable to express their opinions. They also indicated that the small group discussions added to their feelings of safety as people who do not like speaking in public felt less intimidated about expressing their opinions:

"I'm not one really for expressing my opinions. I am kind of … I wouldn't put my hand up the first time, let's say. But I did feel very comfortable expressing my opinion in the small group." (P15, HCP, HCP only meeting)

Conversely, participants from the combined meeting reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room. Both people with diabetes and HCPs said they felt they had to '*hold back'* their opinions. People with diabetes reported feeling that they could not be honest about the '*non-compliant*' (P9, person with diabetes, combined meeting) aspects of managing their diabetes as the HCPs may judge them for it:

"I don't think when you are sitting at a table with HCPs that you're going to be

> discussing the non-compliant things you do… It's probably not the best environment, let's say, to get out some of the smaller things that people do that may not be approved by the other group in the room." (P8, person with diabetes, combined meeting)

On the other hand, HCPs were conscious of confidentiality issues: they were concerned that if they mentioned a particular case, people with diabetes could potentially identify who that patient was, since *'[this location] is a very small place'* (P11, HCP, combined meeting):

> "I felt a bit kind of reticent about how free [I could talk about my experiences as a healthcare professional] … It's different when you are divulging, you know, work practices and difficulties and challenges and personal experiences at work, when it is other medical professionals. But when you have effectively patients there, it is like a big difference." (P13, HCP, combined meeting)

In addition, the HCPs indicated that they did not feel comfortable talking about the service that they worked in as they felt anxious that people with diabetes would confront them on the long waiting times or other issues they had with that particular service.

<u>Going off task to fill the void</u>

Analysis of interview data indicated that participants across all three meetings felt they were able to work together. They reported that the content for discussion was relevant to them as users and providers of health services.

However, the observation data show that although members of the combined meeting appeared to work together, both stakeholder groups were defensive about what intervention components would not work and at times in the meeting nothing seemed feasible. This resulted in each stakeholder group feeling uncomfortable in asserting what they felt the other group should or should not do. To fill this void, participants began to go off task as they focused their discussions on the 'other'. The 'other' took different forms throughout the meeting: the screening service, people with diabetes who were not in the room (e.g. those with type 2 diabetes), and funding and resource limitations in general practice. Even though they were being asked to discuss and make recommendations on how the intervention would work in primary care, the combined meeting participants resorted to making recommendations about how screening uptake could be increased on a national basis through nationwide TV and radio campaigns.

Mixed methods results

The results of the mixed methods analysis are presented in **Table 3.3.** Six key concepts relating to participants' experiences and group dynamics were identified from the datasets: freedom of expression; understanding and respect; balance of participation; learning; productive collaboration; and group cohesion. When key findings were mapped to the overarching concepts, there were four instances of dissonance (where data appeared to contradict each other), two instances of convergence (where data agreed) and two instances of complementarity (where data offered complementary information on the same issue). There were no instances of silence (where data appeared in one method and not in the other).

The four instances of dissonance between quantitative and qualitative data were wholly due to the fact that in the questionnaire participants reported positive experiences of taking part in the meetings, whereas the observation and interview data highlighted some negative experiences and divergent opinions. For example, in relation to *freedom of expression*, the questionnaire data showed that in all three meetings, participants reported feeling comfortable expressing their opinions and reported a sense of trust and openness between group members. In the observation data, participants in the combined meeting did not appear to be comfortable asserting what the other stakeholder group should/should not do as part of the intervention. Furthermore, in the interview's participants reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room.

The instances of complementarity were largely due to the design of the data collection tools. The questionnaire items were designed to be concise and did not require the participants to give any additional details. Whereas in the interviews, participants had the opportunity to expand and give more detail. For example, in the key concept *learning*, the questionnaire item asked participants to indicate how much they agreed with the statement 'I have increased my knowledge about important topics since participating in this group', whereas in the interviews participants had the opportunity to expand and give specific examples of what they had learned (e.g. people with diabetes learned how they can better manage their condition, HCPs leaned about the importance of encouraging their patients to attend screening, etc.).

Table 3.3: Results of mixed methods analysis (triangulation protocol)

Key concept	Quantitative strand	Qualitative strand		
	Questionnaire	**Observation notes**	**Interviews**	
Freedom of expression	In all three meetings, participants were comfortable expressing their opinions and felt a sense of trust and openness between group members	In the combined meeting, participants did not appear to be comfortable asserting what the other stakeholder group should/should not be doing	In the people with diabetes only and HCP only meetings, participants reported that it was an open, honest and relaxed environment where they felt comfortable expressing their opinions In the combined meeting, participants reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room	Dissonance
Understanding and respect	In all three meetings, participants felt their opinions were listened to and considered by other group members, and that they could influence the decisions being made by the group	-	In the combined meeting, participants reported feeling undervalued by the other stakeholder group	Dissonance
Balance of participation	In all three meetings, some participants felt that certain individuals spoke more than others and had more influence over the decision-making process	In all three meetings, some participants spoke more frequently than others and for longer lengths of time	In the people with diabetes only and HCP only meetings, participants were understanding of the unbalanced participation and saw it as a natural consequence of any group dynamic In the combined meeting, HCPs attributed	Convergence, complementarity

Key concept	Quantitative strand	Qualitative strand		
	Questionnaire	Observation notes	Interviews	
			unbalanced participation to people putting too much emphasis on their own personal experiences	
Learning	In all three meetings, most participants felt they increased their knowledge as a result of attending	In all three meetings, participants appeared keen to learn from one another as they asked each other about their experiences	In all three meetings, participants reported learning from one another and provided specific examples of this learning	Convergence, complementarity
Productive collaboration	In all three meetings, participants reported that they were able to work together to influence decisions that affect the research process and people with diabetes	In the combined meeting, although participants appeared to work together, each stakeholder group did not make any comments on what the other stakeholder group should/should not do. Instead, they made recommendations that were not relevant to the intervention (unproductive collaboration).	In all three meetings, participants reported being able to work together as they felt the content for discussion was relevant to them as users and providers of health services	Dissonance
Group cohesion	In all three meetings, participants reported they were part of the group (like they belonged to the group)	In the combined meeting, it was evident that there was a division between both stakeholder groups (e.g. both groups spoke at each other across each table as opposed to with each other around each table).	In the people with diabetes only and HCP only meetings, participants reported that there were some 'small' differences between meeting members, but added that this was a good thing as it allowed them to bring different perspectives to the topics they were discussing	Dissonance

Key concept	Quantitative strand	Qualitative strand		
	Questionnaire	Observation notes	Interviews	
			In the combined meeting, people with diabetes reported that there was a 'complete clash of perspectives' between people with diabetes and HCPs; HCPs reported that people with diabetes and HCPs were 'two different sides of the divide'	

3.5 Discussion

Summary of key findings

The aim of this study was to compare participants' experiences of taking part in the three consensus meetings. The results of the questionnaire suggested that participants had largely positive experiences of taking part in the consensus meetings and there were no differences in participants' experiences between the three meetings. However, results of the observation and interviews highlighted that participants in the combined meeting had different experiences to those in the other two meetings. The perceived lack of common ground between people with diabetes and HCPs in the combined meeting led participants to feel undervalued by the other stakeholder group as they felt that the other group did not understand their perspective. Participants in the combined meeting were reluctant to express their opinions and were defensive about what would/

wouldn't work in terms of developing the intervention. As a result, participants in the combined meeting went off task and made recommendations which were not entirely relevant for the intervention. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately.

Links to existing literature

In the people with diabetes only and the HCP only meetings, participants welcomed their diversity as it allowed them to hear different perspectives on the topics they were discussing. This finding is consistent with existing literature, with many theorists arguing that knowledge diversity can improve group performance by enhancing a group's ability to be creative and to discover novel solutions (146-148). In these meetings, participants focused on their common ground and described being able to come together to make decisions that incorporated a range of perspectives. Previous research suggests that congruent groups- that is, when group members are socially tied and share the same information – are more likely to be productive and successful (149).

The perceived lack of common ground between people with diabetes and HCPs in the combined meeting created a 'them' and 'us' scenario, with participants reluctant to express their opinions. This raises questions about whether too much difference within groups is counterproductive or divisive. Existing research on the productivity of incongruent groups - that is, when social and knowledge subgroups are present within a

group has found that sub-groups can create a divide between group members, undermining the groups' ability to work together and be productive (149).

Some HCPs in the combined meeting felt their contributions were not valued by people with diabetes because the experience of living with diabetes trumped the experience of caring for people with diabetes. This finding may reflect the changing nature of the patient/HCP relationship over the last 20 years – from a paternalistic model where the patient seeks help and is compliant to the professional who makes the decisions, to a more patient-centred approach (150). This approach expects HCPs to enter the patient's world and to see the illness through the patient's eyes (150). This prioritisation of the patient experience has benefited patient outcomes (151). However, as HCPs are often responsible for delivering interventions, their perspectives in the intervention development process are crucial for maximising intervention feasibility. Involving multiple users in the intervention development process is not about understanding which perspective is more valid or more important, it is about understanding all the different perspectives so that the intervention is more acceptable, engaging and feasible to implement.

Strengths and Limitations

One of the strengths of this study was the use of a mixed methods, convergent design which produced a more complete understanding of participants' experiences and group dynamics. It also allowed for the cross-validation of findings from each method resulting in more substantiated findings than sequential designs or quantitative or qualitative

approaches alone (131). The qualitative theme 'needing to feel safe' may explain the instances of dissonance between quantitative and qualitative data as participants completed the questionnaire at the end of each meeting while they were still sitting close to other participants. Some small groups even filled out the questionnaire together. As a result, participants may not have felt comfortable voicing concerns. In the interviews, on the other hand, participants may have felt safer in a one-to-one environment with a researcher who they were already familiar with. The fact that the researcher stressed that she was independent to the consensus meeting research team and her informal approach may have made them more comfortable to speak openly about their experiences of taking part in the meeting. The timing of the questionnaire may have also played an important role. The questionnaire was handed out at the end of the meeting, late in the evening. Participants may have been eager to get home and they may not have fully thought about the responses they were providing. Whereas, in the interviews, participants had time to reflect on their experiences and a provide a more comprehensive account as a result. This is consistent with Krosnick's theory of survey satisficing which is based on the assumption that optimal survey completion involves doing a great deal of cognitive work, so if the participant is not fully motivated to complete the survey, he or she is likely to offer responses that seem reasonable and easy to defend (152). Although questionnaires are a frequently used tool to evaluate consensus meetings, our findings suggest they may not always provide a comprehensive assessment of participants' experiences. This is consistent with a number of previous studies on evaluating participant experiences (153-155).

This study is not without limitations. First, the questionnaire that was used to understand participants' experiences was based on non-validated questionnaire items. We were unable to conduct exploratory factor analysis to validate our questionnaire as our sample size did not meet the minimum criteria of 10 participants per questionnaire item (156). However, given the increasing importance of evaluating PPI and other participatory research activities (14), the questionnaire could be a useful tool in future studies which aim to understand stakeholders' experiences in similar participatory research contexts. Use of the questionnaire in future studies may allow for reliability testing and validation to be carried out (157, 158).

Second, although the experience questionnaire suggested that there were no differences in participants' experiences between the three meetings, due to the number of participants, there was limited power to detect a difference (n=31). Thus, the comparison of participants' questionnaire responses between the groups is used as only an indicator of participants' experiences. Given the small sample, we cannot rule out the possibility that differences between the groups could be detected had a larger sample size been used.

Despite using a range of strategies to recruit a representative sample of people with diabetes, another potential limitation of this study was the absence of people with type 2 diabetes in the combined meeting. As the attendance rate of people with diabetes at the combined meeting (60%) was much lower than the people with diabetes only meeting (100%), it is plausible that people with type 2 diabetes did not attend because they knew there would be HCPs attending. Existing research has established that people

with type 1 and type 2 diabetes have different experiences when managing their condition and engaging with HCPs (159-161). Therefore, the involvement of people with type 2 diabetes in the combined meeting could have potentially changed the nature of the relationship between patients and HCPs and led to different participant experiences and group dynamics.

Finally, participants were given a choice to participate in an in-person or telephone interview. All participants chose telephone interviews due to time constraints and location convenience. This could be another potential limitation as researchers have previously expressed concerns about whether telephone interviews are appropriate for qualitative research (162, 163). These concerns are largely due to the absence of visual cues which may result in the loss of informal communication and contextual information, the inability to develop rapport or to probe and the misinterpretation of responses (163). In this study, the quality of telephone data cannot be compared to in-person data as no in-person interviews were conducted. However, the researcher had considerable experience conducting phone interviews, maintained a friendly and engaging tone throughout and as mentioned previously, participants were found to be open and frank about their experiences.

Implications

The results of this study provide much-needed evidence on how different ways of involving patients and healthcare professionals can lead to differing participant experiences and group dynamics. Patient and public involvement (PPI) in research is

increasingly becoming a requirement in health research and for many research funders. INVOLVE, a national advisory body funded by the National Institute for Health Research (NIHR) in the UK, defines public involvement as research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them (1). In this study, the lines between research participation and involvement were blurred, as is often the case with PPI (164). People with diabetes were research participants in the consensus meetings, experience questionnaire and semi-structured interviews. However, their role in the consensus meeting was to discuss and make decisions about the intervention content and mode of delivery which could be viewed as PPI (33, 165). This study shows that the context and nature of involvement can have important implications for its impact. These findings are not only relevant to health intervention researchers but to all individuals interested in involving patients and members of the public in health research, policy and in the planning and development of health care more broadly.

3.6 Conclusion

Although the results of the experience questionnaire showed no differences in participants experiences across the three meetings, the results of the observation and interviews highlighted that participants in the combined meeting had different experiences. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately. The study provides much-needed evidence on how different ways of involving patients and healthcare professionals can lead to differing participant experiences and group dynamics.

Racine, E. 2020. 'It's a nice thing to do but…': exploring the methods and impact of patient and public involvement (PPI) in trials. PhD Thesis, University College Cork.

Please note that Chapters 4 & 5 (pp. 75-156) are unavailable due to a restriction requested by the author.

6. Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial

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This paper was published in *HRB Open* in 2018 (258)(Appendix 10).

6.1 Abstract

Background

While there is an increasing consensus that clinical trial results should be shared with trial participants, there is a lack of evidence on the most appropriate methods. The aim of this Study Within A Trial (SWAT) is to use a patient and public involvement (PPI) approach to identify, develop and evaluate a patient-based approach to receiving trial results for participants in the Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism Trial (TRUST), a trial of thyroxine versus placebo in people aged 65 years and older.

Methods

Mixed methods study with three consecutive phases. Phase 1 iteratively developed a patient-based approach using semi-structured focus groups and a consensus-orientated-decision model, a PPI group to refine the method and adult literacy review for plain English assessment. Phase 2 was a single-blind parallel group trial. Irish TRUST participants were randomised to the intervention (patient-based approach) and control group (standard approach developed by lead study site). Phase 3 used a patient understanding questionnaire to compare patient understanding of results between the two groups.

Results

Participants want to receive results of clinical trials, with qualitative findings indicating three key themes including 'acknowledgement of individual contribution', 'contributing for a collective benefit' and 'receiving accessible and easy to understand results'. Building on these findings, the patient-based approach was developed. TRUST participants (n=101) were randomised to the intervention (n=51) or control group (n=50). The questionnaire response rate was 74% for the intervention group and 62% for the control group. There were no differences in patient understanding between the two approaches.

Conclusions

We have demonstrated that it is feasible to involve trial participants in the development of result dissemination materials. Although, in this study PPI did not influence patients' understanding of results, it documents the process of conducting PPI within the clinical trial setting.

6.2 Introduction

Patient and public involvement (PPI) is increasingly recognised as an essential component of clinical research. In the UK, the national advisory group supporting active public involvement in health services, public health and social care research (INVOLVE) defines PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to' 'about' or 'for' them'(1). In clinical trials, PPI has been defined as experimenting with participants instead of experimenting on participants (259). PPI may occur at any stage during the research process from priority setting and drafting study protocols right through to conducting the study, interpreting the end results and communicating and disseminating research findings (52, 53). Research funders increasingly expect that PPI is prioritised and resourced within studies. This increasing expectation has heightened the risk of researchers carrying out 'tick-box' PPI rather than 'meaningful' involvement (96). There are many moral and ethical arguments being made for PPI. Many believe that as citizens and taxpayers, members of the public have a right to influence research that is being funded by public money (3). PPI researchers are also making pragmatic arguments for PPI and providing anecdotal accounts about how PPI can make research more relevant, accessible and acceptable to participants (8).The ethical arguments are often seen as sufficient regardless of any pragmatic impact. However, PPI costs time and money, therefore pragmatic claims need scrutiny (114). More substantive evidence is needed to evaluate the potential impact of PPI on the conduct and outcomes of research (96, 260). In 2001, the need to establish if PPI leads to actual, rather than merely perceived benefits for research processes and output was identified. Over fifteen years later, this need remains.

In clinical research, the results of clinical trials have not traditionally been shared with clinical trial participants. A recent survey carried out on a large registry of health research participants, found that while 95.6% of respondents said researchers should always or sometimes offer the results to participants, only 33% of respondents actually received the results of studies in which they had participated (119). An upcoming European Union Clinical Trial Regulation requires sponsors to provide summary results of clinical trials in a format understandable to laypersons, including participants (261). However, there is a lack of evidence on the most appropriate methods of sharing results with participants. Uncertainty persists around what information should be shared, how results should be shared and who should be responsible for sharing the results. Since the findings of clinical research often exist in a complex context of scientific exchange and debate, it is important that the information shared is accessible and relevant to participants (120). The increasing understanding of the importance of sharing research results with study participants is somewhat linked to a wider movement towards transparency in trials. This movement is largely promoted by initiatives such as SPIRIT, CONSORT and AllTrials. The SPIRIT Statement provides guidance to researchers to improve the completeness and quality of trial protocols (262), the Consolidated Standards of Reporting Trials (CONSORT) Statement is an evidence based, minimum set of recommendations for reporting randomised trials (263), and the AllTrials initiative calls for all past and present trials to be registered and their full methods and summary results reported (264). Some of these initiatives also include recommendations for disseminating results to research participants. For example, the SPIRIT statement states that study results must be released to participating physicians, referring physicians, patients and the general

medical community (262). The Thyroid Hormone Replacement for Subclinical Hypothyroidism Trial (TRUST) was a multi-centre, double blind, placebo controlled, phase III clinical trial testing the efficacy of thyroxine replacement in subclinical hypothyroidism in older community dwelling adults (121). The results of the TRUST trial were published in the New England Journal of Medicine on 3rd of April, 2017 (121). This Study Within A Trial (SWAT) was conducted at the Irish TRUST trial site prior to and after publication of results.

The aim of this SWAT was to investigate methods of disseminating trial findings to participants by using a PPI approach to identify, develop, and evaluate a patient-based approach of receiving trial results.

6.3 Methods

Study design

This was a sequential mixed methods study with three phases. In this study, methods were combined for complementarity, where each method addressed a different aspect of the study aim (265). The first phase used a qualitative approach to identify and develop a patient-based approach to disseminating the results, the second phase used a SWAT intervention to compare the dissemination approaches and the third phase used a quantitative patient understanding questionnaire to evaluate the patient-based approach. The full study protocol has been published elsewhere (266) but a summary follows here.

Setting

The study sites for the TRUST trial were the University of Glasgow, Scotland (lead site); Leiden Academy on Vitality and Ageing, The Netherlands; Leiden University Medical Centre, The Netherlands; University of Berne, Switzerland; and University College Cork, Ireland. A total of 738 participants with subclinical hypothyroidism were recruited to the trial over a three-and-a-half year period from 2013–2017 (121). The trial completed recruitment in November 2016 and the results were published in April 2017 (121).

This SWAT was conducted at the Irish TRUST site. The hub centre for the Irish TRUST site was located at the Mercy University Hospital, Cork where 38 participants were recruited. A further 77 participants were recruited from five satellite sites.

Population

As this SWAT was embedded in an ongoing clinical trial the study sample was determined by the TRUST Thyroid trial. There were 115 TRUST participants recruited in the Irish site, 11 of these participants withdrew over the course of the trial. Our study sample included all remaining TRUST participants (n=104).

Phase One: Identification and development of patient-based approach (qualitative and PPI phase)

The first phase of the study used a qualitative approach to iteratively identify and develop a patient-based approach to disseminate the results of TRUST trial. This was

done in three separate stages: qualitative focus groups, a PPI group and an adult literacy review.

Focus groups

Three semi-structured focus groups were conducted with four to eight TRUST trial participants per group. All Cork-based patients (n = 38) were contacted via letter and invited to participate. A €20 shopping voucher was given to all participants to cover travel expenses. Each session was led by trained qualitative researchers (WHS, ER, CH). A topic guide was used to guide the focus groups. The topic guide was reviewed and refined by all members of the SWAT research team (see Appendix 4.1: Focus group topic guide).

The Consensus-Oriented-Decision-Making (CODM) model was used to guide the group to reach a consensus (267). The CODM model is accepted as a flexible model for reaching decisions (267). In this study some of the steps were initiated by the focus group facilitator and others occurred naturally as a follow on from the previous step. Below is an outline of each of the seven steps of the CODM model and how they were used in this study:

1. Framing the topic: The focus group facilitator introduced the idea of sharing results with participants and provided some context on the reasons why results are/ are not shared with participants.

2. Open discussion: The facilitator asked the group whether or not they think results should be shared with trial participants and whether or not they would like to receive the results of the TRUST trial.

3. Identifying underlying concerns: The previous discussion naturally followed on to participants asking questions and expressing concerns about the result method, content and language that would be used.

4. Collaborative proposal building: The group worked together to agree on the important elements of the results in terms of result method, content and language.

5. Choosing a direction: This step occurred naturally as part of the previous step.

6. Synthesizing a final proposal: The facilitator re-iterated the proposal the group had agreed upon and asked the group for feedback.

7. Closure: This step occurred naturally as part of the previous step.

Analysis. Focus group recordings were transcribed verbatim and entered into NVivo Version 11 for data management during thematic analysis. Braun and Clarke guidelines (268) for conducting thematic analysis were followed. Initial focus group transcripts were analysed independently by two researchers (ER and AC). Each transcript was read multiple times (data familiarisation) and initial codes were identified. These codes were then used to identify emerging themes. Both researchers discussed emerging themes

and conducted further refinement. The refined themes were then discussed and agreed upon with other members of the research team (ER, CH, AC, KMS). Researchers (ER, CH, AC) then used the focus group findings to develop an initial draft of a patient-based approach for the dissemination of results (see Appendix 4.2: Draft one of patient-based result letter).

PPI group

A PPI group was established to develop and refine the content of the patient-based approach for the dissemination of results. During the focus groups, three TRUST trial participants volunteered to take part in the PPI group. In addition to these three PPI partners, an additional partner was identified from a previous qualitative research study undertaken by the research team. This individual was keen to learn more about research and expressed an interest in being involved in future projects. While this individual had previous experience of taking part in research (as an interview participant), she had no experience of taking part in a clinical trial or being involved as a PPI partner. Originally, we intended to conduct these sessions in a group format, due to difficulties with PPI partners' schedule commitments, one-to-one sessions were conducted. At the one-one session, a researcher (ER) and the PPI partner discussed the layout, content and language of the initial draft of the result method. Researchers and PPI partners worked together to edit different sections of the document. These discussions were not audio recorded but comprehensive field notes were taken by the researcher (ER). These notes were then collated by the researcher and used to further ensure that the results letter reflected PPI partners' perspectives and preferences.

Adult literacy review

While the PPI group had significant input into the format and language used in the patient-based approach, the research team felt that it would be of additional benefit to collaborate with the National Adult Literacy Agency (NALA) to ensure the document adhered to national 'Plain English' standards. These standards ensured that the information presented to trial participants was sufficiently easy to read and understand (literacy). This would help to ensure that trial participants were able to make sound health decisions based on the information presented (health literacy) (269). This review was an iterative process with several drafts exchanged for editing. Although the review was taken as an additional step to the published protocol for the study, the research team felt it was helpful to further ensure that the document was accessible and easy to understand.

At the end of the first phase of the study, a final draft of the patient-based result letter was approved by researchers, PPI group and adult literacy experts (see Appendix 4.3: Final draft of patient-based result letter).

Phase Two: Dissemination of trial results (intervention phase)

The second phase of the study used a SWAT intervention to disseminate the results of the TRUST Thyroid Trial to trial participants. This was done using a prospective, randomised, single blind, parallel trial design. It is important to note that when the term randomisation is used, it refers to the allocation of patients to intervention/control within the SWAT and not the TRUST Thyroid trial. Irish TRUST participants were

randomised to intervention or control groups using an online random number generator. The intervention group received the patient-based letter format (see Appendix 4.3: Final version patient-based results letter) and the control group received a copy of the TRUST results press release, which was made available by the lead study site on the TRUST Thyroid Trial Website (see Appendix 4.4: Standard results letter). Participants were blinded to their intervention group. One member of the research team was un-blinded in order to perform the randomisation and distribute the results of the trial. As they were un-blinded to perform these two important tasks, they were not involved in the data analysis or interpretation in any way.

Phase Three: Evaluation of patient –based approach (quantitative phase)

The third phase of the study used a quantitative patient understanding questionnaire to evaluate the patient-based approach to disseminating trial results. The questionnaire was developed in consultation with experts in the area of subclinical hypothyroidism and scale (questionnaire) development (PK and KMS). The early development of the questionnaire was guided by a consultation document, which accompanies the EU Clinical Trials Regulation No 536/2014 (270). This document highlights the information which should be presented to trial participants in the trial summary at the end of a trial. However, initial questionnaire items were modified to allow for psychometric testing. The final questionnaire contained 12 questions; six items were measured on a five-point LIKERT scale, there were four multiple-choice questions and two vignettes. The first six items measured patients' perceived understanding of results; the four multiple choice measured patients' actual understanding of results by requiring them to select the

correct answer. To further test participants' understanding of the trial results, two vignettes describing two typical patient case studies of older adults with subclinical hypothyroidism were provided with a question on whether a doctor should prescribe thyroxine for the hypothetical patient described. The questionnaire was reviewed by the PPI group to assess content and face validity. It then underwent further review by NALA to ensure adherence to the national 'Plain English' standard. The final version of the questionnaire can be seen in Appendix 4.5: Patient understanding questionnaire.

The questionnaire was sent to all Irish TRUST participants (intervention and control group) one week after they received the results of the trial. A reminder questionnaire was sent to non-responders 3 weeks later.

Analysis. The primary outcome was the difference in levels of patient understanding between the intervention and control groups. This measured the impact of PPI on patient understanding of end of trial results. The psychometric properties and construct validity of the questionnaire were examined with exploratory factor analysis. Principal component analysis (PCA) was conducted on the six LIKERT scale items. Internal consistency of the questionnaire was investigated using Cronbach's alpha coefficient. Completed questionnaires were entered into SPSS software (version 24) and analysed using descriptive and inferential (Chi-square test and Fishers Exact) statistics. The researcher carrying out data input and analysis was blinded to the participants' allocation status.

Costs of conducting PPI

The lead researcher (ER) kept a detailed account of all direct costs associated with conducting PPI for the purpose of this study. These costs included researcher salary, travel and expenses for PPI participants, adult literacy review and printing and postage costs.

This paper has been written in adherence to the Guidance for Reporting Involvement of Patients and Public 2 (GRIPP 2) (208). The GRIPP 2 checklist is a tool, developed to improve the reporting of patient and public involvement in research and guide the development of a transparent, consistent and high-quality PPI evidence base. The Good Reporting of a Mixed Methods Study (GRAMMS) framework was also used to inform the reporting of the findings (271).

6.4 Results

Characteristics of the trial participants stratified by participation in the different stages of the study are presented in **Table 6.1**.

Table 6.1: Characteristics of trial participants stratified by participation in the different stages of the study.

	Total Irish TRUST participants (n=104)	**Attended SWAT focus groups[1] (n=19)** Total Sample n=38 RR[2]=50%	**Randomised[3] (n=101)**		**Returned SWAT questionnaire (n=69)** Total Sample n=101 RR[2]=68%	
			Intervention Group (n=51)	**Control Group (n=50)**	**Intervention Group (n=38)** RR[2]= 74%	**Control Group (n=31)** RR[2]=62%
Sex						
Male	61 (58.7%)	14 (73.7%)	31 (60.8%)	28 (56%)	26 (68%)	16 (52%)
Female	43 (41.3)	5 (26.3%)	20 (39.2%)	22 (44%)	12 (32%)	15 (48%)
Age						
65-74	57 (54.8%)	12 (63.1%)	32 (62.7%)	24 (48%)	25 (66%)	12 (45%)
75+	47 (45.2%)	7 (36.9%)	19 (37.3%)	26 (52%)	13 (34%)	17 (55%)
Education						
Primary only	22 (21.2%)	2 (10.5%)	12 (23.6%)	9 (18%)	10 (26%)	8 (26%)
Secondary/ Tertiary	47 (45.1%)	12 (63.2%)	24 (47.1%)	22 (44%)	19 (50%)	11 (35%)
Unknown	35 (33.7%)	5 (26.3%)	15 (29.3%)	19 (38%)	9 (24%)	12 (39%)

[1]A subgroup of Irish TRUST participants (n=38) were invited to focus groups.
[2]RR=Response Rate
[3] Total Irish TRUST participants (n=104) excluding PPI partners (n=3) = n=101.

Phase One: Identification and development of patient-based approach (qualitative and PPI phase)

Focus groups

Three focus groups were held with 19 out of 38 participants accepting an invitation to join. Participants who attended the focus groups were similar in age, gender, education level to those who did not attend.

Focus group findings indicate that participants want to receive the results of the trial in which they are taking part. Three main themes emerged in relation to participants' perspectives of and preferences for receiving trial results: 'acknowledgement of individual contribution', 'contributing for a collective benefit' and 'receiving accessible and easy to understand results'.

Acknowledgement of individual contribution

Many participants reported feeling they had made an individual contribution to the trial in terms of their time and personal information while attending the trial study visits. As such, participants felt that receiving the results of the trial would provide an acknowledgement of this individual contribution:

> *'Yes, I mean it's kind of instinctive… when you go into a [clinical trial] and you spend and invest that time in it. I mean okay I had the time to invest but you know at the end of the day, [receiving the result] is kind of like your pay off.'* (FG2 P3)

Contributing for a collective benefit

While participants spoke about making an individual contribution to the trial, they felt that their involvement contributed to a collective benefit or greater good. Participants reported that receiving the results of the trial would help them to feel that they had contributed to this greater good:

> *'I'm not really interested in my own personal results but as the results of the scheme as a whole. You know the idea is, does the study help or hinder old people and that's what I want to know'* (FG2 P1)

This feeling of contributing for a collective benefit was further reinforced when participants discussed their desire to understand how the results of the trial will be implemented by medical experts and ultimately how it will affect others who have the condition:

> *'I would like to know, if they found out, okay, do we treat these people or not. That would be good. Do we treat them, or don't we treat them? I think that is what it's all about' (FG3 P4)*

Receiving accessible and easy to understand results

Participants expressed a clear need to receive the results of the trial in an accessible and easy to understand way. This preference applied to the format, language and content of the patient-based approach.

The majority of participants said they would like to receive the results in a letter format posted to them directly from the TRUST trial. Participants felt that this method would be accessible to them as they could read the results *'in text'* (FG3 P4) and keep a *'hardcopy'* (FG P1). While participants wanted an official statement of the results in a letter format, they also felt it was important to add a personal element to the letter. They suggested this could be done by offering participants a phone number that they could call if they wished to discuss any further issues or concerns with the TRUST study team:

> *'Could you attach a helpline on to it? If you know, somebody had some kind of serious medical question or that they thought was a bit personal element or whatever. That they'd like to talk to a medical person or whatever. Instead of just talking to your GP, maybe that would add another dimension of care around the TRUST'* (FG2 P3)

Participants agreed that the format, content and language of the results letter needed to be easy to read and understand. All participants wanted the letter to be no longer than 2–3 pages and presented in a question and answer format. Participants believed the content of the results letter should include *'pertinent information'* (FG1 P7) relating to the trial itself, the study drug (including side effects) and the results of the trial. They stressed the importance that this information needed to be informed by medical experts and *'from a good authoritative source'* (FG2 P2) but it should be presented to them in a language that fits their current context and could be easily understood by those who do not have scientific or medical backgrounds.

'Just in ordinary language that we can understand ourselves, you know that we don't want big and long explanation or that, just that we can pick it up straight away that it's without any huge number of pages. Just the bare, to me anyway, answers to the questions.' (FG3- P2)

It was evident from the focus groups that participants want to receive the results of the trial both to acknowledge their individual contribution to the trial and also help them to feel that they had contributed to a greater good. Participants expressed a clear preference to receive the results in an accessible and easy to understand way. These results were used by the researcher (ER) to develop an initial draft of the results letter (see Appendix 4.2: Draft one patient-based result letter).

PPI group

The initial draft of the results letter was then further iteratively developed by the PPI group. There were four PPI partners in total (three trial participants and one older adult) Each partner took part in one-to-one session. Each session contained an open discussion between the researcher (ER) and PPI partners on the layout, content and language of the document. Researchers and PPI partners worked together to write, re-write, edit and change different sections of the document.

Health literacy review

This draft was then iteratively reviewed and approved by health literacy experts from the NALA (see Appendix 4.3: Final version patient-based results letter).

Phase Two: Dissemination of trial results (intervention phase)

There was a total of 101 Irish TRUST participants randomised to the SWAT intervention. Trial participants from the PPI group (n = 3) were excluded from randomisation as they reviewed the content of the intervention method prior to the intervention. The intervention group (n=51) received the patient-based letter format (see Appendix 4.3: Final version patient-based results letter) and the control group (n=50) received a copy of the TRUST results press release, which was made available by the lead study site on the TRUST Thyroid Trial Website (see Appendix 4.4: Standard results letter).

Phase Three: Evaluation of patient-based approach (quantitative phase)

The overall response rate for the patient understanding questionnaire was 68% (69/101). The response rate for the intervention group was 74% (38/51) and the response rate for the control group was 62% (31/50). There were no significant differences in age, gender and education between those who returned the questionnaire and those who did not.

Post hoc power calculations showed that the study was underpowered to detect an effect. Power for each of the patient understanding components ranged from .01 to. 58.

Table 6.2 below shows the results of patients' perceived understanding of the purpose and context of the TRUST Thyroid Trial. Due to low participant numbers across the five Likert responses, the questionnaire response bands have been contracted from 'Strongly

Agree' and 'Agree' to 'Yes, 'Strongly Disagree' and 'Disagree' to 'No' and 'Neither agree nor disagree' to 'Neutral'. The results show that patients' perceptions of understanding are similar between the intervention and control groups. Subgroup analysis showed patient's understanding was not significantly impacted by age, gender or educational level.

Table 6.2: Patient perceptions of understanding presented by group[1].

Item	Group	Yes	No	Neutral	p-value
I understand why the TRUST Thyroid Trial took place.	Intervention (n=38)	37 (97.4%)	1 (2.6%)	0 (0%)	0.584
	Control (n=31)	29 (93.5%)	2 (6.5%)	0 (0%)	
I understand why I was invited to the TRUST Thyroid Trial	Intervention (n=38)	38 (100%)	0 (0%)	0 (0%)	0.198
	Control (n=31)	29 (93.5%)	2 (6.5%)	0 (0%)	
I know why the medicine Levothyroxine is used to treat subclinical hypothyroidism	Intervention (n=38)	32 (84.2%)	2 (5.3%)	4 (10.5%)	0.893
	Control (n=31)	25 (80.6%)	3 (9.7%)	3 (9.7%)	
I am aware of the side effects of Levothyroxine	Intervention (n=38)	30 (78.9%)	5 (13.2%)	3 (7.9%)	0.090
	Control (n=31)	17 (54.8%)	7 (22.6%)	7 (22.6%)	
I understand the impact of Levothyroxine on thyroid specific quality of life	Intervention (n=38)	31 (81.6%)	5 (13.2%)	2 (5.3%)	0.281
	Control (n=31)	20 (64.5%)	7 (22.6%)	4 (12.9%)	
I understand how doctors will use the results of the TRUST Thyroid trial to treat people with subclinical hypothyroidism	Intervention (n=38)	33 (86.8%)	2 (5.3%)	3 (7.9%)	0.878
	Control (n=31)	26 (83.9%)	3 (9.7%)	2 (6.5%)	

[1] Patient perceptions of understanding were assessed using a five-point LIKERT scale.

Figure 6.1 shows patients' actual understanding of the primary aim, side effect and results of the TRUST Thyroid Trial. Almost 82% (n=31) of the intervention group and 65% (n=20) of the control group correctly understood the primary aim of the TRUST trial (p=0.108). Almost 40% (n=15) of the intervention group and 36% (n=9) of the control group correctly understood the associated side effects of the active drug (p=0.734). In total 50% of the intervention group (n=19) and 58% of the control group correctly understood the results of the trial (p=0.504). There were no differences in patient understanding of trial results between the intervention and control groups.

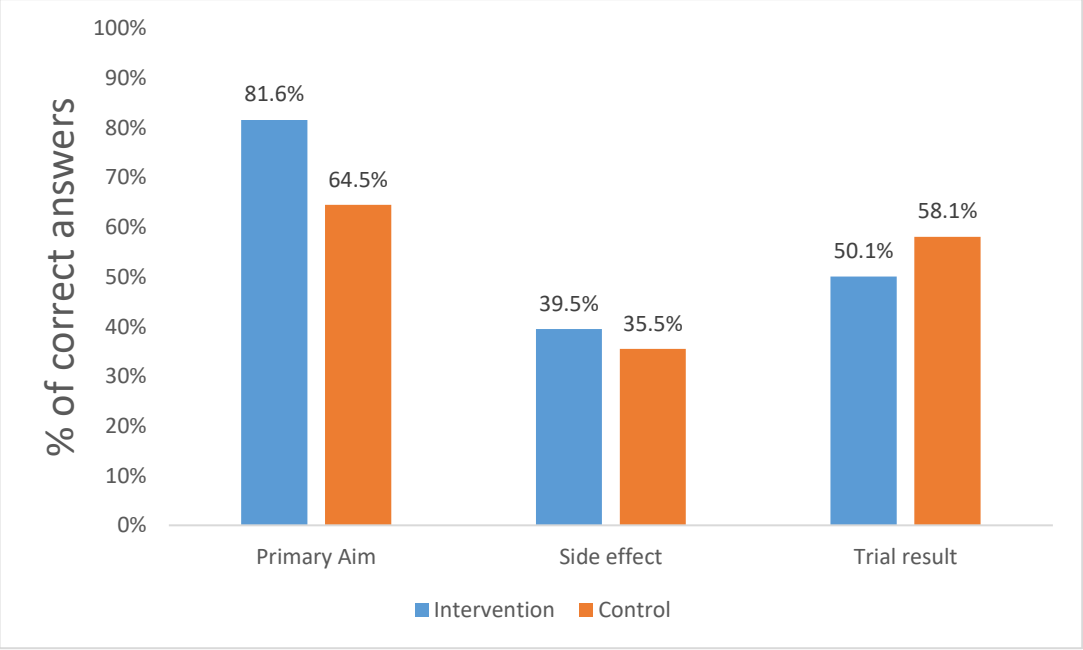

Figure 6.1: Patient understanding of primary aim, side effect and trial result of the TRUST Thyroid Trial presented by group[1].

[1] Patient understanding of primary aim, side effect and trial result was assessed using multiple choice questions.

In terms of patient understanding of hypothetical patient case studies, 43% (n=13) of the intervention group gave the correct answer to Vignette A; this was lower than the control group (62.1%, n=18, p=0.15). In total 77% (n=23) of the intervention group gave the correct answer to Vignette B, this was higher than the control group (66%, n=19, p=0.344).

Psychometric testing

An exploratory principal components analysis (PCA) was conducted on the patient understanding questionnaire to determine its usefulness as a measure of perceived understanding. The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis, KMO= .83. Bartlett's test of sphericity indicated that the correlation matrix was significantly different from an identity matrix, X2 (.852) = 283.92, p<.001. An examination of eigenvalues greater than Kaiser's criterion of one, suggested the extraction of one factor; this was supported by inspection of Cattell's scree plot. An examination of the constituent items for this factor structure also indicated that items loaded most highly on a single factor. This single factor represents a measure of perceived understanding of trial results. PCA was then conducted using an oblique (direct oblimin) rotation, specifying the extraction of one factor. This model explained a combined 69.58% of the variance in patients understanding of the TRUST thyroid trial.

Cost of conducting PPI

The total cost of this study amounted to €8,049 (see Appendix 4.6: Costs of conducting PPI).

6.5 Discussion

While PPI is increasingly recognised as an important element of clinical research, evidence on optimal methods and potential impact is lacking (53, 260). Previous research conducted on the impact of PPI has largely focused on the experiences of participants and researchers (190) and on the research process in broad terms (77). In this study, our primary outcome was specific: a quantitative measure of patient understanding of trial results between those who received the patient-based approach and the standard approach. To our knowledge there has been no previous research conducted on the impact of PPI on patient understanding of trial results.

The involvement of clinical trial participants in this study offered insightful perspectives on the information needs of the study population in terms of receiving end of trial results. Study findings show that trial participants want to receive the results of the clinical trial in which they had participated. This is supported by much of the available literature on patients' preferences of receiving results, with up to 90% of participants in previous studies reporting a desire to receive results (272). Focus group findings showed that participants felt that receiving results would provide an acknowledgement of their individual contribution to the trial. This finding complements previous commentaries about result sharing being an 'ethical imperative or 'moral obligation'. Fernandez et al. points out that many participants place their trust in science and researchers owe a debt to participants to fulfil their trust and recognise their altruism (120, 273).

Unsurprisingly, findings also show that participants want to receive results that are accessible and easy to understand. In this study, the preferred format of receiving results was a letter posted to them directly from the TRUST trial. This preference is also consistent with the literature on patient preferences of receiving results. A previous study investigating preferences of individuals taking part in a cardiac rehabilitation trial found that 80% of trial participants preferred to receive the results by post (274). The patient-based approach identified in this study was feasible for researchers to develop with significant involvement from trial participants and adult literacy experts.

Previous studies exploring participants' reactions found that sharing trial results with participants can cause some negative impacts such as anxiety, anger, guilt, upset and confusion (275-277). As far as researchers in this study are aware, providing results did not cause any negative impacts. This may have been due to the fact that the TRUST trial had a low risk of morbidity or mortality compared to some of the other studies citing negative impacts. Both result methods contained the telephone number, email address and postal address of the research team and participants were urged to contact should they have any questions or concerns relating to the study. The research team did not receive any queries.

Previous systematic reviews highlight the lack of evidence on economic analysis of PPI and call for researchers to consider the costs of its implementation (77, 98). As discussed previously research funders are increasingly demanding that PPI be carried out in research. However, the costs of PPI are often underestimated and can cause a significant financial burden on research project budgets (77, 98, 278, 279). It is extremely important

that researchers plan PPI at the grant proposal stage and estimate the costs appropriately. If these costs are not correctly estimated during the initial stages of developing research proposals, they may cause a financial burden on PPI partners.

Participants in this study were not paid for their time but were provided with a €20 voucher to cover travel expenses. When PPI is not the primary focus of a study, researchers do not consider the cost implications at the beginning of the study and are often tied with limited resources to carry out PPI (278-280). INVOLVE, the national advisory group supporting active public involvement in health services, public health and social care research in the UK, have recommended that PPI partners should be paid for their involvement (281). Despite this, existing research suggests that institutional difficulties make negotiating the mechanisms of paying participants very difficult (278). One study reported that in order for participants to be remunerated for their efforts, they needed to be registered as employees, a process that incurred much paperwork and time delays (278). This study outlines the cost of conducting PPI and includes a full breakdown of costs (see Appendix 4.6: Costs of conducting PPI). This breakdown provides a template to other researchers who plan to carry out and evaluate PPI as part of their research. It is important to note that not all costs associated with carrying out the study were included in this amount. For example, the only salary costed was that of the research assistant. The expertise provided by other members of the study team were not included in the total cost as they were being paid by the University or other research grants. The total cost of conducting this study was €8,049 which is not insignificant but should be considered in the context of the cost of large-scale trials.

6.5.1 Strengths and Limitations of the study

While this study provides important insights into patients' preferences of receiving trial results, it is not without limitations. Firstly, existing PPI literature states that 'to understand the research needs and challenges, PPI has to engage people who are able to offer perspectives from the study population' (52). All PPI partners in this study were active members of the research community as they had taken part in the TRUST trial and had agreed to long-term follow up. This is a strength of the SWAT as they were able to offer perspectives from the study population, however it does have an important implication for their reporting of understanding the results of the trial. They may be more inclined to rate their understanding as high because of their investment in the trial (282), thus potentially minimising differences between the intervention and control conditions and minimising inferences that can be drawn about the intervention. Previous research suggests that people that actively choose to engage in research either as research participants or involvement partners are more likely to be middle-class and highly educated (18, 283). In this study, those that attended the focus groups and PPI group were similar in education level to those that did not attend. This is not surprising considering the entire study sample had already actively volunteered to take part in the TRUST trial.

Secondly, the results of the patient understanding questionnaire show that the levels of patient understanding were similar between the two groups. However, this study was underpowered to detect an effect. As this was a Study Within A Trial (SWAT), the power was limited by the sample size that was available to us from the trial (n=115).

Furthermore, validation of the patient understanding questionnaire was limited by the sample size in this study. While validation of the questionnaire was limited, exploratory factor analysis provided some evidence that the questionnaire is a useful tool for measuring patient understanding of trial results. The developed questionnaire can be tailored for use in other trials in future examinations of patients understanding of trial results. This would provide insight into patient understanding and provide further validation data.

Thirdly, all SWAT participants were aged 65 and over. The layout, format and language of this patient-based approach which was identified and developed may only be relevant for this study population. Other trial populations may prefer to receive the results via email, online or in person from a member of the study team (120). The evidence on patient preferences of receiving trial results is limited, therefore further research is needed to explore patient preferences of receiving trial results amongst different study populations.

It is also important to point out that the control group in this study received a copy of the trial results in a press release format. Most trial participants do not receive this. While this control method was a step further than normal procedure, the researchers in this study felt this was appropriate. The information presented in the press release was similar to that of the patient-based approach. However, the format and layout of the press release was different. Information was written in four long paragraphs separated by individual headings. It was also much shorter (1 page in total) that the patient-based approach (3 pages in total). Given the fact that press releases are written by public

relations professionals with a view to communicating effectively and efficiently, this may have potentially minimised differences between the intervention and control conditions. The primary outcome of this study was assessing the impact of PPI on patient understanding of results, however, this was not the only potential impact. In hindsight, we adopted a limited approach to PPI in this study as we did not involve our PPI partners from the outset of the SWAT. Involving PPI partners in the development of core outcome sets for this SWAT could have identified other more appropriate primary outcome measures (284).

The aim of this SWAT was to investigate methods of disseminating trial findings to trial participants by using a PPI approach to identify, develop and evaluate a patient-based method of receiving trial results. The PPI approach actively involved focus group participants in making decisions about the result method and worked with PPI partners to co-develop the result letter. However, PPI partners were not involved in other aspects of the research process such as research design, data collection or analysis. This is partly due to the fact that PPI is a relatively new concept in clinical trials. As the majority of the literature has only been published in the last 12 months, there is little evidence available on the impact of PPI and no gold standard or comprehensive guidelines for researchers to follow (274). Thornton (259) suggests that in order for PPI to develop it is important to record its social and cultural history by collecting comprehensive databases and undertaking ongoing reviews of the impact of PPI. This paper along with the study protocol have been written in adherence with the Guidelines for Reporting Involvement of Patients and the Public (208), thus providing templates for involving patients and the public in clinical trial design and development. This study is an important step forwards

in documenting the process of conducting PPI as part of a SWAT and evaluating its impact. Future research is needed to further develop PPI in clinical trial settings. As there is currently no gold standard or comprehensive guidelines for researchers to follow when evaluating the impact of PPI, further research is needed. This research should involve PPI partners in the development of core outcome sets for evaluating PPI impact. These would significantly enhance the literature in the area.

6.6 Conclusion

Patient and Public Involvement (PPI) is advocated for every step of the trial process. We have demonstrated that it is feasible to involve PPI partners in the development of dissemination materials. Sharing clinical trial results with participants in a format understandable to laypersons will soon be a legal requirement[11]. However, there is a significant lack of evidence as to the most appropriate methods of sharing results with participants. The study identified and developed a patient-based approach to disseminating clinical trial results for trial participants. Although, in this study PPI did not influence patients' final understanding of results, it documents the process of conducting PPI within the clinical trial setting. This process may be useful for other trialists interested in conducting and evaluating the impact of PPI in clinical trials.

7. Discussion

7.1 Overview

This thesis aims to contribute to the evidence on the methods and impact of PPI by exploring PPI contributors' experiences and contributions at the design, conduct and dissemination stages of trials. Current reports on the methods and impact of PPI are 'piecemeal and inconclusive' (9). This research demonstrates how we can progress from relying solely on researchers' anecdotal reflections and limited qualitative studies on the perceived impact of PPI towards developing a more robust evidence base on the methods and impact of PPI on three specific stages of the trial process: design, conduct and dissemination. To do this, a suite of study designs were used including qualitative, mixed methods, systematic review and narrative synthesis, and an embedded randomised controlled trial. This research also demonstrates that it is feasible to conduct and evaluate PPI in the design, conduct and dessemination of trials while following the core principles of PPI as defined by INVOLVE (23).

This final chapter summarises the main findings, the strengths and limitations of the thesis, the implications, and suggestions for future research. This chapter will also present a reflection on PPI during the COVID-19 outbreak and will close with a brief conclusion.

7.2 Summary of main findings

This research shows that while there are a wide variety of approaches and methods used to involve PPI contributors, the approach or method used can have an important influence on the impact of involvement. The systematic review on the perceived impact

of PPI on trial retention highlighted that PPI contributors are often involved simultaneously with other stakeholders on trial advisory committees and stakeholder advisory boards. However, the research on PPI involvement during intervention development showed that involving PPI contributors simultaneously with other stakeholders (healthcare professionals) can lead to a perceived lack of common ground where both stakeholder groups can feel undervalued by the other group and reluctant to express their opinions. Furthermore, there were more instances of conflicting opinions when both types of stakeholders were involved simultaneously in the same group, than when each stakeholder group was involved separately. In this research, these conflicting opinions were difficult to disentangle and led to researchers being unsure about how to incorporate their opinions into the final intervention that was developed. These findings suggest that it may be more suitable and useful to involve PPI contributors separately rather than simultaneously with other stakeholders. This learning is an important contribution to the existing limited evidence on suitable PPI methodologies and is consistent with existing literature on the productivity of incongruent groups that is, when groups contain smaller subgroups of individuals with conflicting knowledge or experiences. These subgroups can create a division between group members, undermining the groups' ability to work together and be productive (149).

This research found that PPI can influence the research process by creating and fostering trust between researchers and participants. The systematic review showed that researchers perceived PPI to have a positive impact on trial retention as PPI contributors helped research teams to develop trust with participants. When advising on the delivery

and content of trial result dissemination materials, although trial participants wanted the results in a format that was easy to understand, they stressed the importance that this information be informed by medical experts and from a source they could trust. Previous research has highlighted that trust between research participants and researchers is paramount to successful research (250). Factors affecting trust can vary greatly depending on the study target community. For example, a previous study exploring factors affecting trust between research participants and research teams found that non-indigenous people were more likely to base their trust on the general reputation and credentials of the institution in which the research was taking place whereas non-indigenous people tended to base their trust on the face-value and likeability of the researcher (251). PPI contributors also advised the research team on how they can capitalise on existing trusting relationships between patients and healthcare professionals to improve health outcomes. When advising on who should deliver the message to attend retinopathy screening, people with diabetes recommended that the GP deliver the message as people have a relationship with, and trust, their GP and are much more inclined to listen to them. The results contained in this thesis shows how PPI contributors can help researchers to develop and adapt specific trust building measures to suit the particular trial context and target population.

This research found that PPI contributors can help researchers to communicate more effectively with research participants. When advising on how a message to attend diabetic retinopathy screening should be delivered to people with diabetes, PPI contributors recommended that a letter would be the most direct way of contacting participants as it comes to the house so you have to open and read it. They also gave

insights on how the letter would be received from a patient perspective such as sending the letter in a plain envelope so people would not feel apprehensive about opening a letter from their GP practice that could contain negative results. Similarly, the systematic review highlighted that PPI contributors across different trial contests advised on the mode, frequency and format of study reminders and helped to ensure trial materials were understandable when they were not in trial participants primary language. Furthermore, trial participants advising on trial dissemination materials recommended that the results be sent in a letter format. They also stressed the importance of communicating in a format that was accessible to participants and easy to understand.

Throughout this research, the lines between individuals taking part in the research as research participants and being involved in the research as PPI contributors were blurred, as is often the case with PPI (12, 164). For example, although people with diabetes took part in the consensus meetings as research participants, their role was to discuss and make decisions about the intervention content and mode of delivery which fits with the PPI definition and principles (33, 165). Similarly, although TRUST trial participants took part in focus groups, their task was to outline their preferences for trial result dissemination which was ultimately used to develop the result dissemination method which can be defined as PPI. These lines were further blurred with the added complexity of evaluating the impact of PPI. For example, PPI contributors became research participants when they completed the experience survey or patient understanding questionnaire. Researchers evaluating PPI should be mindful of these blurred lines and ensure they establish clear role expectations with PPI contributors so

contributors are fully informed as to when they are expected to be involved as PPI contributors and take part in the research as research participants.

7.3 Strengths and limitations

In addition to the strengths and limitations discussed in each chapter, the overall strengths and limitations of the thesis are outlined below.

This thesis generates evidence on the methods and impact of PPI in randomised trials. This includes qualitative, mixed methods and an embedded randomised controlled trial. The reporting of these designs was strengthened by the use of relevant reporting checklists: Consolidated Criteria for Reporting Qualitative Studies (COREQ), Good Reporting of A Mixed Methods Study (GRAMMS) framework and Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) statement (133, 134, 209). This thesis demonstrates how appropriate methodology can be employed to overcome previous limitations and provide evidence on the methods and impact of PPI in the context of trials.

Secondly, the extensive and continued involvement of PPI contributors is a key strength of this research. Throughout each of the studies presented in this thesis, I employed a combination of the *consultation* and *collaboration* approaches to involvement as defined by the 'levels of involvement' theoretical model originally put forward by Boote, Telford and Cooper and adapted by INVOLVE (7, 32). I consulted and collaborated with PPI contributors to devise the study design and development (Chapters 3, 4 and 5), inform study recruitment materials and processes (Chapters 3 and 4), design study materials

and data collection processes (Chapter 4 and 6), interpret data synthesis (Chapter 5), inform the content, layout, format and delivery of research findings (Chapter 6) and review and contribute to drafts of peer reviewed manuscripts (Chapters 3, 4, 5 and 6). The involvement of PPI contributors also shaped my attitude as an early career researcher. Although trial methodology research ultimately aims to improve the health of patients and the public by improving how trials are carried out, the non-patient facing nature of trial methodology research often meant that I felt disconnected from the community whose health I was aiming to improve. Conducting PPI helped me to connect with that community and realise the purpose of my research.

The results of this research are timely. This research was conducted as the supportive environment for PPI continues to be created by research funders, academic journals and research ethics committees. As many researchers are now required to incorporate PPI as an integral part of their research, there is an emerging appetite amongst the research community to increase their understanding of effective PPI methods and its impact on the research process. This research has contributed to the development of a supportive environment for PPI in health research as I have had the opportunity to present this work at various national and international conferences in the areas of PPI, health services and public health (Appendix 6), record a podcast series to promote PPI in research (see Appendix 5), publish blog posts on the HRB Open and Structured Population and Health Services Education (SPHeRE) platforms (see Appendix 5) and deliver an educational seminar for researchers at University College Cork (see Appendix 5). Throughout these activities, the research was well received and won a number of awards (see Appendix 8).

This research captures the complex nature of PPI. The definition of involvement adopted throughout the duration of this research was the INVOLVE definition of PPI. INVOLVE defines PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them'(1). This broad definition was considered the best fit for this research as it allows for the inclusion of participatory research approaches and activities that share the same principles as PPI but may not necessarily be labelled as PPI in the literature. For example, the inclusion of people with diabetes and healthcare professionals in the studies presented in Chapters 3 and 4 of this thesis fits with the Community Based Participatory Research (CBPR) approach which aims to include all relevant stakeholders as partners, rather than excluding health professionals from the process (285). Additionally, previous systematic reviews have tended to focus their search terms solely relating to 'patient', 'public' and 'involvement', however the systematic review presented in Chapter 5 of this thesis included search terms relating to a wide range of participatory research approaches including CBPR and participatory action research. The inclusion of a wide variety of terms allowed for the transfer of learnings from participatory research approaches which have a much longer history in the social sciences compared to that of the relatively recent tradition of PPI in health research (286).

There are a number of limitations to the research carried out in this thesis. Some of the survey instruments used to collect quantitative data in this thesis were not validated. Given that PPI is a relatively recent phenomenon and the majority of the evidence on evaluating the impact of PPI has been published in the last three years, validated tools to evaluate its impact are non-existent. The questionnaire that was used to understand

participants experiences in Chapter 3 was based on non-validated questionnaire items from a previous study aiming to evaluate dimensions of group dynamics within community-based participatory research partnerships. I was unable to conduct exploratory factor analysis as the study sample size did not meet the minimum criteria for validation. The questionnaire used to evaluate participants' understanding of trial results in Chapter 6 was also not validated. Although exploratory factor analyses provided some evidence that the questionnaire is a useful tool, this would need to be further explored in future studies that aim to evaluate the impact of PPI on participants' understanding of trial results.

Despite using a range of strategies to recruit PPI contributors for each study, another potential limitation is the lack of diverse PPI contributors. For example, the absence of people with type 2 diabetes involved in the combined consensus meeting of people with diabetes and healthcare professionals could have changed the nature of the relationship between patients and HCPs and led to different participant experiences and group dynamics. There was also an absence of individuals from 'hard to reach' or 'seldom heard' communities. For example, although members of the travelling community are twice as likely to have diabetes than members of the general population (287), no members of the travelling community were involved in this research. Diversity has been noted as a significant issue in PPI and needs to be considered in all research studies to allow a broad range of perspectives to be taken into account and to promote equal access to opportunities for public involvement (288). I made significant efforts to promote involvement opportunities (see Appendix 1.2) and ensure the location of involvement activities was easy to find and access. One way to promote involvement

activities would be to collaborate with community representative organisations and community groups and work together with them to promote and conduct involvement activities away from formal project or organisational structures (289).

7.4 Implications

While research funders, ethics committees and academic journals increasingly require PPI (3, 10), the results of this research will have important implications for researchers, patients and members of the public and research funders.

The findings suggest that involving PPI contributors simultaneously with other stakeholder groups may not be the most suitable approach to involvement as it can lead PPI contributors to feel undervalued and reluctant to express their opinions. Although further research is needed to explore whether this finding can be applied to other research contexts, it could potentially be generalizable well beyond randomised controlled trials. Patients and members of the public are increasingly being involved in health and social care research simultaneously with other stakeholders in the form of project steering committees, research advisory boards, community advisory boards etc. (9, 30). Therefore, researchers need to pay careful attention to the methods used to involve PPI contributors to ensure they are comfortable contributing and are enabled to make meaningful contributions to research decisions. This will enable researchers to avoid tokenistic involvement and help research funders to judge the appropriateness and quality of PPI in research proposals (14, 16).

Secondly, the findings show that PPI contributors can make unique and original recommendations that are subsequently incorporated into the intervention development process. The findings also show that PPI can potentially improve trial retention through a number of different mechanisms. Again, these findings can be applied beyond the trial context. Participant follow-up has been identified as a significant barrier in longitudinal study designs (290). Researchers conducting longitudinal studies or otherwise aiming to follow-up research participants can work together with PPI contributors to strengthen these identified mechanisms to ensure successful follow-up.

For researchers that are reluctant to conduct PPI, this evidence on the impact of PPI can help to fully understand the benefits of involvement so they can undertake more than just a tick-box approach to obtaining grants (16, 42). For researchers already conducting PPI, this evidence highlights important opportunities for maximizing PPI impact (14, 16).

For members of the public, Popay et al. states that they will benefit from robust evidence on the impact of PPI as they can understand how their contributions make a difference (12). However, it is likely that the publication format of this research (peer-reviewed publications and academic thesis) will not reach the public domain. Although I made some attempts to ensure that evidence on the methods and impact of this thesis were available to members of the public, including the podcast series, seminar and blog posts presented in Appendix 5, further efforts need to be made to share this robust evidence with members of the public and PPI contributors. For example, researchers are

increasingly designing and delivering PPI training courses for PPI, incorporating evidence-based findings into these training courses would better equip contributors to become involved and help to establish clear expectations around what involvement can achieve.

7.5 Suggestions for future research

This research has important implications for how researchers should approach future evaluations of the methods and impact of PPI. Table 7.1 outlines the specific recommendations for future research from each of the chapters in this thesis. However, this research also has broader implications for future research on the methods and impact of PPI. The complexities of the context and process of involvement can result in researchers thinking that PPI is too controversial or complex to be studied (14). This research suggests that nothing is too complex if we use the right methods. The use of mixed methods designs in this thesis allowed for an in-depth exploration which resulted in a more complete understanding than using qualitative or quantitative methods alone. The systematic review and narrative synthesis identified preliminary mechanisms for PPI impact that can be further tested to generate more robust evidence of impact. Although the results of the embedded randomised trial suggested that PPI did not make a difference to participants' understanding of results, this is not the first embedded randomised controlled SWAT to show limited evidence of the impact of PPI. In 2017, Cockayne et al. conducted a randomised methodology trial to evaluate the effectiveness of optimised patient information sheets on the recruitment of participants in a falls

Table 7.1: Suggestions for future research from each chapter

Chapter #	Finding / Limitation	Suggestions for future research
Chapter 3	Involving people with diabetes and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately.	Given the wide range and complex nature of factors that could have influenced this finding, future research should explore stakeholders' experiences and group dynamics in other participatory research contexts.
	The questionnaire used to evaluate participants' experiences was not validated.	The questionnaire can be easily adapted for use in other research contexts. Future research using the questionnaire would allow for reliability testing and validation to be carried out.
Chapter 4	Involving people with diabetes and HCPs simultaneously in a consensus process was not found to be as useful as involving each stakeholder group separately.	Future research should examine the contributions of different stakeholders in other research settings to determine whether this finding can be generalisable beyond the context of this study.
	Practice administrators were identified as playing a key role for the successful implementation of the intervention.	Future primary care research needs to involve practice administrators to ensure that all voices are heard in the research process.
Chapter 5	Several mechanisms were identified, through which PPI could potentially improve trial retention.	Future embedded randomised trials could compare the involvement of PPI contributors in making decisions about the mechanisms identified in this study (intervention) to standard trial conduct (control) on trial retention (outcome). Such quantitative evaluations could be enhanced using qualitative methods to further explore trial participants' perspectives.
Chapter 6	Participants in this study were aged 65 and over. The layout, format and language of this patient-based approach which was identified and developed may only be relevant for this study population.	Further research is needed to explore patient preferences of receiving trial results amongst different study populations.
	PPI partners were not involved the design of the study.	Future research evaluating the impact of PPI in trials could involve PPI

		partners in the development of core outcome sets for evaluating PPI impact. These would help to ensure that the research questions being asked are of importance to both trial researchers and trial participants.
	Exploratory factor analysis of the patient understanding questionnaire provided some evidence that the questionnaire is a useful tool for measuring patient understanding of trial results.	The developed questionnaire can be easily tailored for use in other trials in future examinations of patients' understanding of trial results. This would provide insight into patient understanding and provide further validation data.

prevention trial which similarly showed PPI to have no effect (202). While it is plausible that PPI did not have an effect, there is also a possibility that purely quantitative approaches to evaluate the impact of PPI that do not incorporate contextual factors and the specific mechanisms by which PPI has an impact into their design may weaken the evidence of impact or produce no evidence of impact (66). Future quantitative methods to evaluate the impact of PPI should be enhanced with qualitative methods to ensure that contextual factors and specific mechanisms for impact/ no impact are considered which would result in a deeper understanding (291). For example, this research identified building trust as a potential mechanism for PPI to have a positive impact on trial retention. Future embedded randomised trials could measure this impact by using a quantitative validated instrument to measure participants' trust and conduct qualitative interviews with trial participants to further explore whether actions taken as a result of PPI input had an influence on their trust levels. Furthermore, realist evaluation is one of several theory-based approaches to evaluation developed within the social sciences, which seeks to address dynamism and context, rather than control for them,

to explain what works for whom in which circumstances (292). As the findings of this research highlight the interplay between the context, process and outcomes of PPI, further evaluations of PPI methods and impact could also adopt a realist approach.

7.6 Reflection on PPI during the COVID-19 outbreak

Just like our lives, our deaths should not be defined by our health conditions.

Written on 25th March, 2020.

444 confirmed cases of COVID 19 on the island of Ireland.

16 people have died as a result of the virus.

As a PhD student, I have spent the last 3 years looking at ways to involve more patients and members of the public in the health research process. I have worked on a number of different research projects aiming to create and facilitate equal partnerships between researchers, healthcare professionals and patients/ members of the public. I have spent this time finding ways for everyone's voices to be heard and bridging the gaps between the people who live with health conditions and the researchers and healthcare professionals who aim to help them. From these experiences, I have learnt that Patient and Public Involvement (PPI) can democratize the research process and lead to real insights that have the potential to improve the quality, relevance, and accessibility of health research to the wider public.

One thing I have learnt during this time is the importance of language. If we really want to work together to achieve common objectives, we need to use the right language. A language that is accepted and understood by all partners involved in the process and a language that does not stigmatize. Language is perhaps the most valuable tool we have as humans. It is important in not only conveying our meaning, but also as an instrument of positive change.

Working with people with diabetes, for example, I have learned to refer to them as 'people with diabetes' and not 'patients' or 'service-users'. They may visit a doctor or 'use' a service for a few hours each year and so they rightly argue that this should not define who they are. The term 'diabetic' has also been dissipated, again their lives should not be defined or labelled by a health condition

Using language that is inclusive and values-based can lower anxiety, build confidence, educate, and empower. Poor communication can be stigmatising, hurtful and disempower. To put it simply, language can be unifying or divisive. Our terminology and tone are vital when trying to level the power dynamic and bring everyone together to work on a common goal.

Throughout this difficult and unprecedented time, it has been so uplifting to see members of the public, healthcare professionals, politicians etc. rally together to achieve a common cause. To fight the COVID-19 virus. Today's trending hashtags on Twitter include #wearewithyou, #proudtobeirish, #weareinthistogether and #weshallovercome.

But if you look closely enough, you can find traces of language which does not have the same unifying effects. Each day, the number of deaths due to COVID-19 are reported on followed by the same three words- 'underlying health condition'. Every time I hear these three words, I ask myself why are they necessary? What is their purpose?

To inform us? So that we can evaluate the potential risk? If the intention is to inform then a mere three words do not give us enough information. What underlying health conditions? How severe were they? Would they have died if they hadn't contracted COVID-19? If the information is intended to inform then it could be presented in a more informed way. Perhaps, a separate statement could be issued which lists each health condition and the percentage of those that have died with each condition.

Of course, it is likely to be the case that some people are more at risk of developing serious complications from the virus than others but sweeping statements that imply that all 'underlying health conditions' carry the same risk do not inform us nor allow us to evaluate our own risk or the risk to those we care about.

Nor do they unite us. These three words are powerful enough to divide the population into two groups. Those with an underlying condition that can live in dread of what may happen to them if they get the virus. And those without an underlying condition that can bask in their increased sense of security. In reality, we are all at risk, whether we have an underlying condition or not. For every death that has occurred as a result of COVID-19, there is a lot more than an 'underlying health condition' lost, there are families, friends, neighbours and colleagues that have been deeply affected.

In a country that is spectacularly working together, we need to get rid of these kinds of daily utterances that tease us apart. Some of us may have an underlying health condition and some of us do not. It doesn't mean that certain people's lives are more expendable or that their right time to die is any nearer or further away.

7.7 Reflection on the INVOLVE definition of PPI

Throughout the course of this research, I adopted the INVOLVE definition of PPI. INVOLVE defines PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them' (1). This definition was integral to my research because although PPI is currently somewhat of a 'buzz' term in the health research community, there is still some confusion around what PPI is (and what it is not!). So, whether I was writing an abstract or manuscript draft, or speaking at conferences or educational seminars, my opening line was always the INVOLVE definition. The INVOLVE definition is everything we want PPI to be -broad ranging, inclusive, non-offensive. Even the use of the phrase 'rather than' instead of 'as opposed to' results in a more friendly and non-confrontational tone. I repeated the definition over and over again. I liked saying it and I always felt that people liked reading and hearing it.

To me, the INVOLVE definition successfully conveys the culture and ethos of PPI. But as I progressed through this PhD research, I started seeing the INVOLVE definition as more of an 'umbrella' definition for PPI. Its broad ranging and inclusive nature meant that it lacked specificity. This was useful at times as it allowed me to explore the methods and impact of a wide variety of involvement approaches and methods, including once-off involvement in Chapters 3 and 4, participatory approaches in Chapter 5 and task-specific

involvement in Chapter 6. But casting such a wide net also caused problems. For example, while conducting the search for the systematic review in Chapter 5, 18,453 articles were identified for title and abstract screening. This felt like a never-ending process. Adopting a broad definition of PPI also shaped the implications of the findings in this thesis. My research shows that the context and nature of involvement can have important implications for its impact but I cannot definitively say 'this type of involvement leads to this type of impact'.

And so, each time I repeated the INVOLVE definition, I became more aware that I also needed to provide some specificity. I followed the definition with information on the different levels or approaches to involvement. I wrote/ spoke about Arnsteins' seminal ladder of involvement (33) which has been recently refined by Boote, Telford and Cooper (7). I wrote/ spoke about what PPI looks like at different stages of the research process. I always highlighted the importance of involvement principles such as respect, support, transparency, responsiveness, diversity, and accountability; all of which are discussed in detail in the background chapter of this thesis. This additional information was given with the aim of creating a clearer picture of what PPI looks like in practice and enabling myself and other researchers to expand beyond the definition to successfully operationalise and evaluate PPI activities.

7.8 Conclusion

This research shows that although there are a wide variety of methods used to involve PPI contributors, the method used can have an important influence on the impact of

involvement. The results suggest that it may be more suitable and useful to involve PPI contributors separately rather than simultaneously with other stakeholder groups. This finding may assist researchers and PPI contributors in designing and conducting more meaningful and effective involvement activities. This research found that PPI can influence the research process by creating and fostering trust between researchers and participants and PPI contributors can help researchers to communicate more effectively with research participants. Although, the results suggest that PPI did not make a difference to participants' understanding of results, suggestions for how researchers should approach future evaluations of the methods and impact of PPI have been put forward. This research paves the way forward for building an evidence base for PPI to ensure that a shared understanding of what works, when, how and why is developed among researchers, patients, members of the public and research funders.

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9. Appendices

Appendix 1- Supplementary Data for Chapter 3

Appendix 1.1: PPI Recruitment Flyer

Appendix 1.2: PPI recruitment strategies

The information flyer was distributed over an 8-week period from 11/08/18 – 11/10/18. The strategies used to circulate the flyer included social marketing (e.g. social media), community outreach (e.g. community and religious groups), health system (e.g. GP practices and hospital waiting rooms), and partnering with community and advocacy organisations (e.g. national organisations and educational institutions).

Recruitment strategy	Details	Number	Timing	Response (n)
Social marketing recruitment	We asked our PPI partner (diabetes advocate and administrator on 'Diabetes in Ireland' Facebook support group) to post the information flyer on Facebook and twitter.	4 social media posts	17/08/18-03/09/18	16
	We posted the information flyer on our research team twitter page (@ESPRIT_UCC).	3 social media posts	12/09/18-08/10/18	
Community outreach recruitment	We circulated the information flyer to local community and religious groups online and asked them to advertise on newsletters etc.	11 'Men's shed' initiatives 6 religious' groups	05/10/18 06/10/18	0
Health system recruitment	We left information flyers in GP practices, hospital waiting rooms and local diabetes clinic	8 GP practice waiting rooms 1 hospital waiting rooms	31/08/18-11/10/18 (6 weeks)	3

	(on tables and noticeboards).			
	We spoke about our research (5-minute overview) at diabetes support groups and distributed information flyers to attendees.	1 diabetes support groups 2 diabetes education sessions	18/09/18 12/09/18 + 14/09/18	17
Partnering with other organisations	We contacted a number of different organisations and asked them to circulate our information flyer to their email list and on their websites.	5 national organisations and educational institutions	27/08/ 2018 05/09/2018	0

Appendix 1.3: PPI Recruitment Survey Results

Table A 1.3: PPI Recruitment Survey Results

Variable	Total (N=20)	%
Age		
25-44	5	25
45-64	9	45
65-84	6	30
Gender		
Female	12	60
Male	8	40
Location		
Urban	14	70
Rural	6	30
Healthcare cover		
Full Medical Card	7	35
GP Visit Card	1	5
Private Health Insurance	13	65
Nationality and Ethnicity		
White Irish	20	100
Education*		
Junior Certificate/ Intercert	1	5.3
Apprenticeship	3	15.8
Leaving Certificate	5	26.3
Diploma	2	10.5
Undergraduate Degree	4	21.1
Master's Degree	2	10.5
Doctorate	2	10.5
Marital status*		
Single	2	10.5
Married	14	73.7
Separated	1	5.3
Widowed	2	10.5
Diabetes		
Type 1	10	50
Type 2	9	45
No diabetes	1	5
Diabetes diagnosis*		
< 12 months	4	21.1
1-5 years	8	42.1
5-10 years	1	5.3

10+ years	6	31.6
Research Experience		
Previous participation in research	6	30
Previous involvement in research	3	15
Diabetes support and education experience		
Previous attendance at diabetes support group	16	80
Previous attendance at diabetes education session	9	45
Diabetic Retinopathy Screening		
Familiar with term 'diabetic retinopathy screening'	19	95
Attended diabetic retinopathy screening*	18	94.7
Attended screening at hospital**	3	16.7
Attended screening at RetinaScreen provider**	11	61.1
Attended screening at local optician**	4	22.2
Other health conditions		
Heart Disease	7	35
Asthma	2	10
Arthritis	4	20
Any emotional or psychiatric problems (such as depression or anxiety)	2	10
Stomach ulcers	1	5
Attitude to medical appointments		
I always attend	20	100

*N=19
**N=18

Appendix 1.4: Experience Questionnaire

A 1.4.1. Additional Information on questionnaire development

Existing validated measures that were deemed unsuitable included questionnaires that measured group dynamics in workplace and organizational settings where participants worked together on an on-going basis (1-3). and a questionnaire that measured participants' experiences of being a research subject (as opposed to being actively involved in a participatory research process) (4). Therefore, we developed our own questionnaire based on sample items from a non-validated survey instrument published by Schulz et al. (5). These questionnaire items were deemed suitable for our research objective and context as they were developed to evaluate individual experiences of group dynamics and short-term measures of partnership effectiveness within community-based participatory research partnerships. Schulz et al. provided 90 sample questionnaire items arranged into 15 categories were developed based on a review of the group dynamics literature, previous instruments (where available) and extensive input from community stakeholders involved in three separate participatory research partnerships. The sample questionnaire items were designed to be used selectively based on their relevance to the topics being evaluated and not combined together as one single instrument. As there were some overlap between items and some items were not relevant to our study objective, we selected 11 items from seven categories. This helped to ensure the questionnaire was relatively short and straightforward to complete at the end of the two-hour consensus meetings (6). Categories included were: (1) comfort level for expressing opinions: communication, (2) level of influence and power

of self and others in the group, (3) perceived level of trust, (4) personal, organizational and community benefits of participation, (5) sense of ownership/ belonging to the group: cohesion, (6) group empowerment and (7) community empowerment. Categories excluded were: (1) Leadership and participation, (2) How well the group recognizes and addresses conflicts and problems, (3) Decision-making procedures, (4) Problem solving processes, (5) Meeting organisation, agenda setting, facilitation and staffing, (6) Accomplishments/impact of the group and (7) Member background and meeting attendance.

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A 1.4.2 Experience questionnaire

EXPERIENCE SURVEY

	Please indicate how much you agree with each statement:	Strongly Disagree	Disagree	Neither Agree Nor Disagree	Agree	Strongly Agree
1.	I felt comfortable expressing my opinion in the group.					
2.	I felt my opinions were listened to and considered by the other group members.					
3.	I felt part of the group (like I belonged to the group).					
4.	I felt pressured to go along with the decisions of the group even though I did not agree.					
5.	I felt a sense of trust and openness between group members.					
6.	I thought that certain individuals spoke more than others in the group.					
7.	I felt that I could influence the decisions that the group made.					
8.	I felt that certain individuals had more influence over the decision-making process than others.					
9.	I have increased my knowledge about important topics since participating in this group.					
10.	By working together, we can influence decisions that affect the research process.					
11.	By working together, we can influence decisions that affect people with diabetes.					

Do you have any other comments/suggestions?

Would you like to participate in a follow-up interview about your experience?

We would like to learn more about your experience of taking part in the meeting. If you are willing to be contacted about taking part in a short follow up interview (either in person or by telephone), please provide your contact details below and a member of the research team will be in touch. If you do not wish to be contacted, please leave this section blank.

Name _____

Phone Number _____

Email Address_____

Appendix 1.5: Observation Guide and Grid

How is the group working overall?
How is the group making decisions

Participation/ Non-participation	**Dominance/ submissiveness**	**Ingroups/Outgroups**
Body language and facial expressions	**Gaze**	**Effect of expert /Lay-knowledge**

Observations

Appendix 1.6: Interview Topic Guide

Interview Topic Guide

Questions	**Prompts**
Did you feel comfortable expressing your opinion in the group?	Why/why not? Did you feel part of the group (like you belonged to the group)?
Did you feel that you could influence the decisions that the group made?	Why/why not? Was your opinion listened to and considered by other group members?
Did everyone in the group have a voice in the decisions that were made?	Did certain individuals talk more at the meeting than others? Did you feel that certain individuals had more influence over the decision-making process than others?
Do you think that the group was able to deal with conflicts that came up during the meeting?	How did the group deal with conflicts?
Did you feel pressured to go along with decisions of the group even though you might not agree?	Did you feel a sense of trust and openness between group members?

Appendix 2- Supplementary Data for Chapter 4

Appendix 2.1 Information flyer

Appendix 2.2 Recruitment survey

Section B: Demographics

B1. Age

- 18-24 ☐
- 25-34 ☐
- 35-44 ☐
- 45-54 ☐
- 55-64 ☐
- 65-74 ☐
- 75-84 ☐
- 85+ ☐

B2. Gender

- Female ☐
- Male ☐

B3. How would you describe the area you live in?

- Urban ☐
- Rural ☐

B4. How would you describe your Nationality and Ethnicity?

- White Irish ☐
- Irish Traveller ☐
- Other White background ☐
- Black or Black Irish of African background ☐
- Any other Black background ☐
- Chinese or Chinese Irish ☐
- Any other Asian background ☐
- Other ☐

Other

B5. Is your healthcare covered by:

- Full Medical Card ☐
- GP Visit Card ☐

Private Health Insurance ☐

None of these ☐

Don't know ☐

B6. What is your highest level of education completed to date?

No formal education ☐

Primary level ☐

Junior Certificate/Intercert ☐

Apprenticeship ☐

Leaving Certificate ☐

Diploma ☐

Undergraduate Degree ☐

Masters Degree ☐

Doctorate ☐

Other ☐

Other

B7. What is your current marital status?

Single ☐

Cohabiting ☐

Married ☐

Separated ☐

Divorced ☐

Widowed ☐

Section C: Research Involvement

C1. Have you previously taken part in research as a research participant?

Yes ☐

No ☐

C2. **If yes, please give details**

C3. **Have you previously been involved in designing or carrying out research?**

- Yes ☐
- No ☐

C4. **If yes please give details:**

C5. **How did you first hear about this research opportunity?**

- Twitter ☐
- Facebook ☐
- Email ☐
- Flyer picked up (eg: waiting room, meeting, etc) ☐
- Flyer given by health care professional ☐
- Flyer given by researcher ☐
- Poster in waiting room ☐
- Poster / stand at confrence ☐
- Other ☐

Other

C6. **Why are you interested in being involved in this research?**

Please tell us the main reasons why you are interested in the box below:

Section D: Diabetes Diagnosis

D1. **Have you been diagnosed with diabetes?**

- ☐ Yes
- ☐ No

Section E: Diabetes Experience

We now have some questions about your experience of diabetes.

E1. **Please select the option that best describes you**

- ☐ I am a carer of someone with diabetes
- ☐ I am a family member of someone with diabetes
- ☐ I am a friend of someone with diabetes
- ☐ Other

Other

E2. **What type of diabetes do you have?**

- ☐ Type 1
- ☐ Type 2

E3. **When were you diagnosed with diabetes?**

- ☐ In the last 12 months
- ☐ 1-5 years ago
- ☐ 5-10 years ago
- ☐ 10+ years ago

E4. Have you ever attended a diabetes support group?

- Yes ☐
- No ☐
- Can't remember ☐

E5. Have you ever attended a diabetes education programme?

- Yes ☐
- No ☐
- Can't remember ☐

E6. Have you heard about diabetic retinopathy screening?

- Yes ☐
- No ☐
- Can't remember ☐

E7. Have you attended a retinopathy screening appointment?

Note: At the retinopathy screening appointment, eye drops are put into your eyes to enlarge your pupils for testing. If you remember this, it is likely that you have attended a retinopathy screening appointment.

- Yes ☐
- No ☐
- Unsure, but I recall getting eye drops put in my eyes at an appointment ☐
- Can't remember ☐

E8. Where was the appointment where you attended screening: click all that apply

- At the hospital ☐
- At a RetinaScreen provider ☐
- At a local optician ☐

E9. Has a doctor ever told you that you have any of the following conditions?

- Heart Disease (including high blood pressure or hypertension or angina or heart attack or congestive heart failure or stroke or ministroke or high cholesterol or heart murmur or an abnormal heart rhythm). ☐
- Chronic lung disease (such as chronic bronchitis or emphysema) ☐
- Asthma ☐
- Arthritis (including osteoarthritis or rheumatism) ☐
- Osteoporosis (sometimes called thin or brittle bones) ☐

Lymphoma (but excluding minor skin cancers) ☐

Parkinson's disease ☐

Any emotional, nervous or psychiatric problems (such as depression or anxiety) ☐

Alcohol or substance abuse ☐

Alzheimer's disease ☐

Dementia, organic brain syndrome, senility ☐

Serious memory impairment ☐

Stomach ulcers ☐

Varicose ulcers (an ulcer due to varicose veins) ☐

Cirrhosis, or serious liver damage ☐

None of the above ☐

E10. Please select the statement which best describes you:

I always attend medical appointments ☐

I try my best to attend medical appointments ☐

I find it hard to attend medical appointments ☐

Appendix 2.3 Summary of existing evidence

ideas

Improving Diabetes Eye-screening Attendance

IMPORTANT INFORMATION

Why are we doing this study?

Diabetes can cause people to lose their sight. This is because diabetes damages the small blood vessels at the back of the eye. This is called retinopathy. Screening to detect and treat early signs of this damage can prevent sight loss. In Ireland, there is a national screening programme, called RetinaScreen, which provides **free** screening. However, not everyone attends this screening.

Only 54% attend

In 2015, only 32% of people consented to be invited to screening. Of those who were invited, only 54% attended their screening appointment.

What we did

We looked at the international evidence

We looked at all the relevant and available evidence on interventions to improve attendance at diabetes eye screening.

We looked at reasons why people attend or do not attend diabetes eye screening

We also looked at all the relevant and available evidence on barriers to and enablers of diabetes eye screening attendance.

We interviewed 48 people with diabetes and 30 healthcare professionals

The healthcare professionals included diabetes nurse specialists, practice nurses and GPs. We asked the patients and the healthcare professionals why people did or did not attend eye screening. We also asked people about their experiences of screening.

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What we found

Interventions to improve screening attendance boost attendance by about 12%.

Interventions can target people with diabetes, professionals or the healthcare system or all of these at the same time.

Common and successful interventions targeting people with diabetes

Give people information.
Interventions give people information on how to attend a screening exam.

Remind people about appointments. Interventions often remind people to attend.

Set goals for people.
For example, treatment targets.

Give feedback on outcomes.
For example, results of their screening

Make people aware of consequences. Give people information about the consequences of diabetes, like how it could lead to eye damage and what that would mean for them.

Make people aware of consequences. Give people information about the consequences of diabetes, like how it could lead to eye damage and what that would mean for them.

Combine awareness and reminders.
Use education to increase awareness of retinopathy or reminders, or both.

Use a source the person trusts.
Give people spoken or written information from a believable or trustworthy source.

Common and successful interventions targeting professionals

Change the environment
For example, adding a new practice team role to contact people with diabetes about their appointments.

Feedback on outcomes
Give professionals feedback on the outcomes, such as, a report on the proportion of patients who attended screening.

Use a source professional trusts
Successful interventions give professionals spoken or written information from a believable or trustworthy source.

Reminders
Interventions which worked often included registration and reminder systems. Reminders worked better if sent to both professionals and people with diabetes.

Why do people attend or not attend screening?

This section is based on our research with Irish people with diabetes and professionals, and international studies.

Practicalities

People's decision to attend screening sometimes depended on:

- the distance to the screening location
- whether or not they were close by
- if they had a way to get there and back.

Forgetting

Reasons people had not attended also included just forgetting and having other health problems.

Eye drops

The eye drops used during the procedure, put some people off attending.

Fear stops some from attending

Being afraid of getting a bad result was suggested as a reason why people did not attend.

Symptoms

People's decision to attend eye screening was also influenced by symptoms. If they had nothing wrong with their eyes they felt they did not need to attend.

Free service

The fact that the new service was free was seen as a reason to attend. People suggested the free aspect of the programme should be emphasised more.

Competing demands

Other demands on their time also made it difficult for people to attend. These included:

- difficulty getting time off work
- an unpredictable work schedule
- having siblings or children to care for

Multiple checks confuse people

People were confused between the routine checks they were already getting for their eyes and the new screening service. People who had been checked elsewhere (sometimes very recently) felt they did not need to attend the new service.

Phoning up to consent

Irish healthcare professionals felt patients may have difficulty phoning up to consent to be invited because of problems with:

- readability of the appointment letter
- IT skills
- dexterity
- hearing.

Some people felt they did not need screening

Some people did not attend if they did not see the need for screening or felt they were not at risk of eye damage. People who felt their eyesight was good or who had a good result from a previous test or another healthcare professional, felt they did not need to attend screening.

Professionals can influence

Some people attended because a GP or nurse recommended they attend screening. People felt screening was something GPs or nurses should highlight. For some people, if a GP or nurse simply asked whether or not they had attended, this encouraged them to do so.

Friends and family also important

A recommendation or prompt from friends or family also encouraged people to attend screening.

Desire to protect eyesight prompted others to attend

Some people went because the consequences of not attending screening were clear to them. They knew other people who had complications, or they already had problems themselves (such as poor eyesight or eye damage).

People who went to screening saw a need for it and felt strongly that screening was important for early detection, or that it would reassure them that everything was ok.

Understanding retinopathy made people more likely to attend

People who knew about the link between diabetes and eye damage usually attended. There was a lack of awareness of this link among people who did not attend, or information did not to start to sink in until they started getting problems with their eyes.

What helps or hinders healthcare professionals to register their patients for screening?

System challenges

There were some things to do with the healthcare system which made it difficult for professionals to register patients:

- time and resources needed to register patients and to check they were on the register
- competing work demands
- not knowing what screening attendance was like in their practice or local area, whether it was good or bad.

Motivation

Some things motivated professionals to register and consent their patients to the screening programme:

- they knew some patients would be unable to do so themselves
- they saw it as part of their professional role
- they saw positive things about screening, like getting a letter back with their patient's results which helped with follow-up care, or the fact patients could get screened locally.

Now please fill out the yellow questionnaire.

If you would like copies of study references, please let us know fiona.riordan@ucc.ie

Appendix 2.4 Self-completion survey of intervention components

Yellow Questionnaire

Here is a list of ways to improve diabetes eye screening attendance. We want your opinion on whether these things are **acceptable** and **feasible**. When we meet in person we will talk about who is the best person to deliver some of these messages, and when and how they should be delivered (e.g. in person, using a leaflet, a letter or a text message).

For each statement, please circle one number in the **acceptable** category and one in the **feasible** category.

Each number represents the following:

1 = Completely disagree
2 = Disagree
3 = Neither disagree or agree
4 = Agree
5 = Completely agree

For example:

Statement	This is acceptable (you like it, and you think it makes sense)	This is feasible (you think it can be done)
Prompt practices to register patients	1 2 3 (4) 5	1 2 3 4 (5)

1. Ways to encourage the person to attend diabetes eye screening

Statement	This is acceptable (you like it, it makes sense)	This is feasible (you think it can be done)
Provide a personal story from someone else with diabetes who…		
• is a similar age and profile to them and explains how screening was a way for them to take charge of their health.	1 2 3 4 5	1 2 3 4 5
• has retinopathy and tells them about the benefits of screening (e.g. reassured all is ok, treatment stops things getting worse)	1 2 3 4 5	1 2 3 4 5
• has retinopathy and tells them it is important to go to screening before it is too late, there may be no symptoms and everyone with diabetes is at risk.	1 2 3 4 5	1 2 3 4 5
• wishes they went to screening sooner who prompts the person to think about the regret they will feel if they do not attend screening.	1 2 3 4 5	1 2 3 4 5
• explains there is no harm from drops used during screening and the overall benefits outweigh the short-term discomfort.	1 2 3 4 5	1 2 3 4 5
• provides an observable example that shows them how to consent or attend.	1 2 3 4 5	1 2 3 4 5
• delivers a message recognising the anxiety people might feel but emphasizes the positive consequences of attending.	1 2 3 4 5	1 2 3 4 5
• prompts the person to imagine the outcomes of attending vs. not attending	1 2 3 4 5	1 2 3 4 5

1

(knowing all is ok, treatment available vs. not knowing, they could have eye damage).										
Statement	**This is acceptable** (you like it, it makes senses)					**This is feasible** (you think it can be done)				
Someone in the practice could…										
• encourage the person to attend screening.	1	2	3	4	5	1	2	3	4	5
• tell the person that they approve of screening and hope the person will attend.	1	2	3	4	5	1	2	3	4	5
• persuade the person they will be able to attend screening (e.g. help them to think about times they successfully managed their diabetes or attended appointments)	1	2	3	4	5	1	2	3	4	5
• send or give a take-home reminder to the person to consent and attend their screening appointment.	1	2	3	4	5	1	2	3	4	5
• explain the difference between routine eye checks and the screening test, what both tests can and cannot tell them, and that routine checks are not a substitute.	1	2	3	4	5	1	2	3	4	5
• explains there is no harm from drops used during screening and the overall benefits outweigh the short-term discomfort.	1	2	3	4	5	1	2	3	4	5
• advise the person how to consent to screening and to ask for help if they are unable/unsure about how to do this.	1	2	3	4	5	1	2	3	4	5
• tell the person that after their appointment they will be reassured or they can get treated in time to stop things getting worse.	1	2	3	4	5	1	2	3	4	5
• explain how it's important to go to screening before it is too late, they personally are at risk and that screening applies to them.	1	2	3	4	5	1	2	3	4	5
• encourage the person to think of screening not as something extra, but as part of the whole package of self-management.	1	2	3	4	5	1	2	3	4	5
• help the person to make a plan about when and where they will consent and how they will attend when they get their appointment.	1	2	3	4	5	1	2	3	4	5
Other ideas to encourage the person with diabetes to consent or attend										
• arrange for support from family/friends (e.g. encouragement to consent/attend).	1	2	3	4	5	1	2	3	4	5
• advise/arrange for practical support from family/friends (e.g. identify transportation).	1	2	3	4	5	1	2	3	4	5
• provide a message from the screening service about why they want the person to attend (e.g. our priority is to preserve your vision) and a reminder the service is free.	1	2	3	4	5	1	2	3	4	5
• draw the person's attention to the number of people like them who have attended.	1	2	3	4	5	1	2	3	4	5
• the person ticks off a checklist when they have consented/attended.	1	2	3	4	5	1	2	3	4	5

2

2. Ways that encourage the practice staff to make sure person attends		
Statement	**This is acceptable** (you like it, and think it makes sense)	**This is feasible** (you think it can be done)
• provide practice with observable example/information on how to check and register people with diabetes.	1 2 3 4 5	1 2 3 4 5
• prompt practice to check the register during consultation and register person if necessary (e.g. electronic reminder)	1 2 3 4 5	1 2 3 4 5
• prompt practice to encourage the person to consent/attend & provide information on the benefits	1 2 3 4 5	1 2 3 4 5
• provide a new resource to the practice (e.g. researcher checks if person registered, consented and/or attended)	1 2 3 4 5	1 2 3 4 5
• provide checklist of ways to encourage consent/attendance	1 2 3 4 5	1 2 3 4 5
• establish a way for the practice to monitor and record their efforts to promote attendance	1 2 3 4 5	1 2 3 4 5
• identify someone in the practice to help the person to register and consent.	1 2 3 4 5	1 2 3 4 5
Tell practices about...		
• the benefits to the practice when their patients attend (e.g. receiving timely results, they have access to local service)	1 2 3 4 5	1 2 3 4 5
• consequences when their patients do not attend (e.g. eye damage, costs of missed appointments).	1 2 3 4 5	1 2 3 4 5
Use a personal story from a patient to tell practices...		
• the benefits and risks to patients of attending/not attending	1 2 3 4 5	1 2 3 4 5
• patients are more likely to attend screening if a health professional prompts or encourages them to do so	1 2 3 4 5	1 2 3 4 5
Give practices feedback on...		
• number of their patients who have not registered, consented or attended	1 2 3 4 5	1 2 3 4 5
• the differences between % attending from their practice and other practices	1 2 3 4 5	1 2 3 4 5
• national or international uptake or targets	1 2 3 4 5	1 2 3 4 5
• use a trusted source to deliver feedback and messages (e.g. colleague)	1 2 3 4 5	1 2 3 4 5

Please write comments or suggestions here about the ideas listed above:

3

Appendix 2.5 Results of self-completion survey of intervention components

Item		Meeting	Acceptable				Feasible			
			Disagree n (%)	Neutral n (%)	Agree n (%)	Total n (%)	Disagree n (%)	Neutral n (%)	Agree n (%)	Total n (%)
1	Provide a personal story from someone else with diabetes who is a similar age and profile to them and explains how screening was a way for them to take charge of their health	People with diabetes	0	1 (10%)	9 (90%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	1 (9.1%)	1 (9.1%)	9 (81.8%)	11 (100%)	1 (9.1%)	1 (9.1%)	9 (81.8%)	11 (100%)
		HCP	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)	1 (16.7%)	0	5 (83.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (6.9%)	4 (13.8%)	23 (79.3%)	29 (100%)	2 (7.1%)	5 (17.9%)	21 (75%)	28 (100%)
2	Provide a personal story from someone else with diabetes who has retinopathy and tells them about the benefits of screening (e.g. reassured all is ok, treatment stops things getting worse) *	People with diabetes	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	0	1 (9.1%)	10 (90.9%)	11 (100%)	1 (9.1%)	1 (9.1%)	9 (81.8%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	0	2 (6.9%)	27 (93.1%)	29 (100%)	1 (3.6%)	3 (10.7%)	24 (85.7%)	28 (100%)
3	Provide a personal story from someone else with diabetes who has retinopathy and tells them it is important to go to screening before it is too late, there may be no symptoms and everyone with diabetes is at risk*	People with diabetes	0	0	10 (100%)	10 (100%)	0	2 (22.2%)	7 (77.8%)	9 (100%) *
		Combined*	0	2 (18.2%)	9 (81.8%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	1 (16.7%)	0	5 (83.3%)	6 (100%)	2 (33.3%)	0	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (200%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	2 (6.9%)	26 (89.7%)	29 (100%)	3 (10.7%)	4 (14.3%)	21 (75%)	28 (100%

4	**Provide a personal story from someone else with diabetes who wishes they went to screening sooner who prompts the person to think about the regret they will feel if they do not attend screening***	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	2 (18.2%)	3 (27.3%)	6 (54.5%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	2 (33.3)	1 (16.7%)	3 (50%)	6 (100%)	3 (50%)	1 (16.7%)	2 (33.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%	2 (100%)
		Total	4 (13.8%)	6 (20.7%)	19 (65.5%)	29 (100%)	4 (14.3%)	6 (21.4%)	18 (64.3%)	28 (100%)
5	**Provide a personal story from someone else with diabetes who explains there is no harm from drops used during screening and the overall benefits outweigh the short-term discomfort**	**People with diabetes**	0	0	10 (100%)	10 (100%)	1 (11.1%)	1 (11.1%)	7 (77.8%)	9 (100%) *
		Combined	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)	1 (9.1%)	1 (9.1%)	9 (81.8%)	11 (100%)
		HCP	1 (16.7%)	0	5 (83.3%)	6 (100%)	1 (16.7%)	0	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (6.9%	2 (6.9%)	25 (86.2%)	29 (100%)	3 (10.7%)	3 (10.7%)	22 (78.6%)	28 (100%)
6	**Provide a personal story from someone else with diabetes who provides an observable example that shows them how to consent or attend***	**People with diabetes only**	0	2 (20%)	8 (80%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	1 (9.1%)	3 (27.3%)	7 (63.6%)	11 (100%)	1 (9.1%)	4 (36.4%)	6 (54.5%)	11 (100%)
		HCP	1 (16.7%)	0	5 (83.3%)	6 (100%)	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (6.9%)	5 (17.2%)	22 (75.9%)	29 (100%)	2 (7.1%)	9 (32.1%)	17 (60.7%)	28 (100%)
7	**Provide a personal story from someone else with diabetes who delivers a message recognising the anxiety people might feel but emphasizes the positive consequences of attending***	**People with diabetes**	0	0	10 (10%)	10 (10%	0	2 (22.2%)	7 (77.8%)	9 (100%) *
		Combined	0	3 (27.3%)	8 (72.7%)	11 (100%	1 (9.1%)	4 (36.4%)	6 (54.5%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	0	3 (10.3%)	26 (89.7%)	29 (100%)	1 (3.6%)	8 (28.6%)	19 (67.9%)	28 (100%)
8	**Provide a personal story from someone else with diabetes who prompts the**	**People with diabetes**	2 (10%)	8 (80%)	0	10 (100%)	0	4 (44.4%)	5 (55.6%)	9 (100%) *
		Combined	3 (27.3%)	8 (72.7%)	0	11 (100%)	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)

	person to imagine the outcomes of attending vs. not attending (knowing all is ok, treatment available vs. not knowing, they could have eye damage) *	**HCP**	3 (50%)	3 (50%)	0	6 (100%)	1 (16.7%)	2 (33.3%)	3 (50%)	6 (100%)
		Did not attend	0	2 (100%)	0	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	8 (27.6%)	21 (72.4%)	0	29 (100%)	3 (10.7%)	8 (28.6%)	17 (60.7%)	28 (100%)
9	**Someone in the practice could encourage the person to attend screening***	**People with diabetes**	0	0	10 (100%)	10 (100%)	0	1 (11.1%)	8 (88.9%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	0	28 (96.6%)	29 (100%)	2 (7.1%)	2 (7.1%)	24 (85.7%)	28 (100%)
10	**Someone in the practice could tell the person that they approve of screening and hope the person will attend***	**People with diabetes only**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	2 (18.2%)	0	9 (81.8%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	2 (6.9%)	0	27 (93.1%)	29 (100%)	1 (3.6%)	2 (7.1%)	25 (89.3%)	28 (100%)
11	**Someone in the practice could persuade the person they will be able to attend screening (e.g. help them to think about times they successfully managed their diabetes or attended appointments)**	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	3 (33.3%)	6 (100%)	9 (100%) *
		Combined	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)	2 (18.2%)	3 (27.3%)	6 (54.4%)	11 (100%)
		HCP	0	3 (50%	3 (50%)	6 (100%)	0	3 (50%)	3 (50%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	7 (24.1%)	21 (72.4%)	29 (100%)	2 (7.1%)	9 (32.1%)	17 (60.7%)	28 (100%)

12	**Someone in the practice could send or give a take-home reminder to the person to consent and attend their screening appointment***	**People with diabetes only**	0	1 (10%)	9 (90%)	10 (100%)	0	1 (11.1%)	8 (88.9%)	9 (100%) *
		Combined	0	1 (9.1%)	10 (90.9%)	11 (100%)	1 (9.1%)	0	10 (90.9%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	1 (16.7%)	0	5 (83.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	1 (50%)	1 (50%)	0	2 (100%)
		Total	0	3 (10.3%)	26 (89.7%)	29 (100%)	3 (10.7%)	2 (7.1%)	23 (82.1%)	28 (100%)
13	**Someone in the practice could explain the difference between routine eye checks and the screening test, what both tests can and cannot tell them, and that routine checks are not a substitute**	**People with diabetes**	0	1 (10%)	9 (90%)	10 (100%)	0	2 (22.2%)	7 (77.8%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	1 (3.4%)	27 (93.1%)	29 (100%)	1 (3.6%)	4 (14.3%)	23 (82.1%)	28 (100%)
14	**Someone in the practice could explain there is no harm from drops used during screening and the overall benefits outweigh the short-term discomfort***	**People with diabetes only**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)	2 (18.2%)	3 (27.3%)	6 (54.5%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	2 (6.9%)	26 (89.7%)	29 (100%)	2 (7.1%)	3 (10.7%)	23 (82.1%)	28 (100%)
15	**Someone in the practice could advise the person how to consent to screening and to ask for help if they are unable/unsure about how to do this**	**People with diabetes**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	2 (18.2%)	0	9 (81.8%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	0	28 (96.6%)	29 (100%)	3 (10.7%)	1 (3.6%)	24 (85.7%)	28 (100%)
16		**People with diabetes**	0	0	10 (100%)	10 (100%)	0	1 (11.1%)	8 (88.9%)	9 (100%) *

	Someone in the practice could tell the person that after their appointment they will be reassured, or they can get treated in time to stop things getting worse	**Combined**	0	0	11 (100%)	11 (100%)	1 (9.1%)	0	10 (90.9%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	2 (33.3%)	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total		1 (3.4%)	28 (96.6%)	29 (100%)	1 (3.6%)	3 (10.7%)	24 (85.7%)	28 (100%)
17	**Someone in the practice could explain how it's important to go to screening before it is too late, they personally are at risk and that screening applies to them***	**People with diabetes only**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	0	1 (9.1%)	10 (90.9%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	1 (16.7%)	0	5 (83.3%)	6 (100%)	1 (16.7%)	0	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	1 (3.4%)	27 (93.1%)	29 (100%)	2 (7.1%)	2 (7.1%)	24 (85.7%)	28 (100%)
18	**Someone in the practice could encourage the person to think of screening not as something extra, but as part of the whole package of self-management***	**People with diabetes**	0	0	9 (100%)	9 (100%) *	0	0	8 (100%)	8 (100%) *
		Combined	0	1 (9.1%)	10 (100%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	0	1 (3.6%)	27 (96.4%)	28 (100%)	1 (3.7%)	3 (11.1%)	23 (85.2%)	27 (100%)
19	**Someone in the practice could help the person to make a plan about when and where they will consent and how they will attend when they get their appointment**	**People with diabetes**	0	3 (30%)	7 (70%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	1 (9.1%)	3 (27.3%)	7 (63.6%)	11 (100%)	3 (27.3%)	5 (45.5%)	3 (27.3%)	11 (100%)
		HCP	0	2 (3.3%)	4 (66.7%)	6 (100%)	1 (16.7%)	0	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	8 (27.6%)	20 (69.0%)	29 (100%)	4 (14.3%)	8 (28.6%)	16 (57.1%)	28 (100%)
20	**Arrange for support from family/friends (e.g. encouragement to consent/attend)**	**People with diabetes only**	1 (10%)	1 (10%)	8 (80%)	10 (100%)	2 (22.2%)	2 (22.2%)	5 (55.6%)	9 (100%) *
		Combined	2 (18.2%)	2 (18.3%)	7 (63.6%)	11 (100%)	5 (45.5%)	3 (27.3%)	3 (27.3%)	11 (100%)

		HCP	0	2 (33.3%)	4 (66.7%)	6 (100%)	2 (33.3%)	0	4 (66.7%)	6 (100%)
		Did not attend	0	2 (100%)	0	2 (100%)	0	2 (100%)	0	2 (100%)
		Total	3 (10.3%)	7 (24.1%)	19 (65.5%)	29 (100%)	9 (32.1%)	7 (25%)	12 (42.9%)	28 (100%)
21	**Advise/arrange for practical support from family/friends (e.g. identify transportation) ***	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	2 (22.2%)	7 (77.8%)	9 (100%) *
		Combined	1 (9.1%)	4 (36.4%)	6 (54.5%)	11 (100%)	6 (54.5%)	2 (18.3%)	3 (27.3%)	11 (100%)
		HCP	2 (33.3%)	0	4 (66.7%)	6 (100%)	2 (33.3%)	2 (33.3%)	2 (33.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	2 (100%)	0	2 (100%)
		Total	3 (10.3%)	7 (24.1%)	19 (65.5%)	29 (100%)	8 (28.6%)	8 (28.6%)	12 (42.9%)	28 (100%)
22	**Provide a message from the screening service about why they want the person to attend (e.g. our priority is to preserve your vision) and a reminder the service is free-**	**People with diabetes only**	0	1 (10%)	9 (90%)	10 (100%)	0	1 (11.1%)	8 (88.9%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	1 (3.4%)	27 (93.1%)	29 (100%)	2 (7.1%)	2 (7.1%)	24 (85.7%)	28 (100%)
23	**Draw the person's attention to the number of people like them who have attended**	**People with diabetes**	1 (10%)	4 (10%)	5 (50%)	10 (100%)	2 (22.2%)	2 (22.2%)	5 (55.6%)	9 (100%) *
		Combined	2 (18.3%)	2 (18.2%)	7 (63.6%)	11 (100%)	2 (18.2%)	4 (36.4%)	5 (45.5%)	11 (100%)
		HCP	1 (16.7%)	2 (33.3%)	3 (50%)	6 (100%)	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	4 (13.8%)	9 (31%)	16 (55.2%)	29 (100%)	5 (17.9%)	8 (28.6%)	15 (53.6%)	28 (100%)
24	**The person ticks off a checklist when they have consented/attended***	**People with diabetes**	0	3 (30%)	7 (70%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	2 (18.2%)	2 (18.2%)	7 (63.6%)	11 (100%)	2 (18.2%)	2 (18.2%)	7 (63.6%)	11 (100%)
		HCP	1 (16.7%)	3 (50%)	3 (50%)	6 (100%)	1 (16.7%)	3 (50%)	2 (33.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)

		Total	3 (10.3%)	9 (31%)	17 (58.6%)	29 (100%)	3 (10.7%)	9 (32.1%)	16 (57.1%)	28 (100%)
25	**Provide practice with observable example/information on how to check and register people with diabetes**	**People with diabetes**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	0	0	11 (100%)	11 (100%)	2 (18.2%)	0	9 (81.8%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	0	0	29 (100%)	29 (100%)	3 (10.7%)	1 (3.6%)	24 (85.7%)	28 (100%)
26	**Prompt practice to check the register during consultation and register person if necessary (e.g. electronic reminder) ***	**People with diabetes**	0	0	10 (100%)	10 (100%	0	0	9 (100%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	2 (18.3%)	2 (18.3%)	7 (63.6%)	11 (100%)
		HCP	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)	2 (33.3%)	0	4 (66.7%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	2 (6.9%)	1 (3.4%)	26 (89.7%)	29 (100%)	4 (14.3%)	2 (7.1%)	22 (78.6%)	28 (100%)
27	**Prompt practice to encourage the person to consent/attend & provide information on the benefits**	**People with diabetes**	0	0	10 (100%)	10 (100%)	0	0	9 (100%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	1 (9.1%)	3 (27.3%)	7 (63.6%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	(100%)
		Total	1 (3.4%)	0	28 (96.6%)	29 (100%)	1 (3.6%)	3 (10.7%)	24 (85.7%)	28 (100%)
28	**Provide a new resource to the practice (e.g. researcher checks if person registered, consented and/or attended) ***	**People with diabetes only**	0	2 (22.2%)	7 (77.8%)	9 (100%) *	0	3 (30%)	7 (70%)	10 (100%)
		Combined	1 (10%)	2 (20%)	7 (70%)	10 (100%) *	4 (36.4%)	3 (27.3%)	4 (36.4%)	11 (100%)
		HCP	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (7.4%)	6 (22.2%)	19 (70.4%)	27 (100%)	5 (17.2%)	8 (27.6%)	16 (55.2%)	29 (100%)
29		**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *

	Provide checklist of ways to encourage consent/attendance	**Combined**	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)	2 (18.2%)	3 (27.3%)	6 (54.5%)	11 (100%)
		HCP	0	2 (33.3%)	4 (66.7%)	6 (100%)	1 (16.7%)	2 (33.3%)	3 (50%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%	2 (100%)
		Total	1 (3.4%	6 (20.7%)	22 (75.9%)	29 (100%)	3 (10.7%)	8 (28.6%)	17 (60.7%)	28 (100%)
30	**Establish a way for the practice to monitor and record their efforts to promote attendance**	**People with diabetes only**	1 (11.1%)	0	8 (88.9%)	9 (100%)*	0	2 (20%)	8 (80%)	10 (100%)
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	3 (27.3%)	3 (27.3%)	5 (45.5%)	11 (100%)
		HCP	1 (16.7%)	0	5 (83.3%)	6 (100%)	1 (16.7%)	2 (33.3%)	3 (50%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	3 (10.7%)	0	25 (89.3%)	28 (100%)	4 (13.8%)	7 (24.1%)	18 (62.1%)	29 (100%)
31	**Identify someone in the practice to help the person to register and consent***	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	2 (22.2%)	7 (77.8%)	9 (100%) *
		Combined	0	3 (27.3%)	8 (72.7%)	11 (100%)	2 (20%)	3 (30%)	5 (50%)	10 (100%) *
		HCP	1 (16.7%)	1 (16.7%)	4 (66.7%)	6 (100%)	1 (16.7%)	2 (33.3%)	3 (50%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	6 (20.7%)	22 (75.9%)	29 (100%)	3 (11.1%)	7 (25.9%)	17 (63%)	27 (100%)
32	**Tell practices about the benefits to the practice when their patients attend (e.g. receiving timely results, they have access to local service)**	**People with diabetes**	0	1 (11.1%)	8 (88.9%)	9 (100%) *	0	2 (20%)	8 (80%)	10 (100%)
		Combined	0	2 (18.2%)	9 (81.8%)	11 (100%)	0	3 (27.3%)	8 (72.7%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	0	5 (17.9%)	23 (82.1%)	28 (100%)	0	7 (24.1%)	22 (75.9%)	29 (100%)
33	**Tell practices about consequences when their patients do not attend (e.g. eye damage, costs of missed appointments)**	**People with diabetes only**	1 (10%)	2 (20%)	7 (70%)	10 (100%)	0	0	8 (100%)	8 (100%) *
		Combined	2 (18.2%)	2 (18.2%)	7 (63.6%)	11 (100%)	0	3 (27.3%)	8 (72.7%)	11 (100%)

		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	2 (33.3%)	4 (66.7%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	3 (10.3%)	6 (20.7%)	20 (69%)	29 (100%)	0	6 (22.2%)	21 (77.8%)	27 (100%)
34	**Use a personal story from a patient to tell practices the benefits and risks to patients of attending/not attending***	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	2 (25%)	6 (75%)	8 (100%) *
		Combined	3 (27.3%)	1 (9.1%)	7 (63.6%)	11 (100%)	1 (9.1%)	3 (27.3%)	7 (63.6%)	11 (100%)
		HCP	2 (33.3%)	1 (16.7%)	3 (50%)	6 (100%)	1 (16.7%)	3 (50%)	2 (33.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	5 (17.2%)	5 (17.2%)	19 (65.5%)	29 (100%)	2 (7.4%)	9 (33.3%)	16 (59.3%)	27 (100%)
35	**Use a personal story from a patient to tell practices patients are more likely to attend screening if a health professional prompts or encourages them to do so***	**People with diabetes**	0	1 (10%)	9 (90%)	10 (100%)	0	1 (12.5%)	7 (87.5%)	8 (100%) *
		Combined	2 (18.2%)	1 (91. %)	8 (72.7%)	11 (100%)	1 (9.1%)	2 (18.2%)	8 (72.7%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	2 (6.9%)	3 (10.3%)	24 (82.8%)	29 (100%)	1 (3.7%)	4 (14.8%)	22 (81.5%)	27 (100%)
36	**Give practices feedback on number of their patients who have not registered, consented or attended**	**People with diabetes**	0	0	10 (100%)	10 (100%)	0	3 (33.3%)	6 (66.7%)	9 (100%) *
		Combined	1 (9.1%)	0	10 (90.9%)	11 (100%)	0	2 (18.2%)	9 (81.8%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	0	2 (100%)	2 (100%)
		Total	1 (3.4%)	0	28 (96.6%)	29 (100%)	0	5 (17.9%)	23 (82.1%)	28 (100%)
37	**Give practices feedback on the differences between % attending from their practice and other practices**	**People with diabetes**	0	3 (30%)	7 (70%)	10 (100%)	0	3 (37.5%)	5 (62.5%)	8 (100%) *
		Combined	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)	0	4 (36.4%)	7 (63.6%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (6.9%)	6 (20.7%)	21 (72.4%)	29 (100%)	0	9 (33.3%)	18 (66.7%)	27 (100%)

38	**Give practices feedback on national or international uptake or targets**	**People with diabetes**	0	2 (20%)	8 (80%)	10 (100%)	0	0	8 (100%)	8 (100%) *
		Combined	2 (18.2%)	0	9 (81.8%)	11 (100%)	0	2 (18.2%)	9 (81.8%)	11 (100%)
		HCP	0	0	6 (100%)	6 (100%)	0	0	6 (100%)	6 (100%)
		Did not attend	0	1 (50%)	1 (50%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (6.9%)	3 (10.3%)	24 (82.8%)	29 (100%)	0	3 (11.1%)	24 (88.9%)	27 (100%)
39	**Use a trusted source to deliver feedback and messages (e.g. colleague)**	**People with diabetes**	0	0	9 (100%)	9 (100%) *	0	2 (20%)	8 (80%)	10 (100%)
		Combined	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)	2 (18.2%)	1 (9.1%)	8 (72.7%)	11 (100%)
		HCP	0	1 (16.7%)	5 (83.3%)	6 (100%)	0	1 (16.7%)	5 (83.3%)	6 (100%)
		Did not attend	0	0	2 (100%)	2 (100%)	0	1 (50%)	1 (50%)	2 (100%)
		Total	2 (7.1%)	2 (7.1%)	24 (85.7%)	28 (100%)	2 (6.9%)	5 (17.2%)	22 (75.9%)	29 (100%)

*Missing data

Appendix 2.6 Changes made to survey of intervention components following PPI feedback

Based on PPI feedback, the following changes were made to the invitation letter:

- Included a sentence at the start to make it sound less of a 'chore'- exciting opportunity to be involved.
- Made the meeting date/time and location clearer.
- Removed any non-essential information and inserted two headings for the two things we were asking them to do before the meeting.
- Changed some of the wording e.g. ~~Evidence Summary~~ Information Summary

Before

Dear

You are invited to take part in a consensus meeting to help us find an acceptable way to improve diabetes eye screening attendance in Ireland. The details of the meeting are as following:

Day: Monday, 15th of October
Time: 6.00- 8.30pm
Location: Room G09, Western Gateway Building, Western Road, Cork.

During the consensus meeting we will discuss and decide on an intervention which we think could improve screening attendance. There will be approximately 15 people at the meeting in total. This will include 10 patients and members of the public, 3 researchers and 2 group facilitators. Further information on the location of the western gateway building (including parking facilities) can be found on the other side of this page.

Before you attend the meeting, it is very important that you read the **evidence summary** included in this pack. This evidence summary will give you important information. Please read this information carefully, as it will be useful for you when taking part in the meeting.

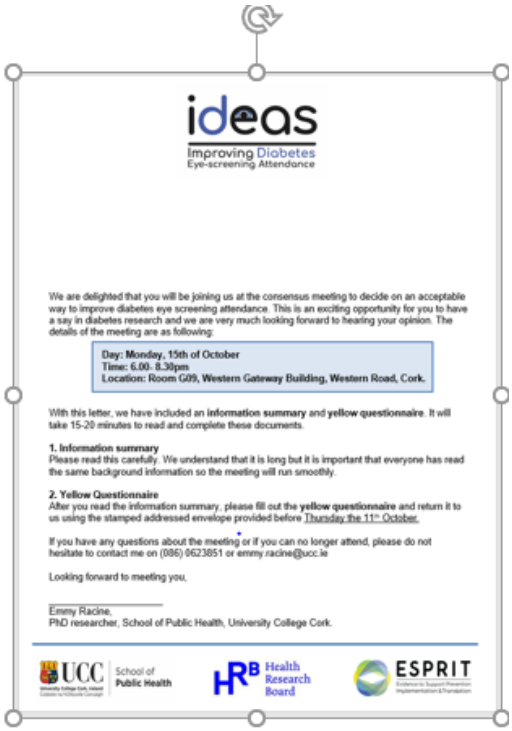
After you read the evidence summary, please fill out the **purple feedback sheet** and return it to us using the stamped addressed envelope provided. We will use the answers you provide on the feedback sheet, together with answers from other people attending the meeting to kick-start our discussion on the day.

If you have any questions about the meeting or if you can no longer attend, please do not hesitate to contact me on (086) 0623851 or emmy.racine@ucc.ie

Best wishes,

Emmy Racine,
PhD researcher, School of Public Health, University College Cork.

After



We are delighted that you will be joining us at the consensus meeting to decide on an acceptable way to improve diabetes eye screening attendance. This is an exciting opportunity for you to have a say in diabetes research and we are very much looking forward to hearing your opinion. The details of the meeting are as following:

Day: Monday, 15th of October
Time: 6.00- 8.30pm
Location: Room G09, Western Gateway Building, Western Road, Cork.

With this letter, we have included an **information summary** and **yellow questionnaire**. It will take 15-20 minutes to read and complete these documents.

1. Information summary
Please read this carefully. We understand that it is long but it is important that everyone has read the same background information so the meeting will run smoothly.

2. Yellow Questionnaire
After you read the information summary, please fill out the **yellow questionnaire** and return it to us using the stamped addressed envelope provided before Thursday the 11th October.

If you have any questions about the meeting or if you can no longer attend, please do not hesitate to contact me on (086) 0623851 or emmy.racine@ucc.ie

Looking forward to meeting you,

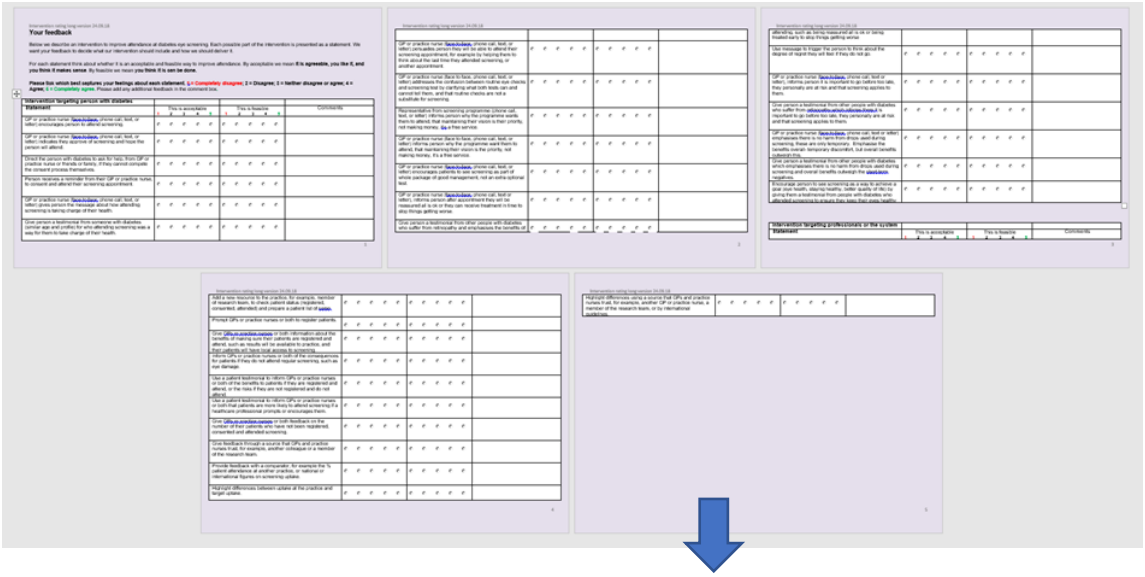
Emmy Racine,
PhD researcher, School of Public Health, University College Cork.

Appendix 2.7 Changes made to invitation letter following PPI feedback

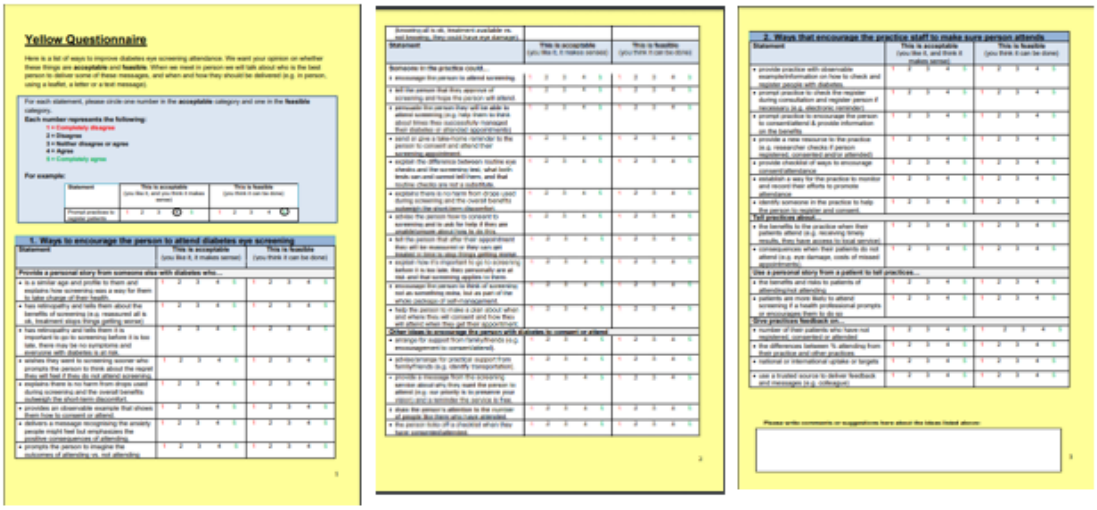
Based on PPI feedback, the following changes were made to the survey of intervention components:

- Changed to 'yellow questionnaire'.
- Reduced from 5 pages to 3 pages.
- Made the instructions clearer and included an example to illustrate how it should be filled out.
- Inserted headings for each section to avoid repetition and make it easier to read.
- Inserted rating headings (e.g. acceptable and feasible) at the top of each page.
- Included definitions of the rating headings to make it more understandable.
- Combined some of the items to make it shorter and easier to complete.
- Changed some of the wording e.g. ~~testimonial~~ = personal story

Before



After



Appendix 2.8 Facilitator Guide

The aim of these discussions is to find out the best way to make the acceptable ideas work in practice – **'what would this look like in practice', thinking about the mode, who should receive or deliver it, and when it should happen.**

TURN ON RECORDER (if not already one)
ASK PEOPLE TO MOVE THEIR PHONES AWAY FROM RECORDER (static on recording)

Group lead: _____

How was that decided (i.e. volunteer, nominated, more than one wanted to be lead):

_____

Small group discussion 1

Focus: other ideas to encourage person to consent or attend screening

If they focus on one particular way of doing things

- Why do you think that?
- If that were not possible what else could be done?
- What could have the most impact?

Specific prompts [put a tick beside the prompts you use] [√]

- Using a checklist – how would that work?
- How to draw people's attention to people who have attended (face to face, letter, leaflet, phone call, text, poster) **[ ]**
- Should screening programme should provide message or not **[ ]**
- How support would be arranged 'what would this look like' – ' how would this work?' **[ ]**

	What did they decide?	**Did everyone agree? Any difference within group?**
Checklist		
Drawing attention to numbers		
Providing messages		
Providing support		

Small group discussion 2

Focus: messages to be given to patients either by personal story or by practice

If they focus on one particular way of doing things

- Why do you think that?
- If that were not possible what else could be done?
- What could have the most impact?

Specific prompts [put a tick beside the prompts you use] [√]

- Would this message be best as a personal story or should it come from the practice? [ ]
- Personal stories
    - who should this person be (i.e. someone who attended or not, someone with eye damage or not, someone the same age?) [ ]
    - when should it be delivered? [ ]
    - how (face to face, letter, leaflet, video, text, phone call)? [ ]
- For messages to be delivered by the practice
    - who should deliver this (GP, practice nurse or someone else?) [ ]
    - when should it be delivered
    - how (face to face, leaflet, letter, text, phone call) [ ]

	What did they decide?	**Did everyone agree? Any difference within group?**
Who delivers message		
When		
How		

Small group discussion 3

Focus: encouraging practices staff to make sure patient attends

If they focus on one particular way of doing things

- Why do you think that?
- If that were not possible what else could be done?
- What could have the most impact?

Specific prompts [put a tick beside the prompts you use] [√]

- What is the best way to prompt or remind practices (e.g. electronic reminder, checklist)? **[ ]**
- How should practices monitor and record efforts? **[ ]**
    - Who should be responsible for this? **[ ]**
- If we provide a resource to practices, how would this work? **[ ]**
- Who should give messages to practice (researchers or using personal story) **[ ]**
    - How (face to face, letter, leaflet, phone call) **[ ]**
- How should feedback be given to practices:
    - who should it be given to (GP, practice nurse) **[ ]**
    - how (face to face, letter, email, phone call) **[ ]**
    - by who (colleague) **[ ]**
    - how often? **[ ]**
    - What comparator is best to use **[ ]**

	What did they decide?	**Did everyone agree? Any difference within group?**
Prompting & reminding practices		
Providing resource		
Providing messages (who, how)		
Feedback (who receives, who delivers, how, how often)		

Appendix 2.9 Recommendations that were within scope but not included in the intervention

Recommendation	Meeting 1	Meeting 2	Meeting 3	Reason for exclusion (based on APEASE criteria)
Practice Level Recommendations				
Have a chart at practice with the % numbers they want to achieve	✓	-	-	This was only put forward by people with diabetes in meeting 1, as it is a practice level recommendation and it was not mentioned by HCPs in meeting 2 or 3, it was not incorporated (acceptability).
Inform practices that they can market themselves as a practice known for good diabetes care	✓	-	-	Same as above.
Patient Level Recommendations				
Who should deliver the message?				
Diabetes Nurse Specialist (DNS)	✓	✓	-	Not all GP practices have DNS. DNS are not employed by HSE (national health service), therefore not possible for practice staff to ask them to deliver message (practicality, equity).
How should the message be delivered?				
SMS	✓	✓/×	-	Wrong individual could access and read text messages. Not all patients may own or use mobile phones. Not all practices use this mode and will have established acceptable consent processes (practicality, acceptability, safety, equity).
Email	✓	✓/×	-	Not all patients use email and have established acceptable consent processes. Participants in meeting 2 and 3 agreed phone call would work better (practicality, equity, safety).
Poster/ TV ad in GP waiting room	-	✓/x	✓/×	Not every practice allows posters/ has a tv. Participants in meeting 2 and 3 felt that it would only reach people that are attending anyway (acceptability, equity).
When should the message be delivered?				
Before patient collects next prescription	-	-	✓	It would not be acceptable to ask practices to do this. Screening attendance is voluntary, not acceptable to coerce people to attend (acceptability).
What should the message contain?				
Personal stories or testimonials -would need to be tailored to be effective	✓	✓	✓	It would be too difficult to tailor personal testimonials to suit all age groups, gender etc. and target them at individual patients (practicality).
Personal story from celebrity	-	-	✓	Put forward by participants in meeting 3 only and not mentioned in meeting 1 and 2. It would not be possible to identify a suitable celebrity and the resources were

				not available to cover the costs (acceptability, practicality, affordability).
GP should recommend that the patient talks to another patient at the practice	✓	-	-	Put forward by participants in meeting 1 only and not mentioned in meeting 2 and 3. It would be difficult to identify and gain consent from another patient at practice. It would not be possible to identify suitable patients for different age groups, gender etc. Also issues re: consent and patient confidentiality (acceptability, practicality, safety).
Distinguish the difference between HBA1c and retinal screening-	✓	✓	-	The research team felt that this recommendation was already captured in the intervention by outlining the asymptomatic nature of retinopathy and outlining the difference between regular eye-checks and retinopathy screening.
Provide a link to further information	-	-	✓	Only put forward by participants in meeting 3 and not mentioned in meeting 1 and 2. This was deemed not feasible as SMS and email were not feasible (see previous).
Ask patients to attend as a favour to the practice to get their numbers up	-	-	✓	It would not be appropriate to ask practice staff to do this and not acceptable to ask all patients to do this. (acceptability).

Appendix 3- Supplementary Data for Chapter 5

Appendix 3.1: Search strategy for each database

MEDLINE

	Search Domain	Search terms	
1	Clinical trials	exp Clinical Trial as Topic/ OR Trial*	926,679
2	PPI	(Consumer* or citizen* or client* or carer* or communit* or lay or patient* or public or service user* or user* or survivor* or stakeholder* or famil* or relative* or parent*) N3 (involv* or collaborat* or engag* or partner* or consult* or advis* or emancipat* or empower* or advocat* or embed* or represent* or test* or driven)	418,794
3	PPI	exp Participatory research/ or exp Patient Participation/ or community-based or participatory research or consumer participation or patient participation or user-tested or participant-developed or consumer-centred or patient-centered	90,982
4	PPI	2 OR 3	492,456
5	Retention	exp Patient Dropout/ or exp follow up/ studies or retention or attrition or follow-up or followup or withdr* or adher* or dropout* or drop-out*	1,350,269
6	PPI outcomes	Impact* or effect* or adapt* or modif* or chang* or develop* or design* or improv* or worse* or increas* or boost* or decreas* or reduc* or differ* or edit* or suggest* or max* or min*	17, 513, 948
7		1 and 4 and 5 and 6	9,207
8		7 (English language only, abstract available only, human only, (exclude animal studies)	7,405

EMBASE

	Search Domain	Search terms	
1	Clinical trials	exp Clinical Trial as Topic/ OR Trial*	2,183,933

2	PPI	(Consumer* or citizen* or client* or carer* or communit* or lay or patient* or public or service user* or user* or survivor* or stakeholder* or famil* or relative* or parent*) N3 (involv* or collaborat* or engag* or partner* or consult* or advis* or emancipat* or empower* or advocat* or embed* or represent* or test* or driven)	2,143
3	PPI	exp Participatory research/ or exp Patient Participation/ or community-based or participatory research or consumer participation or patient participation or user-tested or participant-developed or consumer-centred or patient-centered	50,800
4	PPI	2 OR 3	52,946
5	Retention	exp Patient Dropout/ or exp follow up/ or retention or attrition or follow-up or followup or withdr* or adher* or dropout* or drop-out*	2,394,503
6	PPI outcomes	Impact* or effect* or adapt* or modif* or chang* or develop* or design* or improv* or worse* or increas* or boost* or decreas* or reduc* or differ* or edit* or suggest* or max* or min*	21,830,244
7		1 and 4 and 5 and 6	2,168
8		7 (EMBASE only, English language only, abstract available only, human studies only)	1,294

Cochrane Library

	Search Domain	**Search terms**	
1	Clinical trials	exp Clinical Trial as Topic/ OR Trial*	784,046
2	PPI	(Consumer* or citizen* or client* or carer* or communit* or lay or patient* or public or service user* or user* or survivor* or stakeholder* or famil* or relative* or parent*) N3 (involv* or collaborat* or engag* or partner* or consult* or advis* or emancipat* or empower* or advocat* or embed* or represent* or test* or driven)	172
3	PPI	exp Participatory research/ or exp Patient Participation/ or community-based or participatory research or consumer participation or patient participation or user-tested or participant-developed or consumer-centred or patient-centered	20,491
4	PPI	2 OR 3	20,491
5	Retention	exp Patient Dropout/ or exp follow up/ or retention or attrition or follow-up or followup or withdr* or adher* or dropout* or drop-out*	285,328
6	PPI outcomes	Impact* or effect* or adapt* or modif* or chang* or develop* or design* or improv* or worse* or increas* or boost* or decreas* or reduc* or differ* or edit* or suggest* or max* or min*	1,251,794
7		1 and 4 and 5 and 6	5,934

8		7 (cochrane systematic reviews and trials only)	5,932

Psych Info

	Search Domain	Search terms	
1	Clinical trials	exp Clinical Trial as Topic/ OR Trial*	174,373
2	PPI	(Consumer* or citizen* or client* or carer* or communit* or lay or patient* or public or service user* or user* or survivor* or stakeholder* or famil* or relative* or parent*) N3 (involv* or collaborat* or engag* or partner* or consult* or advis* or emancipat* or empower* or advocat* or embed* or represent* or test* or driven)	147,308
3	PPI	exp Participatory research/ or exp Patient Participation/ or community-based or participatory research or consumer participation or patient participation or user-tested or participant-developed or consumer-centred or patient-centered	41,474
4	PPI	2 OR 3	180,281
5	Retention	exp Patient Dropout/ or exp follow up/ or retention or attrition or follow-up or followup or withdr* or adher* or dropout* or drop-out*	255,162
6	PPI outcomes	Impact* or effect* or adapt* or modif* or chang* or develop* or design* or improv* or worse* or increas* or boost* or decreas* or reduc* or differ* or edit* or suggest* or max* or min*	3,685,244
7		1 and 4 and 5 and 6	2,051
8		7 (English language only, human studies only (exclude animal))	1,906

CINAHL

	Search Domain	Search terms	
1	Clinical trials	exp Clinical Trial as Topic/ OR Trial*	414 846
2	PPI	(Consumer* or citizen* or client* or carer* or communit* or lay or patient* or public or service user* or user* or survivor* or stakeholder* or famil* or relative* or parent*) N3 (involv* or collaborat* or engag* or partner* or consult* or advis* or emancipat* or empower* or advocat* or embed* or represent* or test* or driven)	128 940

3	PPI	exp Participatory research/ or exp Patient Participation/ or community-based or participatory research or consumer participation or patient participation or user-tested or participant-developed or consumer-centred or patient-centered	51 690
4	PPI	2 OR 3	378 665
5	Retention	exp Patient Dropout/ or ~~exp follow-up/~~ research subject retention or retention or attrition or follow-up or followup or withdr* or adher* or dropout* or drop-out*	293 766
6	PPI outcomes	Impact* or effect* or adapt* or modif* or chang* or develop* or design* or improv* or worse* or increas* or boost* or decreas* or reduc* or differ* or edit* or suggest* or max* or min*	2 818 954
7		1 and 4 and 5 and 6	4482
8		7 (CINAHL only (exclude MEDLINE, English language only, abstract available only, human only, (exclude animal studies)	637

Appendix 3.2: Data extraction template

		(1) The RCT				(2) PPI in the trial					
Author	Year	Trial name	Trial subject area	Country	Trial target population	Phrasing in abstract	PPI contributors	Similar to trial target population	Trial Stage	Details of PPI	Training for PPI contributors

(3) Trial retention					(4) Reported Link between PPI and trial retention
Intervention Retention Rate	Control Retention Rate	Overall Retention Rate	Comments on retention	Measured/ Defined as	Link between PPI and retention

Appendix 3.3: Enhancing transparency in reporting the synthesis of qualitative research: the ENTREQ statement

No	Item	Guide and description	Reported on page no.
1	Aim	State the research question the synthesis addresses.	4
2	Synthesis methodology	Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology *(e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis).*	6&7
3	Approach to searching	Indicate whether the search was pre-planned (*comprehensive search strategies to seek all available studies)* or iterative (*to seek all available concepts until they theoretical saturation is achieved)*.	5
4	Inclusion criteria	Specify the inclusion/exclusion criteria *(e.g. in terms of population, language, year limits, type of publication, study type).*	4&5
5	Data sources	Describe the information sources used (e.g. *electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists)* and when the searches conducted; provide the rationale for using the data sources.	5
6	Electronic Search strategy	Describe the literature search *(e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits)*.	5 & Appendix 5.1
7	Study screening methods	Describe the process of study screening and sifting *(e.g. title, abstract and full text review, number of independent reviewers who screened studies).*	6
8	Study characteristics	Present the characteristics of the included studies *(e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions).*	9

No	Item	Guide and description	Reported on page no.
9	Study selection results	Identify the number of studies screened and provide reasons for study exclusion *(e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development).*	8
10	Rationale for appraisal	Describe the rationale and approach used to appraise the included studies or selected findings *(e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings).*	7
11	Appraisal items	State the tools, frameworks and criteria used to appraise the studies or selected findings *(e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope* [25]*; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting).*	7
12	Appraisal process	Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required.	7
13	Appraisal results	Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale.	9
14	Data extraction	Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? *(e.g. all text under the headings "results /conclusions" were extracted electronically and entered into a computer software).*	6&7
15	Software	State the computer software used, if any.	n/a
16	Number of reviewers	Identify who was involved in coding and analysis.	6&7
17	Coding	Describe the process for coding of data *(e.g. line by line coding to search for concepts).*	6&7
18	Study comparison	Describe how were comparisons made within and across studies *(e.g. subsequent*	7

No	Item	Guide and description	Reported on page no.
		studies were coded into pre-existing concepts, and new concepts were created when deemed necessary).	
19	Derivation of themes	Explain whether the process of deriving the themes or constructs was inductive or deductive.	6
20	Quotations	Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author's interpretation.	na
21	Synthesis output	Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. *new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct).*	14-23

Appendix 3.4: GRIPP 2 (Short Form) checklist scores

Study	Aim: Report the aim of PPI in the study	Methods: Provide a clear description of the methods used for PPI in the study	Study results: Outcomes- Report the results of PPI in the study, including both positive and negative outcomes	Discussion and conclusions: Outcome- Comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	Reflections/critical perspective: Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	Total
Adams et al. 2015 (a)	✓	✓	✓	✓	✓	5
Adams et al. 2015 (b)	✓	✓	✓	✓	✓	5
Angell et al. 2003	✓	✓	✓	✓	✓	5
Arean et al. 2003 (a)	✓	✓	✓	✓	-	4
Arean et al. 2003 (b)	✓	✓	✓	✓	-	4
Arjadi et al. 2018	✓	✓	✓	-	-	3
Ashton et al. 2017	✓	✓	-	✓	-	3
August et al. 2006	✓	✓	-	✓	✓	4
Burlew et al. 2011	✓	✓	✓	-	-	3
Buscemi et al. 2015	✓	✓	✓	✓	-	4
Chacko and Scavenius. 2018	✓	✓	-	✓	-	3
Chang et al. 2010	✓	✓	✓	-	-	3
Chhatre et al. 2018	✓	✓	✓	-	-	3
Chung et al. 2017	✓	✓	✓	✓	✓	5
Corbie-Smith et al. 2003	✓	✓	✓	-	-	3
De Marco et al. 2012	✓	✓	✓	✓	-	4
Edwards et al. 2011	✓	✓	✓	✓	✓	5

Elder et al. 2006	✓	✓	✓	-	-	3
Estreet et al. 2017	✓	✓	✓	✓	-	4
Fischer et al. 2017	✓	✓	✓	-	-	3
Foster et al. 2015	✓	✓	-	✓	-	3
Fouad et al. 2014	✓	✓	✓	✓	✓	5
Gappoo et al. 2009	✓	✓	-	✓	-	3
Garcia et al 2016 (a)	✓	✓	✓	✓	✓	5
Garcia et al 2016 (b)	✓	✓	✓	✓	✓	5
Garcia et al 2016 (c)	✓	✓	✓	✓	✓	5
Garcia et al 2016 (d)	✓	✓	✓	✓	✓	5
Harris et al. 2001	✓	✓	✓	✓	-	4
Jeffries et al. 2005	✓	✓	-	✓	-	3
Johnson et al. 2015	✓	✓	✓	✓	-	4
Ka'opua et al. 2011	✓	✓	✓	✓	-	4
Keown et al. 2018	✓	✓	-	✓	-	3
Kogan et al. 2016	✓	✓	✓	✓	✓	5
Koniak-Griffin et al. 2015	✓	✓	✓	✓	✓	4
Lloyd et al. 2017	✓	✓	✓	✓	✓	5
Loughery et al. 2017	✓	✓	✓	✓	✓	5
McGillicuddy et al. 2013	✓	✓	✓	-	-	3
Merriam et al. 2009	✓	✓	✓	✓	✓	5
Okely et al. 2011	✓	✓	✓	✓	-	4
Rhodes et al. 2011	✓	✓	✓	-	-	3

Stineman et al. 2011	✓	✓	✓	✓	-	4
Swartz et al. 2004	✓	✓	-	✓	-	3
Tanjasiri et al. 2015	✓	✓	✓	-	-	3
Vincent et al. 2013	✓	✓	✓	✓	-	4
Williams et al. 2007	✓	✓	✓	-	-	3

Appendix 4- Supplementary Data for Chapter 6

Appendix 4.1: Focus Group Topic Guide

Topic Guide

The objective of this study is to use a public and patient (PPI) strategy to develop a preferred method of receiving end-of-trial information for participants who were enrolled in the TRUST study. The aim is then to compare the information developed through the standard end-of-trail documents developed by the co-ordinating study site in Glasgow.

Thank participants, introduce researcher & study.

Briefly go through information sheet and consent form.

Outline general housekeeping rules which will be said to participants at the beginning of the focus groups.

-If it is ok with you, I will audio record this group discussion so I can give you my full attention. However, the research assistant will be taking field notes during the discussion just to ensure that we don't miss anything.

-Everything we discuss will be confidential and your identity will remain anonymous. We may use direct quotes from this discussion but your identity and position will be kept completely anonymous and your name will not be used on any reports or publications.

-You can choose to withdraw from the discussion at any time.

-Do you have any questions before we get started?

Sign consent and give copy.

Questions	Prompt/probe
Can you each give your first name only please?	-Distribute name tags
Trial Experience How did you first become aware of the TRUST study?	Who told you about it? GP? Other healthcare provider? Advertising campaign? Other?
When you first heard about the trial what did you think?	Were you interested in the trial straight away? Positive/Negative first impression? Did you think it would be useful for you?
How was your experience of the TRUST Thyroid trial?	Tell me about your initial contact with your GP? What was it like when you were first recruited? How did you find the study visits? (research team, doctors etc.) Were they helpful? How do you feel now that the trial has ended? What was your most positive and negative experience of the TRUST study? How do you think this could be improved upon?
In your opinion do you think the information you received at study visits was informative?	Too much information? Too little? What other information would you have liked? Did you seek information from other sources-GP, internet, other? If so what kind of information?
During the trial, did you think you were on the placebo or the active drug?	If you had a choice at the beginning of the trial, which one would you have picked? Why?
Most participants have requested to be un-blinded. Why do you think this is?	Do you think this is important to know? How would you feel if no one ever told you?
Result Dissemination Do you want to find out the final study results? Why?	Do you think this is important? Why?

When the final results of the trial are known, how would you like to find out?	By post/telephone contact/email/face to face meeting?
In your opinion do you think study participants should be involved in formulating information leaflets for research studies?	Yes- in what capacity? No- why not?
Would you be interested in helping to write the information leaflet that we will send to all participants about the final study results?	Is there anything in particular you would like to be included in this leaflet?
Would you participate in another research study?	Yes, why? No, why? Would you have any interest in being part of advisory groups for trials in the future?

Appendix 4.2: Draft one of patient-based result letter

Dear Participant,

Thank you for taking part in the TRUST Thyroid Trial. You may be interested to read the results of the trial which are listed below in question and answer format.

1. **What is subclinical hypothyroidism (SCH)?**

Subclinical hypothyroidism also called mildly underactive thyroid. It affects around one in six people over the age of 65 and has been linked to various health problems, such as heart attacks and strokes, in later life. At this point in time doctors are not sure how to treat these patients because extensive research has never been done.

2. **How is SCH diagnosed?**

A person is said to have SCH if two of their blood tests, taken within a 3month period, show that their TSH level is persistently high (≥4.6 to ≤ 19.9Mu/L) and their free thyroxin (fT4) remains in normal range.

3. **What was the aim of the TRUST Thyroid Trial?**

To test if older community dwelling adults aged ≥65 years with subclinical hypothyroidism (SCH) benefit from Levothyroxine treatment. The main benefit the trial was looking at was an improvement in participant's Thyroid Specific Quality of Life. This was measured using the ThyPRO questionnaire.

4. **What were the secondary benefits examined the in trial?**

During your trial visits, you completed a number of questionnaires, physical tests and weight measurements. The purpose of these tests was to determine if Levothyroxine can prevent cardiovascular disease, improve health-related quality of life, muscle function and cognition in older adults with SCH.

5. **Who took part in the TRUST Thyroid Trial?**

In total 738 participants with SCH from Ireland, the United Kingdom, Netherlands and Switzerland took part in the study.

6. **Why was I chosen to take part in the trial?**

You were asked to take part in the trial as during a routine review of your blood result, your GP found you had an abnormal TSH result and so you would be suitable for the TRUST Thyroid Trial.

7. **How many participants were on the active drug-levothyroxine and how many participants were on the placebo?**

From the total 738 participants recruited to the trial X were on the active drug and X were on the placebo.

8. **How long was the TRUST Thyroid Trial?**

The trial ran from May 2013 to November 2016. Participants had to be in the trial for a minimum of 12months and a maximum of 36 months.

9. Why did the trial stop?

The TRUST trial stopped as it has reached its scheduled completion date.

10. What was the primary result of the TRUST Thyroid Trial?

The trial showed that participants on the active drug had (better or worse) Thyroid specific Quality of Life scores compared to the placebo group

11. What were the secondary results of the trial?

In total the trial included 8 secondary outcomes. The results of these tests are listed in the table below:

Outcome	Questionnaire	Results
	General QOL	
Handgrip strength	Jadaar hand dynamometer	
Cognitive function	Letter Digit Coding Test (LDCT)	
Total mortality and cardiovascular mortality		
Functional ability	Bathel Index and the Older American resources and services (OARS)	
Haemoglobin	Blood test at baseline and 1 year visit	
Blood pressure	Measured at screening and final visit	
Weight and waist circumference	Measured at screening and final visit	

12. Will these results change how treatment for SCH patients?

13. What should I do going forward

If you require more information please contact the TRUST team on (--) ------.

Appendix: 4.3: Final Patient-Preferred Result Letter

TRUST
THYROID TRIAL

Dear Participant,

Your participation in the TRUST Thyroid Trial has helped researchers answer important health questions which will be of benefit to many people in the future.

We are now sending you information about the trial and the final results of the trial. As you know, during the trial you attended a number of study visits. During these visits you completed a number of questionnaires, physical tests and weight measurements. We collected and analysed all of this information and now the final results of the trial are available.

We hope you will take the time to learn the results of the trial and we would like to sincerely thank you for taking part.

Kind regards,

PKearney

Professor Patricia M. Kearney
Principal Investigator TRUST Thyroid Trial
Dept. of Epidemiology and Public Health
University College Cork.

About the Trial…

What was the TRUST Thyroid Trial?
The Thyroid Hormone Replacement for Subclinical Hypothyroidism Trial (TRUST) was set up to better understand how to treat people with subclinical hypothyroidism.

Who was in charge of the trial?
The main study site for the trial was at the University of Glasgow, Scotland. The trial was funded by a €6 million grant from the EU's FP7 programme. This programme is the EU's main funding for research and development in Europe.

What was the aim of the TRUST Trial?
The purpose of the trial was to look at whether or not adults aged over 65 years old with subclinical hypothyroidism benefit from taking the active drug, levothyroxine. Levothyroxine replaces or provides extra thyroid hormone. Thyroid hormone is normally produced by the thyroid gland.

How long was the TRUST trial?
The trial started in May 2013 and ended in November 2016.

Who took part in the TRUST trial?
A total of 738 people took part in the trial from the following countries:

Ireland (115 people)
UK (150)
Netherlands (255)
Switzerland (218)

The hub centre for the Irish site was located at the Mercy University Hospital, Cork. There were also five other Irish sites located at Waterford University Hospital, Bantry General Hospital, Kerry General Hospital, St John's Hospital Limerick and Vista Primary Care Centre, Naas.

Why did you ask me to take part?
You were asked to take part because you are aged over 65 and a routine blood test at your doctor's surgery showed that you may have subclinical hypothyroidism (SCH).

About the Condition…

What is subclinical hypothyroidism (SCH)?

Subclinical hypothyroidism (SCH) is a mildly underactive thyroid. This means that your thyroid gland in your neck may not be producing the right amount of thyroid hormones.

What are the symptoms of SCH?

The condition often shows no symptoms or mild symptoms like:

- fatigue,
- depression
- memory problems
- cold intolerance
- consistent weight gain

In later life, the condition has also been linked to various health problems such as heart attacks and strokes.

How is SCH diagnosed?

There are two important hormones that the body needs for the thyroid to function properly. These hormones are called 'Thyroid Stimulating Hormone' (TSH) and Thyroxin (T4). Subclinical hypothyroidism (SCH) is diagnosed when a person's blood results show that their T4 levels are normal but their TSH level is mildly high (from 4.6 to 19.9 Mu/L).

How is SCH treated?

Before the TRUST trial, doctors were not sure how to treat SCH because previous research was not able to provide any answers.

About the Drug…

What is Levothyroxine?

Levothyroxine is used to treat hypothyroidism. It replaces or provides more thyroid hormone which is normally produced by the thyroid gland. This means that your body has enough thyroid hormone to maintain normal mental and physical activity.

What are the side effects of Levothyroxine?

Many people using this medication do not have any side effects. Most of the side effects are associated with hyperthyroidism (when the thyroid gland makes too much thyroxine). These include:

-temporary hair loss	-sweating	-difficulty sleeping	-vomiting and diarrhoea
-headaches	-weight loss	-chest pain	-high temperature
-flushing	-restlessness	-irregular/fast heartbeat	-muscle cramps

Please contact your GP if you have any questions about the side effects of Levothyroxine.

The Results of the TRUST Thyroid Trial.

How was the trial carried out?

After we asked you to take part in the study, we gave half of the participants the active drug (levothyroxine) and the other half a placebo. A placebo is a substance which has no active ingredient and therefore has no effect. During study visits, you will remember that you completed a number of questionnaires, physical tests and weight measurements. We collected and analysed the information and now are pleased to present you with the final overall results of the study. Please note that these are not your own personal results but the results of the study as a whole.

What were the results of the TRUST trial?

The results of the TRUST Thyroid Trial show that levothyroxine provides no apparent benefits for older people with subclinical hypothyroidism.

Further information

Outcome	Results
Thyroid specific quality of life	No differences found between placebo group and levothyroxine group.
Handgrip strength	No differences found between placebo group and levothyroxine group.
Cognitive function (ability to process thoughts and related to memory)	No differences found between placebo group and levothyroxine group.
Total mortality and cardiovascular mortality	No differences found between placebo group and levothyroxine group.
Functional ability (activities of daily living)	No differences found between placebo group and levothyroxine group.
Haemoglobin (part of red blood cells that carries oxygen)	No differences found between placebo group and levothyroxine group.
Blood pressure	No differences found between placebo group and levothyroxine group.
Weight and waist circumference (width)	No differences found between placebo group and levothyroxine group.

Should doctors treat people with subclinical hypothyroidism?

The results of the TRUST trial show that people with subclinical hypothyroidism do not benefit from taking levothyroxine. However, doctors should prescribe medication on a case-by-case basis.

What should I do now?

If you have any questions about your medical condition and whether or not you should be taking levothyroxine, please speak with your GP.

You can find more information about this study and its results:

Website: www.trustthyroidtrial.com **TRUST Telephone No:** (021) 4205595

Academic publication: http://www.nejm.org/doi/full/10.1056/NEJMoa1603825

Appendix 4.4: Standard Results Letter

TRIAL RESULTS

A mildly underactive thyroid gland (subclinical hypothyroidism) is a common condition in older age, affecting up to one-in-ten older men and women. According to current guidelines, nine of every ten women with the condition receive thyroid hormone tablets, typically levothyroxine, which has become the most prescribed drug in the USA and the third most prescribed drug in the UK. A large 5-year European study now shows that the common treatment of this condition with levothyroxine provides no apparent benefits, calling for a re-evaluation of the guidelines. The main results of the study were launched today with a publication in *The New England Journal of Medicine* along with simultaneous presentation at the Endocrine Society meeting (ENDO 2017) in Orlando, USA.

European 5-year study of 737 older adults

A team of researchers from four European Universities have followed 737 older adults (average age 74 years) to determine if levothyroxine provides clinical benefits for older people with subclinical hypothyroidism. This condition has been linked to various health problems in later life, such as tiredness or lethargy, problems with the blood circulation, muscle weakness, slowed speed of thinking, and increasing blood pressure and weight, but it is also argued that the condition causes little harm. Half of the older adults in the trial were allocated to a placebo and half to levothyroxine, and participants were followed up for at least a year. The 5-year study found that treatment with levothyroxine tablets did effectively restore a normal balance of thyroid function, but did not give any symptomatic benefits. There was also no improvement of muscle strength, speed of thinking or any effect on body weight or blood pressure. Specific advices for the oldest old (over 80 years old) will be available next year, when TRUST results will be combined with an ongoing trial among over 80s.

No worthwhile benefits from levothyroxine treatment

Based on these findings, the team concluded that there is now convincing evidence that older people with a mildly underactive thyroid do not get worthwhile benefits from levothyroxine treatment. Professor David Stott from the University of Glasgow, who led the international study, explains: "Our aim is to significantly improve the health and well-being of older people with subclinical hypothyroidism, by resolving uncertainties about how best to manage this condition. Treatment with levothyroxine is common in clinical practice, but controversial. Our study concludes this treatment provides no apparent benefits for older adults and should therefore no longer be started routinely for this condition. An update of the guidelines is necessary."

About the TRUST research project

Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism Trial (TRUST) is a European research project of experts in ageing, thyroid problems and vascular disease, investigating current treatment practices for people who suffer from a mildly underactive thyroid gland. Professor David Stott from Scotland leads the study, along with collaborators from the Netherlands (lead Professor Jacobijn Gussekloo), Switzerland (Professor Nicolas Rodondi), Ireland (Professor Patricia Kearney) and Denmark (Professor Rudi Westendorp). The data handling was performed at the Robertson Centre for Biostatistics at the University of Glasgow (lead Professor Ian Ford). The study was funded by the European Union and medicines were provided free of charge by Merck KGaA. Please view the **methods paper** and **protocol**.

The article 'Thyroid Hormone Therapy for Older Adults with Subclinical Hypothyroidism' by David Stott, Jacobijn Gussekloo, Nicolas Rodondi, Patricia Kearney, Rudi Westendorp et al. was published by The New England Journal of Medicine on 3 April 2017: ***http://www.nejm.org/doi/full/10.1056/NEJMoa1603825***

Appendix 4.5: Patient Understanding Questionnaire

Questionnaire
Your Understanding of the TRUST Thyroid Trial.

Please read carefully:

We would like to know if the information we gave you about the TRUST Thyroid Trial was useful to you. This questionnaire asks some questions about the TRUST Thyroid Trial, levothyroxine and the results of the trial.

Subject's Understanding

- I understand that my participation is voluntary.
- I understand that I will not be identified by name in the final report.
- I am aware that all documents will be kept confidential in the secure possession of the researcher.
- I understand that I may withdraw from the study at any with no adverse repercussions.

Under the **Data Protection Acts 1998 and 2003** you are entitled to make an access request for a copy of your personal information relating to this study. If you wish to make a request for access to your data, please contact Professor Patricia Kearney (021) 4205502 or patricia.kearney@ucc.ie

Plain English
Approved by NALA

For Office Use only:

Your understanding of the TRUST Thyroid Trial

The TRUST Trial looked at the effects of using the medicine levothyroxine to treat people with subclinical hypothyroidism. Subclinical hypothyroidism (SCH) is a mildly underactive thyroid. This means that the thyroid gland in the neck may not be producing the right amount of thyroid hormones. Please answer the following questions relating to the TRUST trial, levothyroxine and subclinical hypothyroidism.

Q.1 Please tick how much you agree with each statement

	Strongly Disagree	Disagree	Neither Agree or Disagree	Agree	Strongly Agree
I understand why the TRUST Thyroid Trial took place	○	○	○	○	○
I understand why I was invited to take part in the TRUST Thyroid Trial	○	○	○	○	○
I know why the medicine Levothyroxine is used to treat subclinical hypothyroidism	○	○	○	○	○
I am aware of the side effects of Levothyroxine	○	○	○	○	○
I understand the impact of Levothyroxine on thyroid-specific quality of life	○	○	○	○	○
I understand how doctors will use the results of the TRUST Thyroid Trial to treat people with subclinical hypothyroidism	○	○	○	○	○

Please tick <u>one</u> correct answer for each question

Q.2 The primary aim of the TRUST Thyroid Trial was to measure the impact of Levothyroxine on:

○ Heart and circulatory system problems ○ Thyroid-specific quality of life ○ Metabolic rate and weight gain

Q.3 A common side effect linked to Levothyroxine is:

○ Vision impairment ○ Irregular heartbeat ○ Increased sensitivity to the cold

Q.4 The results of the TRUST Thyroid Trial showed that Levothyroxine:

- Improves thyroid-specific quality of life
- Has no effect on thyroid-specific quality of life
- Disimproves thyroid-specific quality of life

Q.5 In the future, would you take Levothyroxine to treat your subclinical hypothyroidism?

Yes No Need more information

Please explain your answer in the comment box below:

Q.6 Do you think doctors should prescribe Levothyroxine based on the results of the trial? Please read the following situation and tick yes or no.

Situation A

Mary is 65 years old. She went to the practise nurse to get a blood test to check her cholesterol. She told the nurse that she has been feeling tired lately and the nurse suggested checking her thyroid level as well. When the blood results returned, the doctor told her that her T4 level (the main thyroid hormone) was normal and her TSH level (a different thyroid hormone) was higher than normal at 8.5 Mu/L meaning she has subclinical hypothryoidism. Should the doctor prescribe Levothyroxine for Mary?

☐ Yes ☐ No

Situation B

John is 80 years old and has been quite forgetful lately. His daughter was worried about him and brought him to the doctor for a routine check-up. When his blood results returned, the doctor rang and told him that his T4 level was normal but his TSH level was high at 7.8. This meant that his thyroid gland was slightly underactive. Should they ask the doctor to prescribe Levothryoxine for him?

☐ Yes ☐ No

Please give reason(s) for your answer:

Thank you for taking the time to fill out this questionnaire.
Your views are important to us.

Appendix 4.6: Costs of Conducting PPI

Phase	PPI Activity	Description	Cost
ALL	Researcher Salary	Research Assistant (3 months)	6588.75
1	Focus Groups (3 separate sessions)	Catering costs	90
		Gift vouchers for participants	400
		Printing and Stationary	11.30
		Postage	27.36
1	Public and Patient Expert Sessions (4 sessions)	Refreshments for study participants	16.00
1	NALA review	Plain English Editing- PPI results letter	230
		Plain English Review –PPI results letter and questionnaire	197
3	Result Dissemination	Printing	17.50
		Postage	104
3	Questionnaire	Printing	36.05
		Postage	332
		TOTAL COST	**€8049.96**

Appendix 5- Activities undertaken to promote PPI in research.

Appendix 5.1: 'LIstening to the Voice of Experience (LIVE)': A podcast series to promote Patient and Public Involvement (PPI) in Research.

Emmy Racine, Samantha Dick, Avril Byrne, Aileen Callanan, Ciaran Dawson, Pawel Hursztyn, Anne O' Leary, Dawn Steacy, John Walsh, Elizabeth Walsh, Sheena McHugh. Patricia M. Kearney.

Introduction

Patient and Public Involvement (PPI) is increasingly recognised as an essential component of research. However, lack of public knowledge about research and PPI has been identified as a barrier (13, 99). Innovative communication methods are needed to raise public awareness and understanding of PPI.
Aim: To develop and broadcast a podcast series about PPI partners' experiences of being involved in research.

Methods

We have two PPI groups in the School of Public Health, University College Cork: the IDEAs group (5 people with diabetes who contribute to an intervention to Improve Diabetes Eye-screening Attendance) and the MiUSE group (10 students who contribute to an intervention to reduce substance use in third level students). Podcasts were developed as following: 1) Development: Researchers and PPI contributors worked together to develop podcast content; 2) Recording: Researchers, contributors and the UCC 98.3fm station manager recorded the podcast. Recording equipment was provided by UCC98.3fm; 3) Editing: Final editing was carried out by UCC 98.3fm; 4) Launch: A public launch event was held in the School of Public Health.

Results

15 contributors were invited and 7 participated. Two podcasts were developed focusing on PPI contributors' perspectives on the research topics and experiences of being involved (see Appendix 5). You can listen to the podcast here: https://soundcloud.com/ucc98-3fm/ppi-podcast. The podcast was launched by Dr Martin Galvin, UCC Community and Civic Engagement Officer in the School of Public Health UCC on the 9/12/19 (see Appendix 5.1). Attendees included PPI contributors, school staff, students, and members of the public. The launch was hosted by Emmy Racine and included talks from Dr Martin Galvin and Dawn Steacy, a PPI contributor who participated in the podcast recording. The podcast has been broadcasted on UCC98.3fm (approx. 10,000 listeners each week) and disseminated widely online.

Conclusion

This podcast, targeting the public, highlights the different aspects of research and the diversity of opportunities available for them to become involved. It is an innovative way to communicate our PPI activities to the public. The podcast is also a useful teaching

resource that can be used to increase researchers' and students' awareness of the value and impact of PPI.

Appendix 5.2: Photos of podcast recording and podcast launch event.

Photo A5.1 LIVE podcast participants during the recording session

Photo A5.2 LIVE podcast participants during the recording session

Photo A5.3 LIVE Podcast participants attending the public event held at the School of Public Health, UCC

Photo A5.4 LIVE Podcast participants attending the public event held at the School of Public Health, UCC

Photo A5.5 Dr Martin Galvin, UCC Civic and Community Engagement Officer speaking at the podcast launch event.

Appendix 5.3: 'Patient and Public Involvement (PPI) in research: from tick-box tokenism to meaningful involvement': An educational seminar for researchers/academic community at University College Cork.

Introduction

Patient and Public Involvement (PPI) has received considerable attention in the last two decades and has now become a prerequisite for some research funders and academic journals. However, the attitudes of academic researchers and the perceived importance of PPI have been identified as important barriers (13).

Aim: To design and deliver an educational seminar on the methods and impact of PPI to researchers based at University College Cork.

Methods

A PowerPoint presentation was developed for the seminar. Slides included information on the definition of PPI, stages of the research cycle that PPI can be conducted, the rationale for PPI, advice for planning PPI activities and preparing the PPI sections of grant applications, a case example of the IDEAs (Improving Diabetes Eye-Screening Attendance study) PPI group, guidance for the payment of PPI contributors and examples of the impact of PPI. One week before the seminar, an invitation email to advertise the seminar was circulated amongst the School of Public Health email list (n=128) and amongst all academic staff within the University (n=3202).

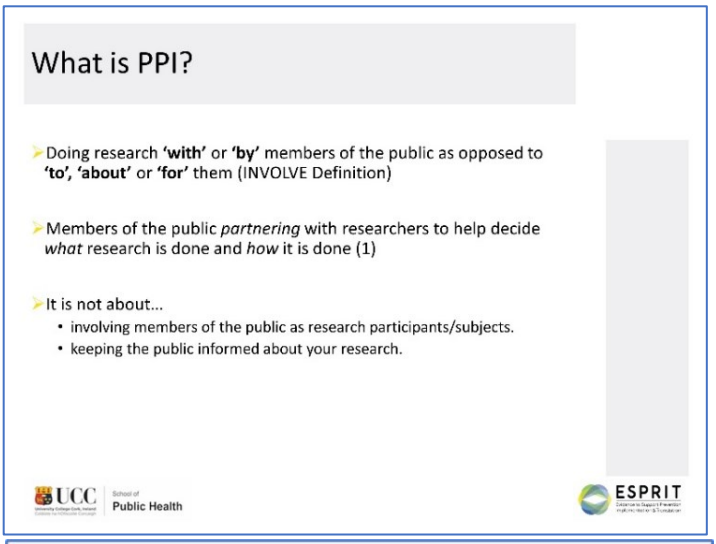
Results

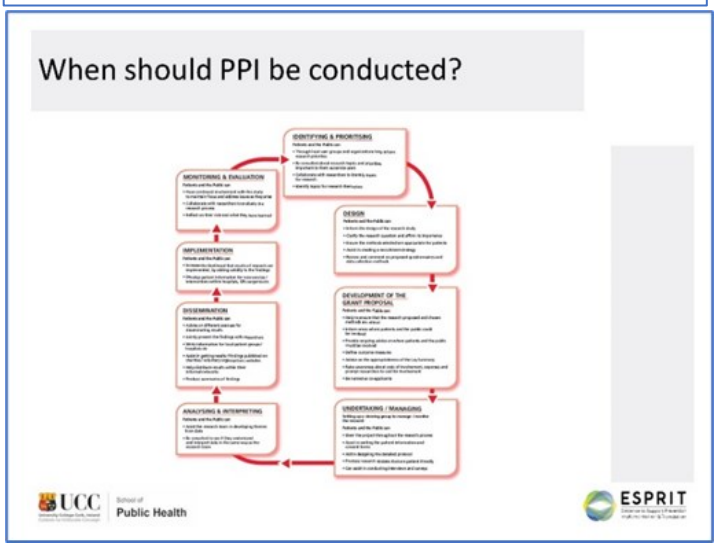
A one-hour seminar was delivered the 27th of November 2019 at the School of Public Health, UCC. The slides used during the seminar are available in Appendix A 5.4. Over 40 people attended the seminar from a variety of University schools including but not limited to the School of Public Health, School of Nursing, School of Applied Social Studies, Cork University Business School, and the School of Applied Psychology. Slides were circulated to attendees after the seminar. Follow-up contact was received from seven individuals who attended the seminar. Two of these individuals sought help preparing the PPI section of funding applications, two asked for input to their previously planned PPI activities, one requested help with facilitating PPI activities, one requested guidance on how to incorporate PPI into a doctoral research proposal and one wanted more insights into developing a podcast as a way to disseminate research to members of the public

Conclusion

This one-hour seminar was a useful way to educate researchers at University College about the methods and impact of PPI. The results show that there is an appetite for learning more about PPI amongst researchers at University College Cork. The slides used during the seminar can be used as a template for future educational seminars on PPI.

Appendix 5.4: Slides used during educational seminar





Why do I need to do PPI?

PPI is increasingly required by:

- Research funders

Public Involvement in Research
The HRB promotes the active involvement of members of the public in the research that it funds where the term 'public' includes patients, potential patients, carers and people who use health and social care services as well as people from organisations that represent people who use services. The HRB recognises that the nature and extent of active public involvement is likely to vary depending on the context of each study. Please provide details of where there has been public involvement in the preparation and/or design of this application and/or provide details of proposed future public involvement in later stages (e.g., conduct, analysis and/or dissemination). Provide information on the individuals/groups and the ways in which they will be involved. If you feel that this is not applicable to your application you are asked to explain why. The word limit is **600 words**.

- Ethics committees
- Academic journals (e.g. BMJ)

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What is the rationale for PPI?

- Moral and ethical argument
  - As taxpayers, citizens have a right to influence research that is being funded by public money (2)
  - Patients rights movements- E.g. Disability movement, HIV/AIDS activism, Breast Cancer Activism etc.

- Pragmatic argument
  - PPI can increase relevance, acceptability and accessibility of research (3,4,5)

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Example: BMJ Open

BMJ Open now requires a PPI statement for all submissions

The editorial team at *BMJ Open* have been inspired by the work of the patient involvement team at *The BMJ*. Following their lead, we are now requiring authors of all submissions to the journal to include a PPI statement.

The PPI statement should appear at the end of the Methods section. It should answer the following questions:

- How was the development of the research question and outcome measures informed by patients' priorities, experience, and preferences?
- How did you involve patients in the design of this study?
- Were patients involved in the recruitment to and conduct of the study?
- How will the results be disseminated to study participants?
- For randomised controlled trials, was the burden of the intervention assessed by patients themselves?
- Patient advisers should also be thanked in the contributorship statement/acknowledgements.

If patients were not involved, authors must state this.

Including PPI statements aligns closely with *BMJ Open*'s values of transparency and inclusiveness. We hope that including PPI statements in all articles is the first step of many for *BMJ Open* in encouraging patient involvement.

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Lets have active involvement..
Patients should be colleagues...
Research
Welcome!
I bring real life expertise..
This keeps the research grounded & realistic
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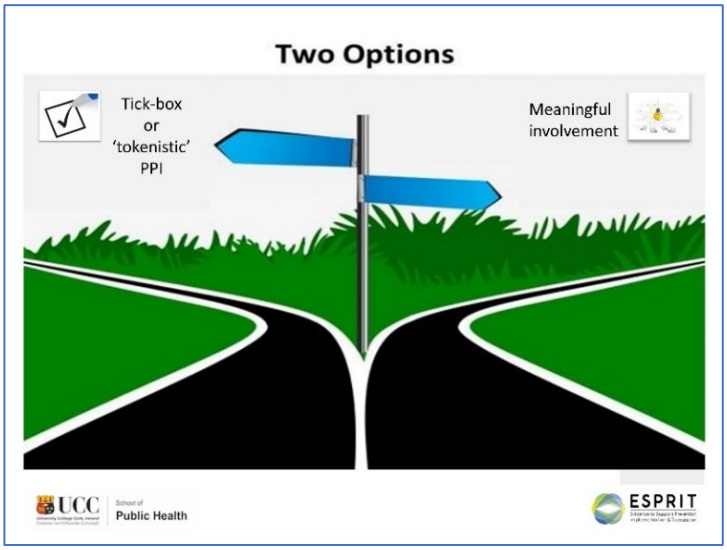
Two Options
Tick-box or 'tokenistic' PPI
Meaningful involvement
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PPI in funding applications
Public Involvement in Research
The HRB promotes the active involvement of members of the public in the research that it funds where the term 'public' includes patients, potential patients, carers and people who use health and social care services as well as people from organisations that represent people who use services. The HRB recognises that the nature and extent of active public involvement is likely to vary depending on the context of each study. Please provide details of where there has been public involvement in the preparation and/or design of this application and/or provide details of proposed future public involvement in later stages (e.g., conduct, analysis and/or dissemination). Provide information on the individuals/groups and the ways in which they will be involved. If you feel that this is not applicable to your application you are asked to explain why. The word limit is 600 words.
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PPI in funding applications

Public Involvement in Research
The HRB promotes the active involvement of members of the public in the research that it funds where the term 'public' includes patients, potential patients, carers and people who use health and social care services as well as people from organisations that represent people who use services. The HRB recognises that the nature and extent of active public involvement is likely to vary depending on the context of each study. Please provide details of where there has been public involvement in the preparation and/or design of this application and/or provide details of proposed future public involvement in later stages (e.g., conduct, analysis and/or dissemination). Provide information on the individuals/groups and the ways in which they will be involved. If you feel that this is not applicable to your application you are asked to explain why. The word limit is **600 words**.

Patients and members of the public will be involved in all aspects of the research project from the generation of the initial research idea right through to the dissemination of results. We will identify patient and public partners using existing networks known to the research team. We will ensure that they are representative of the target research population for this project. Due to ethical concerns regarding the payment of research participants, our partners will not be paid for their time, however parking and out of pocket expenses will be reimbursed. The project Principal Investigator will hold focus groups with the PPI partners to elicit their views and perspectives on how the research should be conducted. ~~Ethical approval and written informed consent will be obtained prior to the focus groups~~. These focus groups will take place when needed throughout the research project and will be ~~recorded and transcribed verbatim~~ to ensure that all perspectives are incorporated into the research being conducted.

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Meaningful PPI- the 5 W's

- Why?
  - Why are you involving people? What do you think they will add/ help with?
- Who?
  - Who are you involving? Representativeness vs Diversity, layers of involvement
  - Who is facilitating/ co-ordinating (power)
- Where?
  - Where are you going to find people? Where are you going to advertise?
  - Where are you going run meetings?
- When?
  - At what stages? (this links back to your why?)
- What?
  - What will they do in your study?

(6)

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What does meaningful involvement look like?

ideas
Improving Diabetes Eye-screening Attendance

Designated PPI coordinator(me!)

How we identified PPI contributors…

- Various strategies used (social media, diabetes support groups and education sessions, partnered with Diabetes Ireland clinics, GP waiting rooms)
- Panel established (35+ people with diabetes, family members, carers)
- 5 from this panel were recruited to be part of the PPI group.

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IDEAs PPI group

PPI contributors

- Good mix of gender, type 1 and type 2 diabetes and urban /rural contributors.

Meetings

- 4 times per year.
- Evening time-from 6-8pm.
- WGB (always the same room).
- Refreshments provided.

- Flexible format, powerpoint slides with:
  - Where the project is at
  - How their suggestions from the last meeting were incorporated
  - 2-3 items to discuss and decide on

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ESPRIT

Payment

- INVOLVE guidelines for payment of contributors: https://www.invo.org.uk/resource-centre/payment-and-recognition-for-public-involvement/

- It is good practice for PPI contributors to be paid for their time, skills and expertise.

- It is inequitable to expect people who are unpaid to work alongside other members of the research team who are paid
  - Reimbursement of expenses (incl. carer, child costs, taxi, parking)
  - Payment for time, skills and expertise

- How will you pay?

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PPI Example 1

BEFORE PPI

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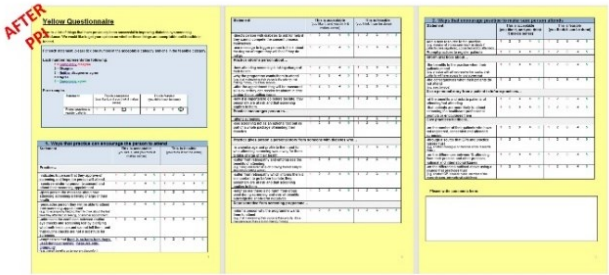

Based on the PPI feedback, we:

- Changed to 'yellow questionnaire'.
- Reduced from 5 pages to 3 pages.
- Made the instructions clearer and included an example to illustrate how it should be filled out.
- Inserted headings for each section to avoid repetition and make it easier to read.
- Inserted rating headings (e.g. acceptable and feasible) at the top of each page.
- Included definitions of the rating headings to make it more understandable.
- Combined some of the items to make it shorter and easier to complete.
- Changed some of the wording e.g. ~~testimonial~~ = personal story
- Learned the importance of a cup of tea!!!!

100% Response Rate

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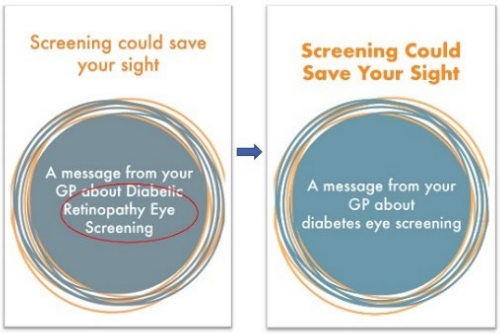
PPI Example 2



Language matters!

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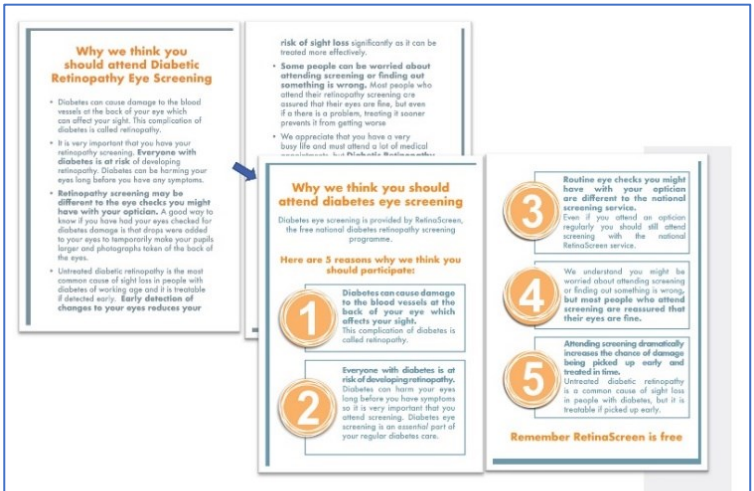

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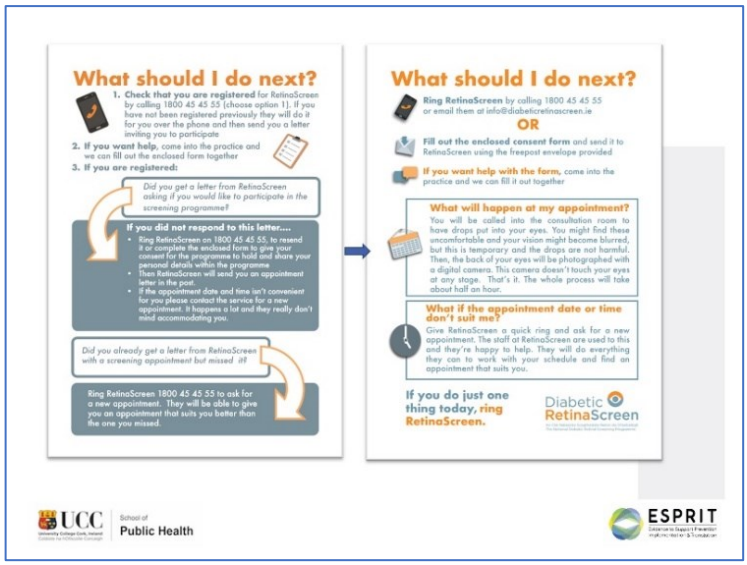
What should I do next?
1. Check that you are registered for RetinaScreen by calling 1800 45 45 55 (choose option 1). If you have not been registered previously they will do it for you over the phone and then send you a letter inviting you to participate
2. If you want help, come into the practice and we can fill out the enclosed form together
3. If you are registered:
Did you get a letter from RetinaScreen asking if you would like to participate in the screening programme?
If you did not respond to this letter....
Ring RetinaScreen on 1800 45 45 55, to resend it or complete the enclosed form to give your consent for the programme to hold and share your personal details within the programme
Then RetinaScreen will send you an appointment letter in the post.
If the appointment date and time isn't convenient for you please contact the service for a new appointment. It happens a lot and they really don't mind accommodating you.
Did you already get a letter from RetinaScreen with a screening appointment but missed it?
Ring RetinaScreen 1800 45 45 55 to ask for a new appointment. They will be able to give you an appointment that suits you better than the one you missed.
What should I do next?
Ring RetinaScreen by calling 1800 45 45 55 or email them at info@diabeticretinascreen.ie
OR
Fill out the enclosed consent form and send it to RetinaScreen using the freepost envelope provided
If you want help with the form, come into the practice and we can fill it out together
What will happen at my appointment?
You will be called into the consultation room to have drops put into your eyes. You might find these uncomfortable and your vision might become blurred, but this is temporary and the drops are not harmful. Then, the back of your eyes will be photographed with a digital camera. This camera doesn't touch your eyes at any stage. That's it. The whole process will take about half an hour.
What if the appointment date or time don't suit me?
Give RetinaScreen a quick ring and ask for a new appointment. The staff at RetinaScreen are used to this and they're happy to help. They will do everything they can to work with your schedule and find an appointment that suits you.
If you do just one thing today, ring RetinaScreen.
Diabetic RetinaScreen
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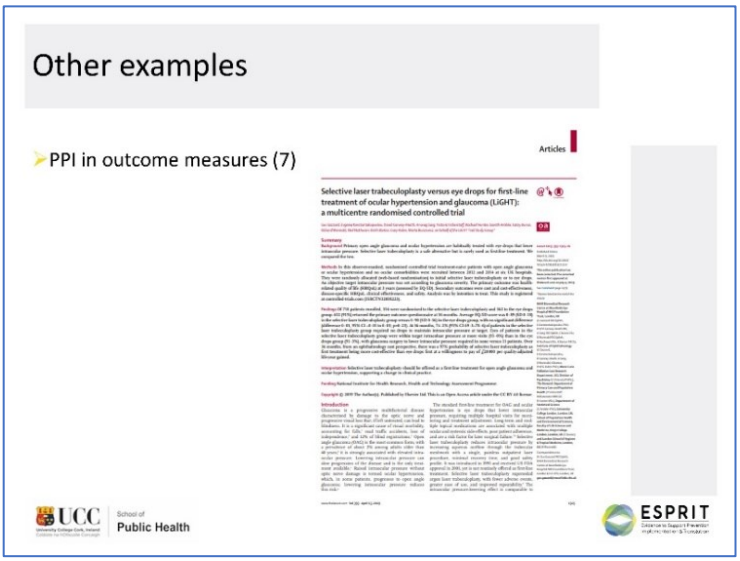
Other examples
PPI in outcome measures (7)
Articles
Selective laser trabeculoplasty versus eye drops for first-line treatment of ocular hypertension and glaucoma (LIGHT): a multicentre randomised controlled trial
UCC School of Public Health
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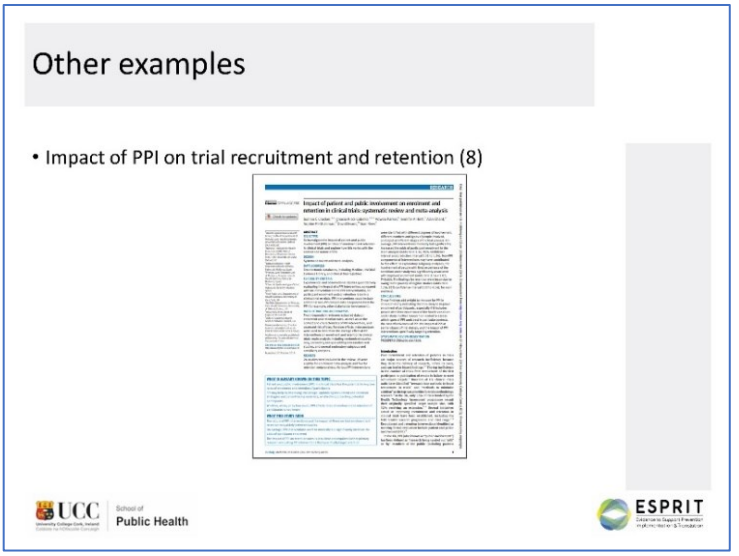
Other examples
• Impact of PPI on trial recruitment and retention (8)
Impact of patient and public involvement on enrolment and retention in clinical trials: systematic review and meta-analysis
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Other Examples

- PPI in systematic reviews (9)

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Top tips!

- Get guidance and support (emmy.racine@ucc.ie)
- Think about the 5 W's
  - Why?
  - Who?
  - Where?
  - When?
  - What?
- Remember that PPI is not research.
- Budget it properly – otherwise it's a 🚩 for funders
- Always give feedback to PPI contributors.

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Date for your diary!

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Thanks for listening!!

Thanks to the IDEAs PPI group and the PCCTNI PPI group in Galway!

More about the IDEAs study and its PPI activities:
https://www.ucc.ie/en/diabeteseyescreening/

emmy.racine@ucc.ie

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Appendix 5.5: 'Participants' Perspectives and Preferences on Clinical Trial Result Dissemination: The TRUST Thyroid Trial Experience': A blog post published on the SPHeRE blog platform.

Published on 4th October, 2018.
Available: http://www.sphereprogramme.ie/participants-perspectives-and-preferences-on-clinical-trial-result-dissemination-the-trust-thyroid-trial-experience/

The results of clinical trials are not traditionally disseminated to clinical trial participants. While there is a growing awareness that participants should receive study results, little is known about the most appropriate methods of doing so. The Thyroid Hormone Replacement for Subclinical Hypothyroidism Trial (TRUST) was a multi-centre, double-blind, randomised, placebo-controlled trial which tested the efficacy of thyroxine replacement in subclinical hypothyroidism in older adults (≥65 years). We recently conducted A Study Within A Trial (SWAT) which used a Public and Patient Involvement (PPI) approach to identify, develop and evaluate a patient-preferred method of receiving the results of the TRUST Thyroid Trial.

Using a mixed methods approach, an intervention study was undertaken at the Irish TRUST site. The first phase of the study used PPI (focus groups and 1-1 sessions with trial participants) to develop a patient-preferred result method. In the second phase, Irish TRUST participants (n=101) were randomised into the intervention (PPI method) and comparison groups (standard method). In the third phase, participants were sent a questionnaire. The primary outcome of the questionnaire was difference in the understanding of results between the two groups.

The results from the first phase clearly established that the preferred method of receiving results was a postal letter containing a 2-3page summary of the trial, condition, treatment and overall aggregated results of the trial. In phase two, the intervention group received the PPI method of results and the comparison group received the standard method as developed by the lead study site in Glasgow, Scotland. In phase three of the study, 67 participants returned a completed questionnaire (response rate 66%). The results of the questionnaire showed no difference in patient understanding between the intervention and comparison groups.

Little is understood about the impact and effectiveness of PPI in clinical trials. While this study found that PPI has no real impact on patient understanding of trial results, it provides empirical evidence on participants' perspectives and preferences of clinical trial result dissemination. It also provides a template for researchers to enhance patient and public involvement in their research

Appendix 5.6: 'The Impact of PPI on clinical trials': A blog post published on the HRB Open blog platform.

Published on 23rd May, 2019.
Available: https://blog.hrbopenresearch.org/2019/05/23/the-impact-of-ppi-on-clinical-trials/

20th May marks International Clinical Trials Day, a day dedicated to what is believed to be the first controlled clinical trial. In 1747, James Lind investigated the link between citrus fruits and scurvy using just 12 men on board the HMS Salisbury of Britain's Royal Navy fleet. Over the years, clinical research has advanced, and is vital in helping us cure illnesses and to improve our health. So, in recognition of the importance of clinical trials and public involvement, we interviewed Emmy Racine, University of Cork, Ireland, to talk about her research article, published on HRB Open Research, investigating patient and public involvement in clinical trials and how the results should be disseminated.

Why should patients and the public be involved in clinical research?
There are many ethical, moral and political arguments for involving patients and members of the public in clinical research. One such argument is that those affected by or paying for research should have a say in how it is done.
There are also pragmatic arguments being made for Patient and Public Involvement (PPI) based on the actual contribution that the public can make to research and its wider acceptability. There is some evidence that PPI can improve the quality, efficiency and impact of research but we really need stronger evidence on the impact of PPI before we can impart these claims.

Why do you think it is important to share trial results?
I think it is important to share trial results because trial participants invest a lot of time, effort and sometimes money (e.g. travel, parking, food expenses) in trials. Not only is it important to share trial results with participants but results should be shared in a way that is easy to read and understand.
A recent European Union Clinical Trial Regulation now requires trial sponsors to provide summary results of clinical trials to participants in a format understandable to lay people. This may sound like an easy thing to do but since the results are interpreted in the context of existing scientific evidence and debate, it is often difficult for trial researchers to present this information in a simple and straightforward way.

Were patients interested in receiving trial results?
Yes, I found that trial participants wanted to receive trial results. Many participants reported that they felt they had made an individual contribution to the trial in terms of their time and personal information and receiving the results of the trial would provide acknowledgement of this individual contribution.
They also believed that their participation contributed to a collective benefit or greater good and they wanted to know the results so they could better understand what this collective benefit was. They wanted to know how the results of the trial would be put into practice by doctors and how it would affect other patients in the future.

How would they like the results to be presented to them?

Participants were very clear that they wanted to receive the results in a way that was accessible and easy to understand. The majority said they would like to receive the results in a letter from the trial researchers posted to them directly. Although participants wanted an official statement of the results in a letter format, they also felt it was important to add a personal element to the letter.

They suggested offering participants a phone number that they could call if they wished to discuss any further issues or concerns with the study team. They were clear that the format, content and language of the results letter should be easy to read and understand. All participants wanted the letter to be no longer than 2-3 pages and presented in a question and answer format.

They stressed that it was important that the information presented to them was informed by medical experts but easily understood by those without a scientific or medical background.

What next?

As part of my research, I developed a questionnaire to evaluate the impact of PPI on patient understanding of trial results. While levels of patient understanding were similar between the group that received the results letter developed by the PPI group (intervention) and the group that received the information developed by researchers at the lead trial site (control), we didn't have enough people to confirm whether the PPI letter made a difference. However, some additional analysis of the questionnaire suggested that it is a useful tool for measuring patient understanding of trial results.

As the questionnaire can be easily adapted for use in other trials, it would be great to see it being used by other trial methodology researchers to add to our results and generate further evidence on the impact of PPI on clinical trials.

Appendix 6- Research Output and Dissemination

Table A6.1: Peer-reviewed publications

Year	Peer-reviewed publication
2020	**Racine E,** Phillip E, Riordan F, McHugh S and Kearney PM. 'It just wasn't going to be heard': A mixed methods study to compare different ways of involving people with diabetes and healthcare professionals in health intervention research. *Health Expectations.* 2020; 00: 1-14. doi: 10.1111/hex.13061.
2020	Riordan F, **Racine E**, Smith SM, Murphy A, Browne J, Kearney PM, Bradley C, James M, Murphy M and McHugh SM. Feasibility of an implementation intervention to increase attendance at diabetic retinopathy screening: protocol for a cluster randomised pilot trial. Pilot and Feasibility Studies. 2020:6:64. doi: 10.1186/s40814.
2020	Riordan F, **Racine E,** Phillip E, Bradley, C, Lorencatto F, Murphy M, Murphy A, Browne J, Smith SM, Kearney PM and McHugh S. Development of an intervention to facilitate implementation and uptake of diabetic retinopathy screening. Implementation Science. 2020. 15:34. doi: 10.1186/s13012.
2020	**Racine E,** Soya A, Barry, P, Cronin F, Hosford O, Moriarty E, O Connor KA, Turvey S, Timmons S, Kearney PM and McHugh SM. 'I've always done what I was told by the Medical people'- a qualitative study of the reasons why older adults attend multifactorial falls risk assessments

	mapped to the Theoretical Domains Framework. BMJ Open. 2020; 10:e033069. doi:10.1136/bmjopen-2019-033069
2019	Tracey M, **Racine E,** Riordan F, McHugh S and Kearney PM. Understanding the uptake of a national retinopathy screening programme: An audit of people with diabetes in two large primary care centres. HRB Open. 2019, 2, 17. doi: 10.12688/hrbopenres.12926.3
2019	**Racine E,** Hurley C, Cheung A, Sinnott C, Matvienko-Sikar, K, Smithson WH, Kearney PM. Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience. *HRB Open.* 2019. 1:14. doi:10.12688/hrbopenres.12817.2
2018	McHugh S, Sinnott C, **Racine** E, Timmons S, Byrne M and Kearney PM. 'Around the edges': using behaviour change techniques to characterise a multilevel implementation strategy for a fall prevention programme. Implementation Science. 2018; 13:113. Doi: 10.1186/s13012-018-0798-6
2017	**Racine E**, Hurley C, Cheung A. et al. Study within a trial (SWAT) protocol. Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience. Contemporary Clinical Trials Communications. 2017. 163-165. Doi: 10.1016/j.conctc.2017.07.001.
2017	Hurley C, Sinnott C, Clarke M, Kearney PM, **Racine E,** Eustace J and Shiely F**.** Perceived barriers and facilitators to Risk Based Monitoring in academic-led clinical trials: a mixed methods study. Trials. 2017; 18-423. Doi: 10.1186/s13063-017-2148-4.

2015	**Racine E,** Chen YW and Collins N. Gone but not forgotten: the (re-)making of diaspora strategies. Asian Ethnicity. 2015; 6; 3; 371-379. Doi: 10.1080/14631369.2013.878210.

Table A 6.2: Other outputs

Year	Output
2019	LIstening to the Voice of Experience (LIVE) Podcast. Available to listen: https://soundcloud.com/ucc98-3fm/ppi-podcast.
2019	Public event to launch the LIstening to the Voice of Experience (LIVE) Podcast. School of Public Health, University College Cork.
2019	Lunchtime seminar: ''Patient and Public Involvement (PPI) in research: from tick-box tokenism to meaningful involvement' at the School of Public Health, University College Cork.
2019	**Racine E,** Cahill D, Sheehan C and Smithson HW. The Studying age Friendly Environments (SAFE) Project Report. Report prepared for the Age Friendly Alliance (unpublished). 2019.
2018	HRB Open Blog. The Impact of PPI on clinical trials. Available: https://blog.hrbopenresearch.org/2019/05/23/the-impact-of-ppi-on-clinical-trials/
2018	SPHeRE Blog. Participants' Perspectives and Preferences on Clinical Trial Result Dissemination: The TRUST Thyroid Trial Experience. Available: http://www.sphereprogramme.ie/participants-perspectives-and-preferences-on-clinical-trial-result-dissemination-the-trust-thyroid-trial-experience/
2018	Soye A, **Racine E** and McHugh S. Report Title. Report prepared for the Falls Project Steering Group, HSE (unpublished). 2019.

Table A 6.3. Conference presentations during PhD

Year	Title		Conference
2019	LIstening to the Voice of Experience (LIVE): A podcast series to promote Patient and Public Involvement (PPI) in Research.	Poster	Psychology, Health and Medicine Conference. University College Cork, Ireland
2020	LIstening to the Voice of Experience (LIVE): A podcast series to promote Patient and Public Involvement (PPI) in Research.	Oral	6th Annual Structured Population and Health-services Research Education (SPHeRE) Conference. 25th Feb. Dublin, Ireland.
2019	'Us' and 'Them': Identifying the most suitable approach to involving patients and healthcare professionals in a consensus process to inform intervention development.	Oral (Rapid Fire Session)	Annual Scientific Meeting, Society of Social Medicine. 4th-6th Sep. University College Cork, Ireland.
2019	The Study of Age Friendly Environments (SAFE) Project.	Oral	ISS21 Ageing Cluster Research Day, 10th April,

			University College Cork, Ireland.
2019	'Us' and 'Them': Identifying the most suitable approach to involving patients and healthcare professionals in a consensus process to inform intervention development.	Poster	5th Annual Structured Population and Health-services Research Education (SPHeRE) Conference. 26th Feb. Dublin, Ireland.
2018	Seldom heard: Evaluating the impact of involving patients and the public in a consensus process to inform intervention development.	Poster	New Horizons Research Conference, School of Medicine Research Committee. 6th Dec. University College Cork, Ireland.
2018	Seldom heard: Evaluating the impact of involving patients and the public in a consensus process to inform intervention development.	Poster	International Perspectives on the Evaluation of Patient and Public Involvement in Research, 15th and 16th November. Newcastle, UK.
2018	Participants' perspectives and preferences on clinical trial result	Poster	International Perspectives on the

	dissemination: The TRUST Thyroid Trial experience.		Evaluation of Patient and Public Involvement in Research, 15th and 16th November. Newcastle, UK.
2018	Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience.	Oral (Rapid Fire Session)	Annual Scientific Meeting, Society of Social Medicine. 5th-7th Sep. University of Glasgow, Scotland.
2018	'I've always done what I was told by the Medical people'- a mixed methods study of older peoples reasons for attendance at a new fall prevention clinic.	Oral (Rapid Fire Session)	Annual Scientific Meeting, Society of Social Medicine. 5th-7th Sep. University of Glasgow, Scotland.
2018	Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience.	Oral	4th Annual Structured Population and Health-services Research Education (SPHeRE) Conference. 11th Jan. Dublin, Ireland.

2018	Older Adults Experiences of Attending Falls Risk Assessment Clinics.	Oral	4th Annual Structured Population and Health-services Research Education (SPHeRE) Conference. 11th Jan. Dublin, Ireland.
2017	Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience.	Poster	New Horizons Research Conference, School of Medicine Research Committee. 7th Dec. University College Cork, Ireland.
2017	Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience.	Poster	INFANT Research Day. 26th Oct. Cork, Ireland.

Table A 6.4: Other conferences attended during PhD

Year	Conference
2019	European Patients Forum Congress 'Advancing meaningful patient involvement: A path to more effective health systems', 12th-14th November, Crowne Plaza Hotel, Brussels, Belgium.
2018	Gender Equality in Higher Education, 20th-22nd August, Trinity College, Dublin, Ireland.
2018	'Every voice matters', PPI in Research Conference, 25th April, National University of Ireland Galway, Ireland.
2017	'Transparency in Trials', 3rd Annual Trial Methodology Symposium 2017, 20th October. Radisson Blu, St. Helens, Dublin, Ireland.

Appendix 7- PhD Education & Training

Table A7.1 Training and workshops attended during PhD

Year	Course
	Extra-credit modules, University College Cork
2018	PG7016 Systematic Reviews for the Health Sciences (5 credits)
2018	ST6013 Statistics and Data Analysis for Postgraduate Research Students (10 credits)
2018	PG6025 Community-Based Participatory Research (5 credits)
	Other training completed during PhD
2020	The impact of unpaid work on self-reported mental wellbeing, Kingston and St Georges Faculty of Health and Social Care Education (online workshop).
2020	Digital Badge in Responsible Conduct of Research. Research Integrity Training, Epigeum (online component) and University College Cork (face-to-face component).
2019	Developing and Evaluating Complex Interventions Workshop, DECIPHer (Development and Evaluation of Complex Interventions for Public Health Improvement) research group held at University College Cork.
2019	PPI in Clinical Trials: Design, Recruitment and Retention, Edinburgh Clinical Research Facility, Edinburgh, Scotland.
2019	ICH- Good Clinical Practice (ICH- GCP)
2018	The Odyssey Programme, Department of Human Resources, UCC.

2018	Training Institute for Dissemination and Implementation Research in Health (TIDIRH) Ireland, School of Public Health in collaboration with TIDIRH US, UCC.
2018	Guidance for Developing and Evaluating Complex Interventions. Medical Research Council, University of Glasgow.
2018	Writing lay summaries for research proposals. Health Research Board. National University of Ireland, Galway.
2018	Training in Patient and Public Involvement in Research, Centre for Public Engagement, Kingston and St Georges University of London, England.
2017	Introduction to Applied Biostatistics: Statistics for Medical Research, Osaka University, EdX Online Learning Platform.
2017	PredictionX: John Snow and the Cholera Epidemic of 1854, Harvard University, EdX Online Learning Platform.
2018	Normalisation Process Theory, School of Nursing and Midwifery, UCC.
2018	Turbocharge your writing, Graduate Studies Office, UCC.
2018	How to plan your PhD, Graduate Studies Office, UCC.
2018	The Seven Secrets of Highly Successful Research Students, Graduate Studies Office, UCC.

Appendix 8- Awards and additional funding obtained

Table A 8.1 Awards obtained during PhD

Year	Award
2019	Best Rapid-Fire Presentation. Joint Society for Social Medicine and Population Health 63rd Annual Scientific Meeting and European Congress of Epidemiology Meeting, 4-6th September 2019, University College Cork, Ireland.
2019	Best Poster. 5th Annual Structured Population and Health-services Research Education (SPHeRE) Conference. 26th Feb. Dublin, Ireland.
2018	Best Rapid-Fire Presentation. Annual Scientific Meeting, Society of Social Medicine. 5th-7th Sep 2018. University of Glasgow, Scotland.

Table A 8.2 Additional funding/ bursaries obtained during PhD

Month/year	Award	Amount
April 2019	Travel bursary: College of Medicine and Health, UCC.	€1000.00
April 2019	Irish Research Council Postgraduate Scholarship Scheme: *It's a nice thing to do but…. Exploring the impact of Patient and Public Involvement on Randomised Controlled Trials.*	€18,250.00
March 2019	Irish Research Council New Foundations Scheme: *Listening to liVed Experience (LIVE) Podcast Series*	€4,950.00
September 2018	Free place bursary: Annual Scientific Meeting, Society of Social Medicine. 5th-7th Sep 2018. University of Glasgow, Scotland.	€870.00
March 2018	Health Research Board- Trial Methodology Research Network (HRB TMRN), SWAT Funding Scheme: *Seldom Heard: Listening to Patients and the Public during intervention development*.	€24, 186.38

Appendix 9- Committee membership, contributions to teaching and funding applications

Table A 9.1 Committee membership

Year	Committee	Role
2019	Local Organising Committee for the Joint Society for Social Medicine and Population Health 63rd Annual Scientific Meeting and European Congress of Epidemiology Meeting, 4-6th September 2019, University College Cork.	Early Career Researcher Representative.
2019	Local Organising Committee for the Society for Social Medicine Early Career Researcher Event, 3rd September 2019, University College Cork.	Member.
2018 - present	Working Group Committee for the Athena SWAN Bronze Award Application.	Member.
2018- present	Organisation and Culture Subcommittee for the Athena SWAN Bronze Award Application.	Member.

Table A 9.2 Contributions to teaching, marking and supervision

Academic Year	Contribution
2019-2020	Supervision: Bsc Public Health Sciences Virtual work placement with the School of Public Health, University College Cork (5 students in total).
2019-2020	Teaching: EH 1006 Perspectives on Public Health. Lecture on Poverty, Social Exclusion and Community Development.
2019-2020	Marking: MPH Public Health-MPH Thesis (3 in total)
2018-2019	Teaching: EH 1006 Perspectives on Public Health. 2 hr lecture on Poverty, Social Exclusion and Community Development.
2018-2019	Tutorial: EH 1006 Perspectives on Public Health. The Young Offenders (Film and discussion).
2018-2019	Marking: BSc Public Health Sciences-Work Placement portfolios (2 in total)
2018-2019	Marking: MPH Public Health- MPH Thesis (3 in total)

Table A9.3 Contribution to research funding applications during PhD

Month/year	Funding Body	Title of Research Project	Principal Investigator	Contribution
August 2020	Health Research Board and Irish Research Council	National PPI Network	Prof Sean Dineen (NUIG)	Contributed to overall network application, designed UCC site activities, developed UCC site budget, coordinated partners contributions and processed relevant partner paperwork, prepared UCC Gantt chart.
December 2019	Heath Research Board	Improving Diabetes Eye Screening Attendance (IDEAs)	Dr Sheena McHugh	Wrote the PPI section (including costings) and coordinated a PPI review of the application.

November 2019	HRCI/ HRB	The role of Microbiota in the immune response in colorectal cancer.	Prof Paul O'Toole	Wrote the PPI section (including costings).
June 2019	Health Research Board	Centre for Understanding Tailored Methods of Implementation involving Stakeholders, Evidence and skills Development (CUSTOMISED) for policy and practice	Dr Sheena McHugh	Wrote the PPI section (including costings) and coordinated a PPI review of the application.
March 2019	Irish Research Council	LIstening to liVed Experience (LIVE)	Dr Sheena McHugh	Came up with and developed the idea for the project, wrote and submitted the application.
March 2019		Evidence for Policies to Prevent Chronic Conditions (EPICC)	Prof Patricia Kearney	Wrote the PPI section (including costings) and coordinated a PPI

				review of the application.
November 2018	Irish Research Council	Postgraduate Scholarship Scheme	Emmy Racine	Came up with and developed the idea for the project, wrote and submitted the application.
October 2018		Children's fOod Marketing Exposure (COMET): "Can I have…."	Dr Janas Harrington	Wrote the PPI section (including costings).
March 2018	Health Research Board	Reducing Maternal Stress in Ireland	Dr Karen Matvienko Sikar	Wrote the PPI section (including costings).
March 2018		Seldom Heard: Listening to patients and the public during intervention development	Prof Patricia Kearney	Came up with and developed the idea for the project, wrote and submitted the application.
December 2017	Health Research Board	A feasibility study of a social support intervention to improve mental	Dr Darren Dahly	Wrote the PPI section (including costings).

		health outcomes in older adults who experienced abuse as children		
November 2017	Health Service Executive	Learning Neighbourhoods are Active Neighbourhoods: A Community Designed and Led Physical Activity Intervention in four UNESCO Learning Neighbourhoods in Cork City	Dr Janas Harrington	Wrote the PPI section (including costings).

Appendix 10- Published Papers

Received: 5 November 2019 | Revised: 20 February 2020 | Accepted: 24 March 2020

DOI: 10.1111/hex.13061

WILEY

ORIGINAL RESEARCH PAPER

'It just wasn't going to be heard': A mixed methods study to compare different ways of involving people with diabetes and health-care professionals in health intervention research

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Abstract

Background: Guidelines recommend involving intervention users in the intervention development process. However, there is limited guidance on how to involve users in a meaningful and effective way.

Objective: The aim of this Study within a trial was to compare participants' experiences of taking part in one of three types of consensus meetings—people with diabetes-only, combined people with diabetes and health-care professionals (HCPs) or HCP-only meeting.

Design: The study used a mixed methods convergent design. Quantitative (questionnaire) and qualitative (observation notes and semi-structured telephone interviews) data were collected to explore participants' experiences. A triangulation protocol was used to compare quantitative and qualitative findings.

Participants: People with diabetes (recruited via multiple strategies) were randomly assigned to attend the people with diabetes or combined meeting. HCPs (recruited through professional networks) attended the HCP or combined meeting based on their availability.

Results: Sixteen people with diabetes and 15 HCPs attended meetings, of whom 18 participated in a telephone interview. Participants' questionnaire responses suggested similar positive experiences across the three meetings. Observation and semi-structured interviews highlighted differences experienced by participants in the combined meeting relating to: perceived lack of common ground; feeling empowered versus undervalued; needing to feel safe and going off task to fill the void.

Conclusions: The qualitative theme 'needing to feel safe' may explain the dissonance (disagreement) between quantitative and qualitative data. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately.

KEYWORDS
consensus process, intervention development, patient and public involvement, user involvement

1 | INTRODUCTION

For interventions to be successfully implemented in practice, they need to be acceptable, engaging and feasible to implement.[1] Intervention development guidelines recommend involving all appropriate intervention users to maximize the chances of successful implementation.[2] User involvement is a broad term that includes (but is not limited to) those receiving, eg patients and members of the public and delivering the intervention, eg healthcare professionals (HCPs).

Consensus methods are a way of involving multiple users simultaneously in the intervention development process.[3-5] Different users may have different priorities and preferences when making decisions about the content and delivery of an intervention.[6,7] For example, patients and members of the public may be concerned about how an intervention will be received by the target population, whereas HCPs may be more concerned about the cost involved (both time and money).[7] Group dynamics are complex, and some user groups may find it more difficult to voice their priorities and perspectives compared with others.[8] Despite increasing emphasis on user involvement, limited guidance exists on how to involve users in a meaningful and effective way. To our knowledge, no research has been conducted on patients and HCPs experiences of being involved in consensus methods and whether their experiences differ according to group composition.

The aim of this Study Within A Trial was to compare participants' experiences of taking part in one of three types of consensus meetings—people with diabetes-only, combined people with diabetes and HCPs or HCP-only meeting.

1.1 | METHODS

This Study Within A Trial (SWAT) was conducted within the on-going Improving Diabetes Eye-screening Attendance (IDEAs) study. IDEAs is a feasibility study of a multifaceted intervention in general practice targeting HCPs and people with diabetes to improve the uptake of retinopathy screening. As part of the development phase of IDEAs, three separate consensus meetings were held to discuss the acceptability and feasibility of the proposed intervention content and suitable modes of delivery. Recommendations from each meeting were used to refine intervention components that could be delivered in general practice. The first consensus meeting consisted of people with diabetes only; the second meeting consisted of a combination of people with diabetes and HCPs and the third meeting consisted of HCPs only.

1.2 | Study design

The SWAT used a mixed methods convergent design to understand and compare participants' experiences of taking part in the consensus meetings (Figure 1). A one-phase design was used, where quantitative (experience survey) and qualitative (observation notes and semi-structured interviews) methods were used during the same timeframe and were given equal weight in the analysis.[9]

Quantitative and qualitative data were collected and analysed separately. Results were merged during interpretation (mixed methods phase). A triangulation protocol was used in this phase to compare key concepts identified in each dataset that related to participants' experiences of taking part in the meetings.[9,10] The Good Reporting of A Mixed Methods Study (GRAMMS) framework and the Consolidated Criteria for Reporting Qualitative Studies (COREQ) were used to guide reporting of the findings.[11,12]

1.3 | Recruitment of participants

People with diabetes were recruited using an information flyer developed by the research team and a graphic designer (Supplementary File 1). The flyer was distributed using a range of recruitment strategies previously identified by Vat et al[13] (Supplementary File 2).

All individuals who contacted the study team about involvement were sent a 26-item demographic survey (Supplementary File 3 for survey questions and results). The individuals who returned a demographic survey were randomly assigned (using an online random number generator) to the meeting for people with diabetes-only or the combined meeting.

HCPs were recruited through professional networks known to the SWAT and IDEAs study teams. HCPs were initially sent an email or letter inviting them to take part in the consensus meeting. This was followed by a phone call to confirm their attendance. HCPs were either allocated to the HCP-only or combined meeting based on their availability to attend.

1.4 | Semi-structured consensus meetings

Before the meetings, the IDEAs study team (FR, SMH) developed (a) a short summary of existing evidence on barriers to and enablers of attendance at diabetic retinopathy screening, and interventions to address non-attendance and (b) a survey asking participants to rate intervention components according to acceptability (like it, think it makes sense) and feasibility (think it can be done).

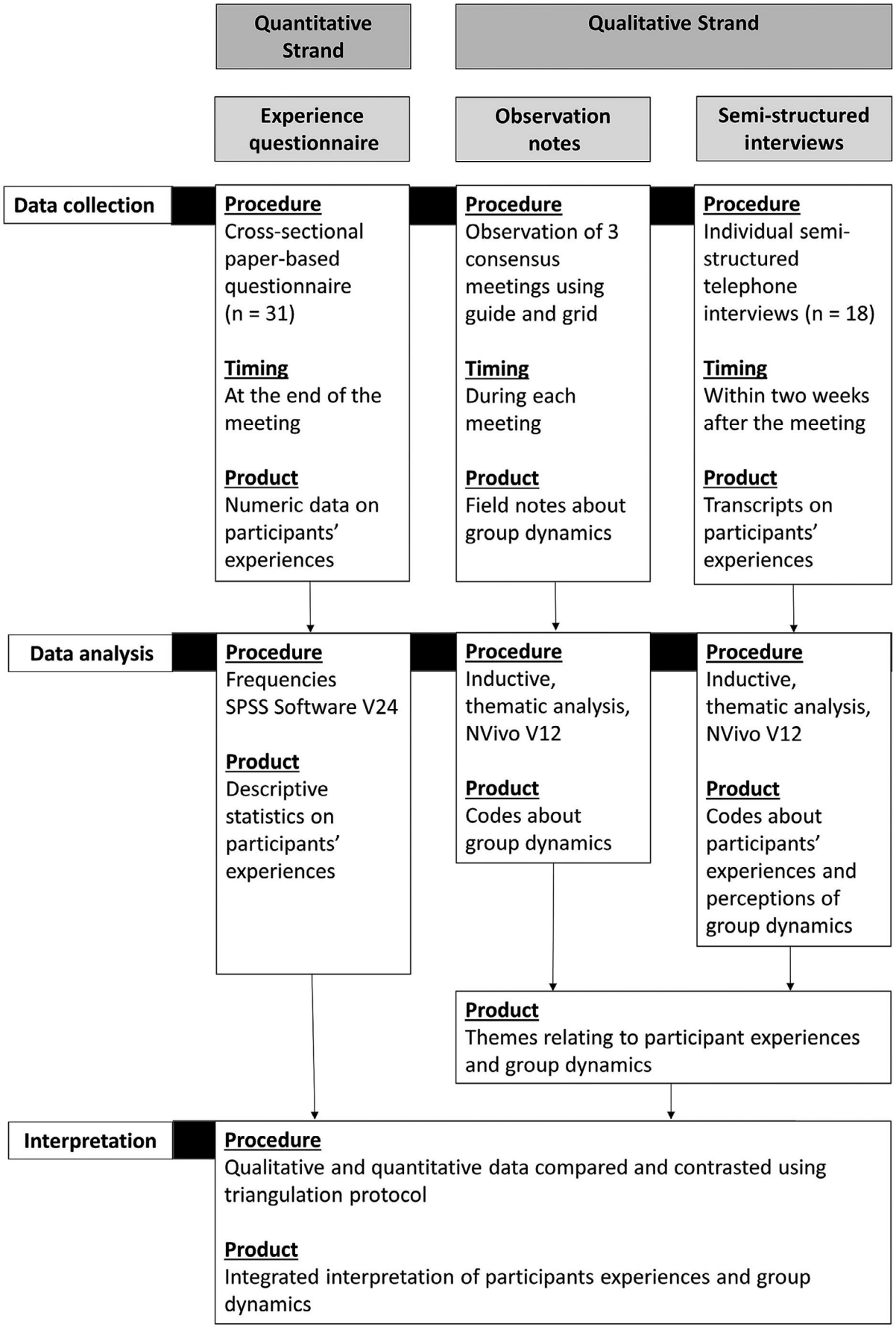

FIGURE 1 Procedural diagram of the convergent study design

The survey was based on measures developed by Weiner et al[14] Materials were reviewed by adult literacy experts (Irish National Adult Literacy Agency) and a Patient and Public Involvement (PPI) group from another research project and revised based on their feedback. Before the meeting, the evidence summary and survey was sent to all meeting participants in electronic or paper format depending on participants' preferences. Survey responses were collated and analysed descriptively by a member of the IDEAs study team (FR) and a summary of the results was prepared to be presented at each meeting.

Each consensus meeting was held from 6.30 to 8.30 pm in University College Cork. Before each meeting (at 6 pm), the lead SWAT researcher (ER) held an informal briefing for people with diabetes on key medical and research terms, the aim of the meeting and their role as patient contributors. Each meeting was facilitated by an experienced facilitator (male). During the meetings, a summary of the survey results was presented to participants, followed by a series of small group discussions facilitated by FR, SMH and EP. Participants were asked how each intervention component would work in practice and which mode of delivery would work best. Each small group was asked to nominate a lead to feed back their discussion to the larger group. Each group discussion was audio recorded.

1.5 | Quantitative strand

1.5.1 | Experience questionnaire

At the end of each meeting, all participants were asked to complete a questionnaire about their experience of the meeting. The objective of the questionnaire was to understand individual experiences of taking part in the meeting, asking them to rate how they felt about their participation and the participation of other group members; how decisions were made by the group; and the potential impact of the decisions that were made. We were unable to find a suitable validated instrument that was appropriate for our questionnaire objective and context (one-off participatory research process). Therefore, we developed our own questionnaire based on sample items from a non-validated survey instrument published by Schulz et al[15] For additional information on the questionnaire development, please see Supplementary File 4. The original phrasing of the sample items was maintained, with the exception of some questions that were changed to statements to fit with a Likert scale format. Agreement with each statement was measured on a 5-point Likert scale ranging from 'strongly disagree' to 'strongly agree'. The questionnaire also contained an open-ended comment box for any other comments or suggestions. At the bottom of the questionnaire, participants were invited to 'opt in' if they were interested in participating in a follow-up interview on their experiences of taking part in the meeting.

1.5.2 | Quantitative data analysis

Questionnaire responses were entered into SPSS software (version 24) and analysed using descriptive statistics. The five response categories were collapsed into three categories—'Agree', 'Neither agree nor disagree' and 'Disagree'.

1.6 | Qualitative strand

1.6.1 | Observation notes

The SWAT lead researcher (ER) observed each consensus meeting and took comprehensive field notes. The objective of the observation was to understand how members participated and interacted with other meeting members and how they made decisions for the development of the intervention (group dynamics and decision-making processes). An observation guide and grid were used to guide note taking and as a reminder of the events and issues of most importance (Supplementary File 5).[16] The observation guide contained two overarching questions: 'How is the group working overall?' and 'How is the group making decisions?'. The observation grid contained six constructs informed by group dynamics and decision-making processes literature.[17-20] These constructs were as follows: participation/non-participation, dominance/submissiveness, in-groups/out-groups,[1] body language and facial expressions, gaze, and effect of expert/lay knowledge. After each meeting, the researcher met with the group facilitators to discuss and document their experiences and perspectives as supplementary information.

1.6.2 | Semi-structured interviews

Within 2 weeks of the consensus meetings, semi-structured telephone interviews were conducted with the consensus meeting participants who agreed to take part in an interview in the experience questionnaire. The objective of the interviews was to gain insights into individual experiences of taking part in the meeting in terms of: how comfortable they felt in the meeting; how they felt members of the group interacted with each other and how they felt they worked together to make decisions (ie, whether there was agreement, conflict, synergy). Interviews were audio-recorded (see Supplementary File 6 for Interview Topic Guide). Interviews were conducted by ER, a young female PhD candidate. All participants were familiar with ER as she facilitated the briefing session prior to the consensus meetings. At the beginning of each interview, the SWAT lead researcher (ER) stressed to participants that she was independent to the trial study team that

[1]An in-group is a social group to which a person psychologically identifies as being a member. An out-group is a social group with which a person does not identify.

were running the consensus meetings and therefore would not be offended if they described negative experiences.

1.6.3 | Qualitative data analysis

Field notes were collated, and audio recordings were transcribed verbatim. All qualitative data were managed using NVivo software (version 12). Thematic analysis was carried out following Braun and Clarke guidelines.[21] Firstly, an extensive familiarization process was conducted by two researchers (ER, EP), where notes and transcripts were read and re-read multiple times. ER open coded all the observation notes and transcripts (using semantic and latent codes) and developed three separate sets of codes—one set for each meeting. The pattern and meanings of codes were then examined across the three meetings to identify one set of candidate or potential themes relating to participants' experiences and group dynamics. Themes were developed using a conventional or 'bottom-up' approach, whereby themes were developed directly from the data.[21] ER discussed each theme with EP to revise, refine and define themes.

1.7 | Mixed methods phase

After separate analysis of quantitative and qualitative data (as described above), the data were compared using a triangulation protocol. Triangulation provides a visual and tabular representation of the findings from qualitative and quantitative data, allowing for a clearer comparison and broader interpretation.[22] The steps taken to create the triangulation protocol are outlined in Table 1 below.

1.8 | Patient and Public Involvement (PPI) component

A PPI partner (GF) was involved in the SWAT from the outset. The PPI partner is a person with diabetes, previously known to the lead author (ER). She contributed to the initial discussions about the study which ultimately informed the SWAT grant application, reviewed the application and made changes to its content. GF was also involved in the development of materials used to recruit PPI contributors and assisted the research team with recruitment by posting recruitment flyers online via social media networks. In addition, she contributed to and reviewed each draft of this manuscript and is a co-author on this publication.

1.9 | Ethics

The study received ethical approval from the Social Research Ethics Committee (SREC) at University College Cork. Written informed consent was obtained from all participants prior to taking part in the consensus meetings and completing the questionnaire. Telephone consent was obtained from participants prior to taking part in the interviews.

2 | RESULTS

2.1 | Participants

A total of 36 people contacted the research team expressing an interest in the SWAT. Of these, 20 completed the recruitment survey

TABLE 1 Steps taken to create triangulation protocol

	Step	Activity
1.	Collate key findings from each dataset	This was done by examining the original data, interpretation and reports of analysis. For quantitative data, each questionnaire item was deemed as a separate key finding. For qualitative data, multiple key findings were identified within each theme, as themes were too broad in their descriptions to compare directly to quantitative findings
2.	Group key findings into concepts	Key quantitative and qualitative findings were grouped together into concepts according to how they related to participants' experiences and group dynamics (eg freedom of expression, balance of participation)
3.	Create table for triangulation protocol	A table was created with each column representing the data source (questionnaire, observation and interview) and each row representing a key concept
4.	Map key findings to table	Key findings were then mapped to the table to examine where findings from each method agreed (convergence), offered complementary information on the same issue (complementarity), appeared to contradict each other (dissonance) or appeared in one method and not the other (silence)[46]
5.	Explore intermethod discrepancies	This was done by examining the methodological rigour of each method and re-examining the data in light of the discrepancy[47]

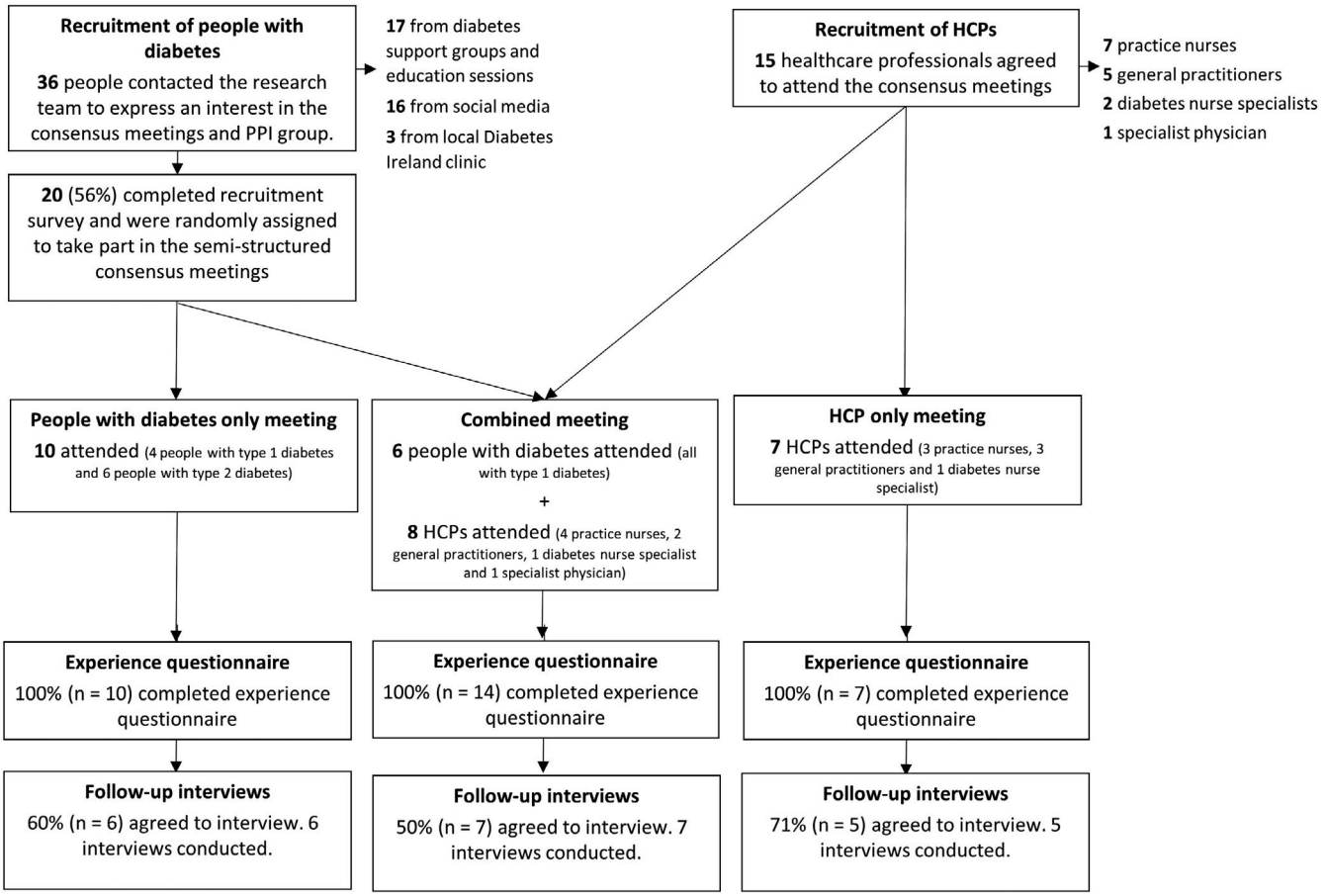

FIGURE 2 Flow diagram of recruitment and response rates

(see Supplementary File 3 for recruitment survey results). These 20 people were randomly assigned to either the people with diabetes-only meeting (4 with type 1 diabetes and 6 with type 2 diabetes) or the combined meeting (6 with type 1 diabetes, 3 with type 2 diabetes and 1 carer). All 10 people attended the people with diabetes-only meeting (attendance rate 100%) and 6 people with diabetes attended the combined meeting (attendance rate 60%). An invitation to attend was sent out to 50 HCPs (practice nurses, diabetes nurse specialists, general practitioners and specialist physicians), of whom 8 attended the combined meeting and 7 attended the HCP-only meeting (attendance rate 30%). Further details on the recruitment and response rates for each stage of the data collection are shown in Figure 2 below.

2.2 | Quantitative results

All consensus meeting participants (n = 31) completed the experience questionnaire (response rate 100%). Table 2 shows the results of the questionnaire stratified by meeting type (people with diabetes only, combined and HCP only). The descriptive statistics presented in Table 2 demonstrate that there were no differences in participants' self-reported experiences of the three meetings. All participants across the three groups agreed with the statements '*I felt comfortable expressing my opinion in the group*', '*I felt my opinions were listened to and considered by other group members*' and '*I did not feel pressured to go along with the decisions of the group even though they did not agree*'. Some participants agreed with the statements that '*I thought that certain individuals spoke more than others in the group*' and '*I felt that certain individuals had more influence over the decision-making process than others*'. A number of participants expressed doubt that they could influence the decisions made during the meeting.

2.3 | Qualitative results

In total, 18 questionnaire respondents agreed to be contacted for a follow-up interview. Interviews were conducted with participants from the people with diabetes-only (n = 6), combined (n = 7) and HCP-only (n = 5) meetings. Interviews were, on average, 34 minutes in duration (range 18-56 minutes).

Four themes were developed from the qualitative data relating to participants' experiences and group dynamics: perceived lack of common ground; feeling empowered versus undervalued; needing to feel safe and going off task to fill the void.

TABLE 2 Results of the participant experience questionnaires stratified by meeting type

Item	Meeting	Agree N (%)	Disagree N (%)	Neither agree nor disagree N (%)
I felt comfortable expressing my opinion in the group	People with diabetes	10 (100)	-	-
	Combined	14 (100)	-	-
	HCP	7 (100)	-	-
I felt my opinions were listened to and considered by other group members	People with diabetes	10 (100)	-	-
	Combined	14 (100)		-
	HCP	7 (100)	-	-
I felt part of the group (like I belonged to the group)	People with diabetes	10 (100)	-	-
	Combined[a]	12 (92.3)	-	1 (7.7)
	HCP	7 (100)	-	-
I felt pressured to go along with the decisions of the group even though I did not agree	People with diabetes	-	10 (100)	-
	Combined	-	14 (100)	-
	HCP	-	7 (100)	-
I felt a sense of trust and openness between group members	People with diabetes	10 (100)	-	-
	Combined	13 (92.9)	-	1 (7.1)
	HCP	7 (100)	-	-
I thought that certain individuals spoke more than others in the group	People with diabetes only	3 (30)	6 (60)	1 (10)
	Combined	4 (28.6)	6 (42.8)	4 (28.6)
	HCP	3 (42.9)	3 (42.9)	1 (14.2)
I felt that I could influence the decisions made by the group	People with diabetes	7 ((70)	-	3 (30)
	Combined	8 (57.1)	1 (7.1)	5 (35.7)
	HCP[a]	4 (66.7)	-	2 (33.3)
I felt that certain individuals had more influence over the decision-making process than others	People with diabetes	3 (30)	6 (60)	1 (10)
	Combined	2 (14.3)	9 (64.3)	3 (21.4)
	HCP	1 (14.3)	3 (42.9)	3 (42.9)
I have increased my knowledge about important topics since participating in this group	People with diabetes[a]	8 (88.9)	-	1 (11.1)
	Combined	10 (71.4)	1 (7.1)	3 (21.4)
	HCP	6 (85.7)	-	1 (14.3)
By working together, we can influence decisions that affect the research process	People with diabetes only	10 (100)	-	-
	Combined	13 (92.9)	-	1 (7.1)
	HCP	7 (100)	-	-
By working together, we can influence decisions that affect people with diabetes	People with diabetes	10 (100)	-	-
	Combined	14 (100)	-	-
	HCP	7 (100)	-	-

[a]Missing data.

2.4 | Perceived lack of common ground

In the people with diabetes-only meeting, there were differences between participants in terms of diabetes type, length of diagnosis and education level. In the HCP-only meeting, differences included profession (eg medical doctor, practice nurse, diabetes nurse specialist), experience of working with people with diabetes, and size, location and nature of their practices. During the interviews, participants from these two meetings described these demographic, geographical and clinical differences as *'small'* differences, which they welcomed as they felt it allowed them to bring different perspectives to the topics they were discussing. They focused on the common ground they shared with other meeting participants and identified with one another based on the shared experience of living with diabetes or caring for people with diabetes. They felt that they were all *'singing from the same hymn sheet'* (P3, person with diabetes, people with diabetes-only meeting) and described being able to come together to make decisions that incorporated different perspectives:

> *It was interesting to hear their views. We were all on the same page, but we were coming from different angles and we used that then; we came together and made the decisions together.*
>
> *(P2, person with diabetes, person with diabetes-only meeting)*

In contrast, a lack of common ground was reported by participants in the combined meeting. This created a division in the group, a 'them' and 'us' attitude, which was evident in the interview and observation data. In the interview data, people with diabetes stated that there was *a 'complete clash of perspectives'* (P9, person with diabetes, combined meeting) between people who lived with the condition and HCPs who cared for people with diabetes. HCPs reported that people with diabetes and HCPs were '*two different sides of the divide'* (P11, HCP, combined meeting). The observation data also suggested a division between people with diabetes and HCPs in the combined meeting. At the beginning of the meeting, people with diabetes and HCPs sat on opposite sides of each small table. During the small group discussions, participants expressed their opinions as collective opinions of their stakeholder group. Rather than expressing individual opinions (eg 'I think that…'or 'My experience is…'), people with diabetes spoke on behalf of all the people with diabetes in the group, and HCPs spoke on behalf of all HCPs in the group (eg 'We feel that… don't we?' and 'As people with diabetes, we think that…'). Moreover, during the small group discussions, each stakeholder group focused their gaze on the other stakeholder group, resulting in people with diabetes and HCPs talking at each other, at opposite sides of each table. This was in contrast to the people with diabetes-only and HCP-only meeting, where members focused their gaze on all members around the table.

Participants' lacking a sense of shared experience was accompanied by differences in perceptions around the balance of participation. During all three meetings, it was observed that some participants spoke more frequently and for longer than others. In the interviews, participants from the people with diabetes-only and HCP-only meetings perceived this unbalanced participation as a natural consequence of any group dynamic. They mainly attributed it to different personalities. In contrast, HCPs from the combined meeting attributed the unbalanced participation to people putting too much emphasis on their own personal experiences:

> *It was very much centred around them [people with diabetes] and a lot of the offerings that I had in terms of experience were nothing in comparison to what they felt as people that have the problem. Which is fine. But that wasn't really the point. The point is that I don't have diabetes, that is not my personal experience. But I am still the one left in the room everyday trying to deal with patients… But I just couldn't come out with it on the night. I just didn't. It wasn't going to be heard.*
>
> *(P12, HCP, combined meeting)*

2.5 | Feeling empowered versus undervalued

In the interviews, participants from the people with diabetes- and HCP-only meetings reported learning from other meeting members and feeling empowered by the event. In the people with diabetes-only meeting, participants stated that they learned from one another about how they can better manage their condition and about the difference between type 1 and type 2 diabetes. Those who had been diagnosed with diabetes for a long time described gaining a renewed compassion for those who were newly diagnosed. Participants from the HCP-only meeting reported learning about the importance of encouraging their patients to attend screening, about the roles of different HCPs and about the cultural difficulties and language barriers that some practices face due to a high number of non-English speaking patients.

There were also some reports of learning in the combined meeting. People with diabetes said they gained a new insight into the work practices of HCPs—in particular, the increased workload experienced by HCPs. The HCPs reported gaining an insight into the struggles of having to live with a medical condition:

> *I put in a couple of thousand eye drops a year, it doesn't mean anything to me like. But it obviously means something for patients who are having to go through this – and you know the awkwardness of getting appointments and driving to and from appointments and getting a lift and all that side of things.*
>
> *(P14, HCP, combined meeting)*

However, participants from the combined meeting reported feeling undervalued by the other stakeholder group. People with diabetes

felt that HCPs did not understand how it feels to live with a chronic illness, describing *'a complete clash of the reality of living with diabetes versus a medical professional's perspective'* (P7, person with diabetes, combined meeting). Among some of the HCPs, there was a sense that any contributions they made during the meeting were not valued by people with diabetes because the experience of living with diabetes was deemed more important than the experience of caring for people with diabetes:

> *I've worked in four different GP practices at this stage and all very different. And yet I felt like as if any value that I had to add to the conversation was kind of almost either misheard or not really heard, or almost felt not quite as relevant because of their personal experiences. Which is fair enough. But that was not what the meeting was about.*
>
> *(P13, HCP, combined meeting)*

2.6 | Needing to feel safe to express honest opinions

In the interviews, participants from the people with diabetes-only and HCP-only meetings reported an open, honest, relaxed and non-judgemental environment, where everyone had a voice and was heard. This environment made participants feel safe and comfortable to express their opinions. They also indicated that the small group discussions added to their feelings of safety as people who do not like speaking in public felt less intimidated about expressing their opinions:

> *I'm not one really for expressing my opinions. I am kind of … I wouldn't put my hand up the first time, let's say. But I did feel very comfortable expressing my opinion in the small group.*
>
> *(P15, HCP, HCP-only meeting)*

Conversely, participants from the combined meeting reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room. Both people with diabetes and HCPs said they felt they had to *'hold back'* their opinions. People with diabetes reported feeling that they could not be honest about the *'non-compliant'* (P9, person with diabetes, combined meeting) aspects of managing their diabetes as the HCPs may judge them for it:

> *I don't think when you are sitting at a table with HCPs that you're going to be discussing the non-compliant things you do… It's probably not the best environment, let's say, to get out some of the smaller things that people do that may not be approved by the other group in the room.*
>
> *(P8, person with diabetes, combined meeting)*

On the other hand, HCPs were conscious of confidentiality issues: they were concerned that if they mentioned a particular case, people with diabetes could potentially identify who that patient was, since *'[this location] is a very small place'* (P11, HCP, combined meeting):

> *I felt a bit kind of reticent about how free [I could talk about my experiences as a healthcare professional]… It's different when you are divulging, you know, work practices and difficulties and challenges and personal experiences at work, when it is other medical professionals. But when you have effectively patients there, it is like a big difference.*
>
> *(P13, HCP, combined meeting)*

In addition, the HCPs indicated that they did not feel comfortable talking about the service that they worked in as they felt anxious that people with diabetes would confront them on the long waiting times or other issues they had with that particular service.

2.7 | Going off task to fill the void

Analysis of interview data indicated that participants across all three meetings felt they were able to work together. They reported that the content for discussion was relevant to them as users and providers of health services.

However, the observation data show that although members of the combined meeting appeared to work together, both stakeholder groups were defensive about what intervention components would not work and at times in the meeting nothing seemed feasible. This resulted in each stakeholder group feeling uncomfortable in asserting what they felt the other group should or should not do. To fill this void, participants began to go off task as they focused their discussions on the 'other'. The 'other' took different forms throughout the meeting: the screening service, people with diabetes who were not in the room (eg those with type 2 diabetes), and funding and resource limitations in general practice. Even though they were being asked to discuss and make recommendations on how the intervention would work in primary care, the combined meeting participants resorted to making recommendations about how screening uptake could be increased on a national basis through nationwide TV and radio campaigns.

2.8 | Mixed methods results

The results of the mixed methods analysis are presented in Table 3. Six key concepts relating to participants' experiences and group dynamics were identified from the datasets: freedom of expression; understanding and respect; balance of participation; learning; productive collaboration and group cohesion. When key findings were mapped to the overarching concepts, there were four instances of

TABLE 3 Results of mixed methods analysis (triangulation protocol)

	Quantitative strand	Qualitative strand		
Key concept	**Questionnaire**	**Observation notes**	**Interviews Interviews**	
Freedom of expression	In all three meetings, participants were comfortable expressing their opinions and felt a sense of trust and openness between group members	In the combined meeting, participants did not appear to be comfortable asserting what the other stakeholder group should/should not be doing	In the people with diabetes-only and HCP-only meetings, participants reported that it was an open, honest and relaxed environment where they felt comfortable expressing their opinions In the combined meeting, participants reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room	Dissonance
Understanding and respect	In all three meetings, participants felt their opinions were listened to and considered by other group members, and that they could influence the decisions being made by the group	-	In the combined meeting, participants reported feeling undervalued by the other stakeholder group	Dissonance
Balance of participation	In all three meetings, some participants felt that certain individuals spoke more than others and had more influence over the decision-making process	In all three meetings, some participants spoke more frequently than others and for longer lengths of time	In the people with diabetes-only and HCP-only meetings, participants were understanding of the unbalanced participation and saw it as a natural consequence of any group dynamic In the combined meeting, HCPs attributed unbalanced participation to people putting too much emphasis on their own personal experiences	Convergence, complementarity
Learning	In all three meetings, most participants felt they increased their knowledge as a result of attending	In all three meetings, participants appeared keen to learn from one another as they asked each other about their experiences	In all three meetings, participants reported learning from one another and provided specific examples of this learning	Convergence, complementarity
Productive collaboration	In all three meetings, participants reported that they were able to work together to influence decisions that affect the research process and people with diabetes	In the combined meeting, although participants appeared to work together, each stakeholder group did not make any comments on what the other stakeholder group should/should not do. Instead, they made recommendations that were not relevant to the intervention (unproductive collaboration)	In all three meetings, participants reported being able to work together as they felt the content for discussion was relevant to them as users and providers of health services	Dissonance

(Continues)

TABLE 3 (Continued)

	Quantitative strand	Qualitative strand		
Key concept	**Questionnaire**	**Observation notes**	**Interviews Interviews**	
Group cohesion	In all three meetings, participants reported they were part of the group (like they belonged to the group)	In the combined meeting, it was evident that there was a division between both stakeholder groups (eg both groups spoke at each other across the each table as opposed to with each other around each table).	In the people with diabetes-only and HCP-only meetings, participants reported that there were some 'small' differences between meeting members, but added that this was a good thing as it allowed them to bring different perspectives to the topics they were discussing In the combined meeting, people with diabetes reported that there was a 'complete clash of perspectives' between people with diabetes and HCPs; HCPs reported that people with diabetes and HCPs were 'two different sides of the divide'	Dissonance

dissonance (where data appeared to contradict each other), two instances of convergence (where data agreed) and two instances of complementarity (where data offered complementary information on the same issue). There were no instances of silence (where data appeared in one method and not in the other).

The four instances of dissonance between quantitative and qualitative data were wholly due to the fact that in the questionnaire participants reported positive experiences of taking part in the meetings, whereas the observation and interview data highlighted some negative experiences and divergent opinions. For example, in relation to *freedom of expression*, the questionnaire data showed that in all three meetings, participants reported feeling comfortable expressing their opinions and reported a sense of trust and openness between group members. In the observation data, participants in the combined meeting did not appear to be comfortable asserting what the other stakeholder group should/ should not do as part of the intervention. Furthermore, in the interviews participants reported feeling uncomfortable and unable to express their opinions as they were conscious of the other stakeholder group in the room.

The instances of complementarity were largely due to the design of the data collection tools. The questionnaire items were designed to be concise and did not require the participants to give any additional details. Whereas in the interviews, participants had the opportunity to expand and give more detail. For example, in the key concept *learning*, the questionnaire item asked participants to indicate how much they agreed with the statement 'I have increased my knowledge about important topics since participating in this group', whereas in the interviews participants had the opportunity to expand and give specific examples of what they had learned (eg people with diabetes learned how they can better manage their condition, HCPs leaned about the importance of encouraging their patients to attend screening, etc).

3 | DISCUSSION

3.1 | Summary of key findings

The aim of this study was to compare participants' experiences of taking part in the three consensus meetings. The results of the questionnaire suggested that participants had largely positive experiences of taking part in the consensus meetings and there were no differences in participants' experiences between the three meetings. However, results of the observation and interviews highlighted that participants in the combined meeting had different experiences from those in the other two meetings. The perceived lack of common ground between people with diabetes and HCPs in the combined meeting led participants to feel undervalued by the other stakeholder group as they felt that the other group did not understand their perspective. Participants in the combined meeting were reluctant to express their opinions and were defensive about what would/ wouldn't work in terms of developing the intervention. As a

result participants in the combined meeting went off task and made recommendations which were not entirely relevant for the intervention. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately.

3.2 | Links to existing literature

In the people with diabetes-only and the HCP-only meetings, participants welcomed their diversity as it allowed them to hear different perspectives on the topics they were discussing. This finding is consistent with existing literature, with many theorists arguing that knowledge diversity can improve group performance by enhancing a group's ability to be creative and to discover novel solutions.[23-25] In these meetings, participants focused on their common ground and described being able to come together to make decisions that incorporated a range of perspectives. Previous research suggests that congruent groups—ie when group members are socially tied and share the same information—are more likely to be productive and successful.[26]

The perceived lack of common ground between people with diabetes and HCPs in the combined meeting created a 'them' and 'us' scenario, with participants reluctant to express their opinions. This raises questions about whether too much difference within groups is counterproductive or divisive. Existing research on the productivity of incongruent groups—ie when social and knowledge subgroups are present within a group has found that subgroups can create a divide between group members, undermining the groups' ability to work together and be productive.[26]

Some HCPs in the combined meeting felt their contributions were not valued by people with diabetes because the experience of living with diabetes trumped the experience of caring for people with diabetes. This finding may reflect the changing nature of the patient/HCP relationship over the last 20 years—from a paternalistic model where the patient seeks help and is compliant to the professional who makes the decisions, to a more patient-centred approach.[27] This approach expects HCPs to enter the patient's world and to see the illness through the patient's eyes.[27] This prioritization of the patient experience has benefited patient outcomes.[28] However, as HCPs are often responsible for delivering interventions, their perspectives in the intervention development process are crucial for maximizing intervention feasibility. Involving multiple users in the intervention development process is not about understanding which perspective is more valid or more important, it is about understanding all the different perspectives so that the intervention is more acceptable, engaging and feasible to implement.

3.3 | Strengths and limitations

One of the strengths of this study was the use of a mixed methods, convergent design which produced a more complete understanding of participants' experiences and group dynamics. It also allowed for the cross-validation of findings from each method resulting in more substantiated findings than sequential designs or quantitative or qualitative approaches alone.[9] The qualitative theme 'needing to feel safe' may explain the instances of dissonance between quantitative and qualitative data as participants completed the questionnaire at the end of each meeting while they were still sitting close to other participants. Some small groups even filled out the questionnaire together. As a result, participants may not have felt comfortable voicing concerns. In the interviews, on the other hand, participants may have felt safer in a one-to-one environment with a researcher who they were already familiar with. The fact that the researcher stressed that she was independent to the consensus meeting research team and her informal approach may have made them more comfortable to speak openly about their experiences of taking part in the meeting. The timing of the questionnaire may have also played an important role. The questionnaire was handed out at the end of the meeting, late in the evening. Participants may have been eager to get home and they may not have fully thought about the responses they were providing. However, in the interviews, participants had time to reflect on their experiences and provide a more comprehensive account as a result. This is consistent with Krosnick's theory of survey satisficing which is based on the assumption that optimal survey completion involves doing a great deal of cognitive work, so if the participant is not fully motivated to complete the survey, he or she is likely to offer responses that seem reasonable and easy to defend.[29] Although questionnaires are a frequently used tool to evaluate consensus meetings, our findings suggest that they may not always provide a comprehensive assessment of participants' experiences. This is consistent with a number of previous studies on evaluating participant experiences.[30-32]

This study is not without limitations. First, the questionnaire that was used to understand participants' experiences was based on non-validated questionnaire items. We were unable to conduct exploratory factor analysis to validate our questionnaire as our sample size did not meet the minimum criteria of 10 participants per questionnaire item.[33] However, given the increasing importance of evaluating PPI and other participatory research activities,[34] the questionnaire could be a useful tool in future studies which aim to understand stakeholders' experiences in similar participatory research contexts. Use of the questionnaire in future studies may allow for reliability testing and validation to be carried out.[35,36]

Second, although the experience questionnaire suggested that there were no differences in participants' experiences between the three meetings, due to the number of participants, there was limited power to detect a difference (n = 31). Thus, the comparison of participants' questionnaire responses between the groups is used as only an indicator of participants' experiences. Given the small sample, we cannot rule out the possibility that differences between the groups could be detected had a larger sample size been used.

Despite using a range of strategies to recruit a representative sample of people with diabetes, another potential limitation of this study was the absence of people with type 2 diabetes in the combined meeting. As the attendance rate of people with diabetes at the combined meeting (60%) was much lower than the people with

diabetes-only meeting (100%), it is plausible that people with type 2 diabetes did not attend because they knew there would be HCPs attending. Existing research has established that people with type 1 and type 2 diabetes have different experiences when managing their condition and engaging with HCPs.[37-39] Therefore, the involvement of people with type 2 diabetes in the combined meeting could have potentially changed the nature of the relationship between patients and HCPs and led to different participant experiences and group dynamics.

Finally, participants were given a choice to participate in an in-person or telephone interview. All participants chose telephone interviews due to time constraints and location convenience. This could be another potential limitation as researchers have previously expressed concerns about whether telephone interviews are appropriate for qualitative research.[40,41] These concerns are largely due to the absence of visual cues which may result in the loss of informal communication and contextual information, the inability to develop rapport or to probe and the misinterpretation of responses.[41] In this study, the quality of telephone data cannot be compared with in-person data as no in-person interviews were conducted. However, the researcher had considerable experience conducting phone interviews, maintained a friendly and engaging tone throughout and as mentioned previously, participants were found to be open and frank about their experiences.

3.4 | Implications

The results of this study provide much needed evidence on how different ways of involving patients and health-care professionals can lead to differing participant experiences and group dynamics. Patient and public involvement (PPI) in research is increasingly becoming a requirement in health research and for many research funders. INVOLVE, a national advisory body funded by the National Institute for Health Research (NIHR) in the UK, defines public involvement as research being carried out 'with' or 'by' members of the public rather than 'to', 'about' or 'for' them.[42] In this study, the lines between research participation and involvement were blurred, as is often the case with PPI.[43] People with diabetes were research participants in the consensus meetings, experience questionnaire and semi-structured interviews. However, their role in the consensus meeting was to discuss and make decisions about the intervention content and mode of delivery which could be viewed as PPI.[44,45] This study shows that the context and nature of involvement can have important implications for its impact. These findings are not only relevant to health intervention researchers but to all individuals interested in involving patients and members of the public in health research, policy and in the planning and development of health care more broadly.

4 | CONCLUSION

Although the results of the experience questionnaire showed no differences in participants' experiences across the three meetings, the results of the observation and interviews highlighted that participants in the combined meeting had different experiences. In this study, involving patients and HCPs simultaneously in a consensus process was not found to be as suitable as involving each stakeholder group separately. The study provides much needed evidence on how different ways of involving patients and health-care professionals can lead to differing participant experiences and group dynamics.

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CONFLICT OF INTEREST

The authors have no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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RESEARCH ARTICLE

REVISED Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience [version 2; peer review: 2 approved]

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Abstract

Background: While there is an increasing consensus that clinical trial results should be shared with trial participants, there is a lack of evidence on the most appropriate methods. The aim of this Study Within A Trial (SWAT) is to use a patient and public involvement (PPI) approach to identify, develop and evaluate a patient-based approach to receiving trial results for participants in the Thyroid Hormone Replacement for Subclinical Hypo-Thyroidism Trial (TRUST), a trial of thyroxine versus placebo in people aged 65 years and older.

Methods: Mixed methods study with three consecutive phases. Phase 1 iteratively developed a patient-based approach using semi-structured focus groups and a consensus-orientated-decision model, a PPI group to refine the method and adult literacy review for plain English assessment. Phase 2 was a single-blind parallel group trial. Irish TRUST participants were randomised to the intervention (patient-based approach) and control group (standard approach developed by lead study site). Phase 3 used a patient understanding questionnaire to compare patient understanding of results between the two groups.

Results: Participants want to receive results of clinical trials, with qualitative findings indicating three key themes including 'acknowledgement of individual contribution', 'contributing for a collective benefit' and 'receiving accessible and easy to understand results'. Building on these findings, the patient-based approachwas developed. TRUST participants (n=101) were randomised to the intervention (n=51) or control group (n=50). The questionnaire response rate was 74% for the intervention group and 62% for the control group. There were no differences in patient understanding between the two approaches.

Conclusions: We have demonstrated that it is feasible to involve trial

Open Peer Review

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	1	**2**
version 2 (revision) 22 Mar 2019	✓ report	
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Any reports and responses or comments on the article can be found at the end of the article.

participants in the development of result dissemination materials. Although, in this study PPI did not influence patients' understanding of results, it documents the process of conducting PPI within the clinical trial setting.

Keywords
Patient and public involvement, patient involvement in clinical trials, study within a trial, SWAT, Clinical trial result dissemination, study results, research dissemination, trial results.

This article is included in the HRB-TMRN collection.

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REVISED **Amendments from Version 1**

This improved version contains some minor revisions as suggested by peer-reviewers.

Throughout the manuscript, the following changes have been made:

- "patient- preferred" has been changed to "patient-based".
- "patient-preferred method" has been changed to "patient-based approach".
- "Standard method" has been changed to "standard approach".

Within the Abstract, the aim of the study has been re-worded to clarify that all TRUST participants were aged 65 and over.

Within the Introduction section, additional background information has been provided on the need to evaluate the impact of PPI. This serves as a rationale for doing the study. We have also introduced the recent movement towards transparency in trials including references to the SPIRIT, CONSORT and AllTrials initiatives.

Within the Methods section, additional details have been provided on the PPI group and how PPI partners were identified and recruited. Further information has also been provided on the Consensus Oriented Decision Making (CODM) model and how the model was specifically used in this study. We have also provided a clear distinction between adult literacy and health literacy.

Within the Results section, a footnote has been added to Table 1 to clarify that only a subgroup of Irish participants were invited to the focus groups. A footnote has also been added to Table 2 to clarify how patient understanding was assessed.

Within the Discussion, the section entitled 'Limitations of the study' has now been reworded to 'Strengths and limitations of the study' and the paragraph that discusses how PPI partners were participants in the trial has been rephrased as a strength of the study.

See referee reports

Introduction

Patient and public involvement (PPI) is increasingly recognised as an essential component of clinical research. In the UK, the national advisory group supporting active public involvement in health services, public health and social care research (INVOLVE) defines PPI as 'research being carried out 'with' or 'by' members of the public rather than 'to' 'about' or 'for' them'[1]. In clinical trials, PPI has been defined as experimenting with participants instead of experimenting on participants[2]. PPI may occur at any stage during the research process from priority setting and drafting study protocols right through to conducting the study, interpreting the end results and communicating and disseminating research findings[3,4]. Research funders increasingly expect that PPI is prioritised and resourced within studies. This increasing expectation has heightened the risk of researchers carrying out 'tick-box' PPI rather than 'meaningful' involvement[5]. There are many moral and ethical arguments being made for PPI. Many believe that as citizens and taxpayers, members of the public have a right to influence research that is being funded by public money[6]. PPI researchers are also making pragmatic arguments for PPI and providing anecdotal accounts about how PPI can make research more relevant, accessible and acceptable to participants[7]. The ethical arguments are often seen as sufficient regardless of any pragmatic impact. However, PPI costs time and money, therefore pragmatic claims need scrutiny[8]. More substantive evidence is needed to evaluate the potential impact of PPI on the conduct and outcomes of research[5,9]. In 2001, the need to establish if PPI leads to actual, rather than merely perceived benefits for research processes and output was identified. Over fifteen years later, this need remains.

In clinical research, the results of clinical trials have not traditionally been shared with clinical trial participants. A recent survey carried out on a large registry of health research participants, found that while 95.6% of respondents said researchers should always or sometimes offer the results to participants, only 33% of respondents actually received the results of studies in which they had participated[10]. An upcoming European Union Clinical Trial Regulation requires sponsors to provide summary results of clinical trials in a format understandable to laypersons, including participants[11]. However, there is a lack of evidence on the most appropriate methods of sharing results with participants. Uncertainty persists around what information should be shared, how results should be shared and who should be responsible for sharing the results. Since the findings of clinical research often exist in a complex context of scientific exchange and debate, it is important that the information shared is accessible and relevant to participants[12]. The increasing understanding of the importance of sharing research results with study participants is somewhat linked to a wider movement towards transparency in trials. This movement is largely promoted by initiatives such as SPIRIT, CONSORT and AllTrials. The SPIRIT Statement provides guidance to researchers to improve the completeness and quality of trial protocols[13], the Consolodated Standards of Reporting Trials (CONSORT) Statement is an evidence based, minimum set of recommendations for reporting randomised trials[14] and the AllTRials iniative calls for all past and present triasl to be registered and their full methods and summary results reported[15]. Some of these initiatives also include recommendations for disseminating results to research participants. For example, the SPIRIT statement states that study results must be released to participating physicians, referring physicians, patients and the general medical community[13].

The Thyroid Hormone Replacement for Subclinical Hypothyroidism Trial (TRUST) was a multi-centre, double blind, placebo controlled, phase III clinical trial testing the efficacy of thyroxine replacement in subclinical hypothyroidism in older community dwelling adults[16]. The results of the TRUST trial were published in the New England Journal of Medicine on 3rd of April, 2017[16]. This Study Within A Trial (SWAT) was conducted at the Irish TRUST trial site prior to and after publication of results. The aim of this SWAT was to investigate methods of disseminating trial findings to participants by using a PPI approach to identify, develop and evaluate a patient-based approach of receiving trial results.

Methods

Study design

This was a sequential mixed methods study with three phases. In this study, methods were combined for complementarity, where

each method addressed a different aspect of the study aim[17]. The first phase used a qualitative approach to identify and develop a patient-based approach to disseminating the results, the second phase used a SWAT intervention to compare the dissemination approaches and the third phase used a quantitative patient understanding questionnaire to evaluate the patient-based approach. The full study protocol has been published elsewhere[18], but a summary follows here.

Setting

The study sites for the TRUST trial were the University of Glasgow, Scotland (lead site); Leiden Academy on Vitality and Ageing, The Netherlands; Leiden University Medical Centre, The Netherlands; University of Berne, Switzerland; and University College Cork, Ireland. A total of 738 participants with subclinical hypothyroidism were recruited to the trial over a three-and-a-half year period from 2013–2017[16]. The trial completed recruitment in November 2016 and the results were published in April 2017[16].

This SWAT was conducted at the Irish TRUST site. The hub centre for the Irish TRUST site was located at the Mercy University Hospital, Cork where 38 participants were recruited. A further 77 participants were recruited from five satellite sites.

Population

As this SWAT was embedded in an ongoing clinical trial the study sample was determined by the TRUST Thyroid trial. There were 115 TRUST participants recruited in the Irish site, 11 of these participants withdrew over the course of the trial. Our study sample included all remaining TRUST participants (n=104).

Phase One: Identification and development of patient-based approach (qualitative and PPI phase)

The first phase of the study used a qualitative approach to iteratively identify and develop a patient-based approach to disseminate the results of TRUST trial. This was done in three separate stages: qualitative focus groups, a PPI group and an adult literacy review.

Focus groups

Three semi-structured focus groups were conducted with four to eight TRUST trial participants per group. All Cork-based patients (n = 38) were contacted via letter and invited to participate. A €20 shopping voucher was given to all participants to cover travel expenses. Each session was led by trained qualitative researchers (WHS, ER, CH). A topic guide was used to guide the focus groups. The topic guide was reviewed and refined by all members of the SWAT research team (see Supplementary File 1: Focus group topic guide).

The Consensus-Oriented-Decision-Making (CODM) model was used to guide the group to reach a consensus[19]. The CODM model is accepted as a flexible model for reaching decisions[19]. In this study some of the steps were initiated by the focus group facilitator and others occurred naturally as a follow on from the previous step. Below is an outline of each of the seven steps of the CODM model and how they were used in this study:

1. Framing the topic: The focus group facilitator introduced the idea of sharing results with participants and provided some context on the reasons why results are/ are not shared with participants.

2. Open discussion: The facilitator asked the group whether or not they think results should be shared with trial participants and whether or not they would like to receive the results of the TRUST trial.

3. Identifying underlying concerns: The previous discussion naturally followed on to participants asking questions and expressing concerns about the result method, content and language that would be used.

4. Collaborative proposal building: The group worked together to agree on the important elements of the results in terms of result method, content and language.

5. Choosing a direction: This step occurred naturally as part of the previous step.

6. Synthesizing a final proposal: The facilitator re-iterated the proposal the group had agreed upon and asked the group for feedback.

7. Closure: This step occurred naturally as part of the previous step.

Analysis. Focus group recordings were transcribed verbatim and entered into NVivo Version 11 for data management during thematic analysis. Braun and Clarke guidelines[20] for conducting thematic analysis were followed. Initial focus group transcripts were analysed independently by two researchers (ER and AC). Each transcript was read multiple times (data familiarisation) and initial codes were identified. These codes were then used to identify emerging themes. Both researchers discussed emerging themes and conducted further refinement. The refined themes were then discussed and agreed upon with other members of the research team (ER, CH, AC, KMS). Researchers (ER, CH, AC) then used the focus group findings to develop an initial draft of a patient-based approach for the dissemination of results (see Supplementary File 2: Draft one of patient-based result letter).

PPI group

A PPI group was established to develop and refine the content of the patient-based appproach for the dissemination of results. During the focus groups, three TRUST trial participants volunteered to take part in the PPI group. In addition to these three PPI partners, an additional partner was identified from a previous qualitative research study undertaken by the research team. This individual was keen to learn more about research and expressed an interest in being involved in future projects. While this individual had previous experience of taking part in

research (as an interview participant), she had no experience of taking part in a clinical trial or being involved as a PPI partner. Originally, we intended to conduct these sessions in a group format, due to difficulties with PPI partners' schedule commitments, one-to-one sessions were conducted. At the one-one session, a researcher (ER) and the PPI partner discussed the layout, content and language of the initial draft of the result method. Researchers and PPI partners worked together to edit different sections of the document. These discussions were not audio recorded but comprehensive field notes were taken by the researcher (ER). These notes were then collated by the researcher and used to further ensure that the results letter reflected PPI partners' perspectives and preferences.

Adult literacy review

While the PPI group had significant input into the format and language used in the patient-based approach, the research team felt that it would be of additional benefit to collaborate with the National Adult Literacy Agency (NALA) to ensure the document adhered to national "Plain English" standards. These standards ensured that the information presented to trial participants was sufficiently easy to read and understand (literacy). This would help to ensure that trial participants were able to make sound health decisions based on the information presented (health literacy)[21]. This review was an iterative process with several drafts exchanged for editing. Although the review was taken as an additional step to the published protocol for the study, the research team felt it was helpful to further ensure that the document was accessible and easy to understand.

At the end of the first phase of the study, a final draft of the patient-based result letter was approved by researchers, PPI group and adult literacy experts (see Supplementary File 3: Final draft of patient-based result letter).

Phase Two: Dissemination of trial results (intervention phase)

The second phase of the study used a SWAT intervention to disseminate the results of the TRUST Thyroid Trial to trial participants. This was done using a prospective, randomised, single blind, parallel trial design. It is important to note that when the term randomisation is used, it refers to the allocation of patients to intervention/control within the SWAT and not the TRUST Thyroid trial. Irish TRUST participants were randomised to intervention or control groups using an online random number generator. The intervention group received the patient-based letter format (see Supplementary File 3: Final version patient-based results letter) and the control group received a copy of the TRUST results press release, which was made available by the lead study site on the TRUST Thyroid Trial Website (see Supplementary File 4: Standard results letter). Participants were blinded to their intervention group. One member of the research team was un-blinded in order to perform the randomisation and distribute the results of the trial. As they were un-blinded to perform these two important tasks, they were not involved in the data analysis or interpretation in any way.

Phase Three: Evaluation of patient –based approach (quantitative phase)

The third phase of the study used a quantitative patient understanding questionnaire to evaluate the patient-based approach to disseminating trial results. The questionnaire was developed in consultation with experts in the area of subclinical hypothyroidism and scale (questionnaire) development (PK and KMS). The early development of the questionnaire was guided by a consultation document, which accompanies the EU Clinical Trials Regulation No 536/2014[22]. This document highlights the information which should be presented to trial participants in the trial summary at the end of a trial. However, initial questionnaire items were modified to allow for psychometric testing. The final questionnaire contained 12 questions; six items were measured on a five point LIKERT scale, there were four multiple-choice questions and two vignettes. The first six items measured patients' perceived understanding of results, the four multiple choice measured patients' actual understanding of results by requiring them to select the correct answer. To further test participants' understanding of the trial results, two vignettes describing two typical patient case studies of older adults with subclinical hypothyroidism were provided with a question on whether a doctor should prescribe thyroxine for the hypothetical patient described. The questionnaire was reviewed by the PPI group to assess content and face validity. It then underwent further review by NALA to ensure adherence to the national 'Plain English' standard. The final version of the questionnaire can be seen in Supplementary File 5: Patient understanding questionnaire.

The questionnaire was sent to all Irish TRUST participants (intervention and control group) one week after they received the results of the trial. A reminder questionnaire was sent to non-responders 3 weeks later.

Analysis. The primary outcome was the difference in levels of patient understanding between the intervention and control groups. This measured the impact of PPI on patient understanding of end of trial results. The psychometric properties and construct validity of the questionnaire were examined with exploratory factor analysis. Principal component analysis (PCA) was conducted on the six LIKERT scale items. Internal consistency of the questionnaire was investigated using Cronbach's alpha coefficient. Completed questionnaires were entered into SPSS software (version 24) and analysed using descriptive and inferential (Chi-square test and Fishers Exact) statistics. The researcher carrying out data input and analysis was blinded to the participants' allocation status.

Costs of conducting PPI

The lead researcher (ER) kept a detailed account of all direct costs associated with conducting PPI for the purpose of this study. These costs included researcher salary, travel and expenses for PPI participants, adult literacy review and printing and postage costs.

This paper has been written in adherence to the Guidance for Reporting Involvement of Patients and Public 2 (GRIPP 2)[23].

The GRIPP 2 checklist is a tool, developed to improve the reporting of patient and public involvement in research and guide the development of a transparent, consistent and high-quality PPI evidence base. The Good Reporting of a Mixed Methods Study (GRAMMS) framework was also used to inform the reporting of the findings[24].

Results

Characteristics of the trial participants stratified by participation in the different stages of the study are presented in Table 1.

Phase One: Identification and development of patient-based approach (qualitative and PPI phase)

Focus groups

Three focus groups were held with 19 out of 38 participants accepting an invitation to join. Participants who attended the focus groups were similar in age, gender, education level to those who did not attend.

Focus group findings indicate that participants want to receive the results of the trial in which they are taking part. Three main themes emerged in relation to participants' perspectives of and preferences for receiving trial results: 'acknowledgement of individual contribution', 'contributing for a collective benefit' and 'receiving accessible and easy to understand results'.

Acknowledgement of individual contribution

Many participants reported feeling they had made an individual contribution to the trial in terms of their time and personal information while attending the trial study visits. As such, participants felt that receiving the results of the trial would provide an acknowledgement of this individual contribution:

> *'Yes, I mean it's kind of instinctive… when you go into a [clinical trial] and you spend and invest that time in it. I mean okay I had the time to invest but you know at the end of the day, [receiving the result] is kind of like your pay off. '* (FG2 P3)

Contributing for a collective benefit

While participants spoke about making an individual contribution to the trial, they felt that their involvement contributed to a collective benefit or greater good. Participants reported that receiving the results of the trial would help them to feel that they had contributed to this greater good:

> *'I'm not really interested in my own personal results but as the results of the scheme as a whole. You know the idea is, does the study help or hinder old people and that's what I want to know'* (FG2 P1)

This feeling of contributing for a collective benefit was further reinforced when participants discussed their desire to understand how the results of the trial will be implemented by medical experts and ultimately how it will affect others who have the condition:

> *'I would like to know, if they found out, okay, do we treat these people or not. That would be good. Do we treat them or don't we treat them? I think that is what it's all about' (FG3 P4)*

Receiving accessible and easy to understand results

Participants expressed a clear need to receive the results of the trial in an accessible and easy to understand way. This preference applied to the format, language and content of the patient-based approach.

Table 1. Characteristics of trial participants stratified by participation in the different stages of the study.

	Total Irish TRUST participants (n=104)	Attended SWAT focus groups[1] (n=19) Total Sample n=38 RR[2] =50%	Randomised[3] (n=101)		Returned SWAT questionnaire (n=69) Total Sample n=101 RR[2]=68%	
			Intervention Group (n=51)	Control Group (n=50)	Intervention Group (n=38) RR= 74%	Control Group (n=31) RR=62%
Sex						
Male	61 (58.7%)	14 (73.7%)	31 (60.8%)	28 (56%)	26 (68%)	16 (52%)
Female	43 (41.3)	5 (26.3%)	20 (39.2%)	22 (44%)	12 (32%)	15 (48%)
Age						
65–74	57 (54.8%)	12 (63.1%)	32 (62.7%)	24 (48%)	25 (66%)	12 (45%)
75+	47 (45.2%)	7 (36.9%)	19 (37.3%)	26 (52%)	13 (34%)	17 (55%)
Education						
Primary only	22 (21.2%)	2 (10.5%)	12 (23.6%)	9 (18%)	10 (26%)	8 (26%)
Secondary/Tertiary	47 (45.1%)	12 (63.2%)	24 (47.1%)	22 (44%)	19 (50%)	11 (35%)
Unknown	35 (33.7%)	5 (26.3%)	15 (29.3%)	19 (38%)	9 (24%)	12 (39%)

[1]A subgroup of Irish TRUST participants (n=38) were invited to focus groups.

[2]RR=Response Rate

[3]Total Irish TRUST participants (n=104) excluding PPI partners (n=3)= n=101.

The majority of participants said they would like to receive the results in a letter format posted to them directly from the TRUST trial. Participants felt that this method would be accessible to them as they could read the results *'in text'* (FG3 P4) and keep a *'hardcopy'* (FG P1). While participants wanted an official statement of the results in a letter format, they also felt it was important to add a personal element to the letter. They suggested this could be done by offering participants a phone number that they could call if they wished to discuss any further issues or concerns with the TRUST study team:

> *'Could you attach a helpline on to it? If you know, somebody had some kind of serious medical question or that they thought was a bit personal element or whatever. That they'd like to talk to a medical person or whatever. Instead of just talking to your GP, maybe that would add another dimension of care around the TRUST'* (FG2 P3)

Participants agreed that the format, content and language of the results letter needed to be easy to read and understand. All participants wanted the letter to be no longer than 2–3 pages and presented in a question and answer format. Participants believed the content of the results letter should include *'pertinent information'* (FG1 P7) relating to the trial itself, the study drug (including side effects) and the results of the trial. They stressed the importance that this information needed to be informed by medical experts and *'from a good authoritative source'* (FG2 P2) but it should be presented to them in a language that fits their current context and could be easily understood by those who do not have scientific or medical backgrounds.

> *'Just in ordinary language that we can understand ourselves, you know that we don't want big and long explanation or that, just that we can pick it up straight away that it's without any huge number of pages. Just the bare, to me anyway, answers to the questions.'* (FG3- P2)

It was evident from the focus groups that participants want to receive the results of the trial both to acknowledge their individual contribution to the trial and also help them to feel that they had contributed to a greater good. Participants expressed a clear preference to receive the results in an accessible and easy to understand way. These results were used by the researcher (ER) to develop an initial draft of the results letter (see Supplementary File 2: Draft one patient-based result letter).

PPI group

The initial draft of the results letter was then further iteratively developed by the PPI group. There were four PPI partners in total (three trial participants and one older adult) Each partner toook part in one-to-one session. Each session contained an open discussion between the researcher (ER) and PPI partners on the layout, content and language of the document. Researchers and PPI partners worked together to write, re-write, edit and change different sections of the document.

Health literacy review

This draft was then iteratively reviewed and approved by health literacy experts from the NALA (see Supplementary File 3: Final version patient-based results letter).

Phase Two: Dissemination of trial results (intervention phase)

There were a total of 101 Irish TRUST participants randomised to the SWAT intervention. Trial participants from the PPI group (n = 3) were excluded from randomisation as they reviewed the content of the intervention method prior to the intervention. The intervention group (n=51) received the patient-based letter format (see Supplementary File 3: Final version patient-based results letter) and the control group (n=50) received a copy of the TRUST results press release, which was made available by the lead study site on the TRUST Thyroid Trial Website (see Supplementary File 4: Standard results letter).

Phase Three: Evaluation of patient-based approach (quantitative phase)

The overall response rate for the patient understanding questionnaire was 68% (69/101). The response rate for the intervention group was 74% (38/51) and the response rate for the control group was 62% (31/50). There were no significant differences in age, gender and education between those who returned the questionnaire and those who did not.

Post hoc power calculations showed that the study was underpowered to detect an effect. Power for each of the patient understanding components ranged from .01 to. 58.

Table 2 below shows the results of patients' perceived understanding of the purpose and context of the TRUST Thyroid Trial. Due to low participant numbers across the five Likert responses, the questionnaire response bands have been contracted from 'Strongly Agree' and 'Agree' to 'Yes, 'Strongly Disagree' and 'Disagree' to 'No' and 'Neither agree nor disagree' to 'Neutral'. The results show that patients' perceptions of understanding are similar between the intervention and control groups. Subgroup analysis showed patient's understanding was not significantly impacted by age, gender or educational level.

Figure 1 shows patients' actual understanding of the primary aim, side effect and results of the TRUST Thyroid Trial. Almost 82% (n=31) of the intervention group and 65% (n=20) of the control group correctly understood the primary aim of the TRUST trial (p=0.108). Almost 40% (n=15) of the intervention group and 36% (n=9) of the control group correctly understood the associated side effects of the active drug (p=0.734). In total 50% of the intervention group (n=19) and 58% of the control group correctly understood the results of the trial (p=0.504). There were no differences in patient understanding of trial results between the intervention and control groups.

In terms of patient understanding of hypothetical patient case studies, 43% (n=13) of the intervention group gave the correct answer to Vignette A; this was lower than the control group (62.1%, n=18, p=0.15). In total 77% (n=23) of the intervention group gave the correct answer to Vignette B, this was higher than the control group (66%, n=19, p=0.344).

Psychometric testing

An exploratory principal components analysis (PCA) was conducted on the patient understanding questionnaire to determine

Table 2. Patient perceptions of understanding presented by group[1].

Item	Group	Yes	No	Neutral	p-value
I understand why the TRUST Thyroid Trial took place.	Intervention (n=38)	37 (97.4%)	1 (2.6%)	0 (0%)	0.584
	Control (n=31)	29 (93.5%)	2 (6.5%)	0 (0%)	
I understand why I was invited to the TRUST Thyroid Trial	Intervention (n=38)	38 (100%)	0 (0%)	0 (0%)	0.198
	Control (n=31)	29 (93.5%)	2 (6.5%)	0 (0%)	
I know why the medicine Levothyroxine is used to treat subclinical hypothyroidism	Intervention (n=38)	32 (84.2%)	2 (5.3%)	4 (10.5%)	0.893
	Control (n=31)	25 (80.6%)	3 (9.7%)	3 (9.7%)	
I am aware of the side effects of Levothyroxine	Intervention (n=38)	30 (78.9%)	5 (13.2%)	3 (7.9%)	0.090
	Control (n=31)	17 (54.8%)	7 (22.6%)	7 (22.6%)	
I understand the impact of Levothyroxine on thyroid specific quality of life	Intervention (n=38)	31 (81.6%)	5 (13.2%)	2 (5.3%)	0.281
	Control (n=31)	20 (64.5%)	7 (22.6%)	4 (12.9%)	
I understand how doctors will use the results of the TRUST Thyroid trial to treat people with subclinical hypothyroidism	Intervention (n=38)	33 (86.8%)	2 (5.3%)	3 (7.9%)	0.878
	Control (n=31)	26 (83.9%)	3 (9.7%)	2 (6.5%)	

[1]Patient perceptions of understanding were assessed using a five point LIKERT scale.

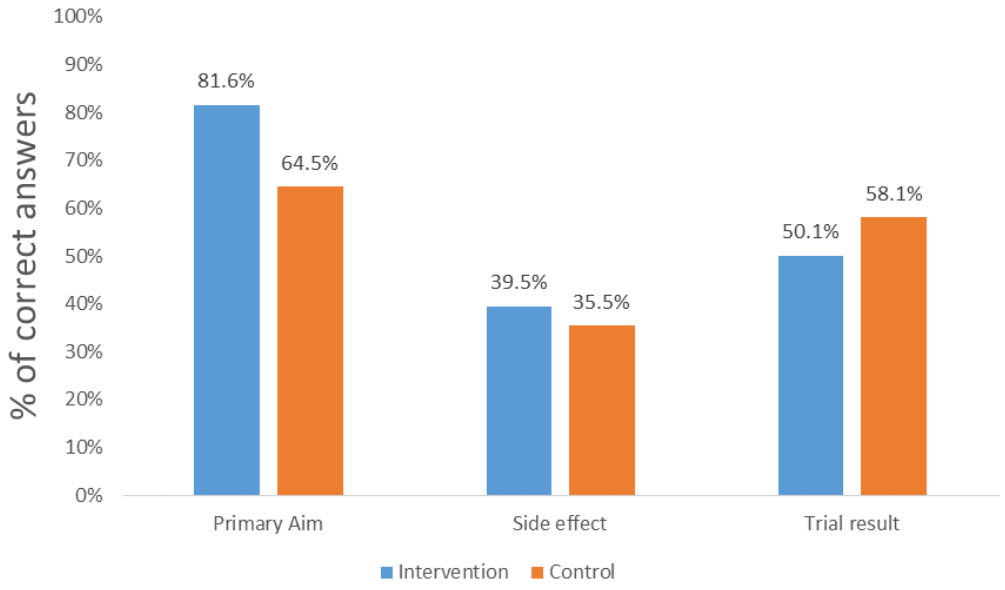

Figure 1. Patient understanding of primary aim, side effect and trial result of the TRUST Thyroid Trial presented by group[1].

[1]Patient understanding of primary aim, side effect and trial result was assessed using multiple choice questions.

its usefulness as a measure of perceived understanding. The Kaiser-Meyer-Olkin (KMO) measure verified the sampling adequacy for the analysis, KMO= .83. Bartlett's test of sphericity indicated that the correlation matrix was significantly different from an identity matrix, X2 (.852) = 283.92, p<.001. An examination of eigenvalues greater than Kaiser's criterion of one, suggested the extraction of one factor; this was supported by inspection of Cattell's scree plot. An examination of the constituent items for this factor structure also indicated that items loaded most highly on a single factor. This single factor represents a measure of perceived understanding of trial results. PCA was then conducted using an oblique (direct oblimin) rotation, specifying the extraction of one factor. This model explained a combined 69.58% of the variance in patients understanding of the TRUST thyroid trial.

Cost of conducting PPI

The total cost of this study amounted to €8,049 (see Supplementary File 6: Costs of conducting PPI).

Discussion

While PPI is increasingly recognised as an important element of clinical research, evidence on optimal methods and potential impact is lacking[4,9]. Previous research conducted on the impact of PPI has largely focused on the experiences of participants and researchers[25] and on the research process in broad terms[26]. In this study, our primary outcome was specific: a quantitative measure of patient understanding of trial results between those who received the patient-based approach and the standard approach. To our knowledge there has been no previous research conducted on the impact of PPI on patient understanding of trial results.

The involvement of clinical trial participants in this study offered insightful perspectives on the information needs of the study population in terms of receiving end of trial results. Study findings show that trial participants want to receive the results of the clinical trial in which they had participated. This is supported by much of the available literature on patients' preferences of receiving results, with up to 90% of participants in previous studies reporting a desire to receive results[27]. Focus group findings showed that participants felt that receiving results would provide an acknowledgement of their individual contribution to the trial. This finding complements previous commentaries about result sharing being an 'ethical imperative or 'moral obligation'. Fernandez et al. points out that many participants place their trust in science and researchers owe a debt to participants to fulfil their trust and recognise their altruism[12,28].

Unsurprisingly, findings also show that participants want to receive results that are accessible and easy to understand. In this study, the preferred format of receiving results was a letter posted to them directly from the TRUST trial. This preference is also consistent with the literature on patient preferences of receiving results. A previous study investigating prefrences of individuals taking part in a cardiac rehabilitation trial found that 80% of trial participants preferred to receive the results by post[29]. The patient-based approach identified in this study was feasible for researchers to develop with significant involvement from trial participants and adult literacy experts.

Previous studies exploring participants' reactions found that sharing trial results with participants can cause some negative impacts such as anxiety, anger, guilt, upset and confusion[30–32]. As far as researchers in this study are aware, providing results did not cause any negative impacts. This may have been due to the fact that the TRUST trial had a low risk of morbidity or mortality compared to some of the other studies citing negative impacts. Both result methods contained the telephone number, email address and postal address of the research team and participants were urged to contact should they have any questions or concerns relating to the study. The research team did not receive any queries.

Previous systematic reviews highlight the lack of evidence on economic analysis of PPI and call for researchers to consider the costs of its implementation[26,33]. As discussed previously research funders are increasingly demanding that PPI be carried out in research. However, the costs of PPI are often underestimated and can cause a significant financial burden on research project budgets[26,33–35]. It is extremely important that researchers plan PPI at the grant proposal stage and estimate the costs appropriately. If these costs are not correctly estimated during the initial stages of developing research proposals, they may cause a financial burden on PPI partners.

Participants in this study were not paid for their time but were provided with a €20 voucher to cover travel expenses. When PPI is not the primary focus of a study, researchers do not consider the cost implications at the beginning of the study and are often tied with limited resources to carry out PPI[34–36]. INVOLVE, the national advisory group supporting active public involvement in health services, public health and social care research in the UK, have recommended that PPI partners should be paid for their involvement[37]. Despite this, existing research suggests that institutional difficulties make negotiating the mechanisms of paying participants very difficult[34]. One study reported that in order for participants to be remunerated for their efforts, they needed to be registered as employees, a process that incurred much paperwork and time delays[34]. This study outlines the cost of conducting PPI and includes a full breakdown of costs (see Supplementary File 6: Costs of conducting PPI). This breakdown provides a template to other researchers who plan to carry out and evaluate PPI as part of their research. It is important to note that not all costs associated with carrying out the study were included in this amount. For example, the only salary costed was that of the research assistant. The expertise provided by other members of the study team were not included in the total cost as they were being paid by the University or other research grants. The total cost of conducting this study was €8,049 which is not insignificant but should be considered in the context of the cost of large-scale trials.

Strengths and Limitations of the study

While this study provides important insights into patients' preferences of receiving trial results, it is not without limitations. Firstly,

existing PPI literature states that 'to understand the research needs and challenges, PPI has to engage people who are able to offer perspectives from the study population'[3]. All PPI partners in this study were active members of the research community as they had taken part in the TRUST trial and had agreed to long-term follow up. This is a strength of the SWAT as they were able to offer perspectives from the study population, however it does have an important implication for their reporting of understanding the results of the trial. They may be more inclined to rate their understanding as high because of their investment in the trial[38], thus potentially minimising differences between the intervention and control conditions and minimising inferences that can be drawn about the intervention. Previous research suggests that people that actively choose to engage in research either as research participants or involvement partners are more likely to be middle-class and highly educated[39,40]. In this study, those that attended the focus groups and PPI group were similar in education level to those that did not attend. This is not surprising considering the entire study sample had already actively volunteered to take part in the TRUST trial.

Secondly, the results of the patient understanding questionnaire show that the levels of patient understanding were similar between the two groups. However, this study was underpowered to detect an effect. As this was a Study Within A Trial (SWAT), the power was limited by the sample size that was available to us from the trial (n=115). Furthermore, validation of the patient understanding questionnaire was limited by the sample size in this study. While validation of the questionnaire was limited, exploratory factor analysis provided some evidence that the questionnaire is a useful tool for measuring patient understanding of trial results. The developed questionnaire can be tailored for use in other trials in future examinations of patients understanding of trial results. This would provide insight into patient understanding and provide further validation data.

Thirdly, all SWATparticipants were aged 65 and over. The layout, format and language of this patient-based approach which was identified and developed may only be relevant for this study population. Other trial populations may prefer to receive the results via email, online or in person from a member of the study team[12]. The evidence on patient preferences of receiving trial results is limited, therefore further research is needed to explore patient preferences of receiving trial results amongst different study populations.

It is also important to point out that the control group in this study received a copy of the trial results in a press release format. Most trial participants do not receive this. While this control method was a step further than normal procedure, the researchers in this study felt this was appropriate. The information presented in the press release was similar to that of the patient-based approach. However, the format and layout of the press release was different. Information was writtern in four long paragraphs separated by individual headings. It was also much shorter (1 page in total) that the patient-based approach (3 pages in total). Given the fact that press releases are written by public relations professionals with a view to communicating effectively and efficiently, this may have potentially minimised differences between the intervention and control conditions. The primary outcome of this study was assessing the impact of PPI on patient understanding of results, however, this was not the only potential impact. In hindsight, we adopted a limited approach to PPI in this study as we did not involve our PPI partners from the outset of the SWAT. Involving PPI partners in the development of core outcome sets for this SWAT could have identified other more appropriate primary outcome measures[41].

The aim of this SWAT was to investigate methods of disseminating trial findings to trial participants by using a PPI approach to identify, develop and evaluate a patient-based method of receiving trial results. The PPI approach actively involved focus group participants in making decisions about the result method and worked with PPI partners to co-develop the result letter. However, PPI partners were not involved in other aspects of the research process such as research design, data collection or analysis. This is partly due to the fact that PPI is a relatively new concept in clinical trials. As the majority of the literature has only been published in the last 12 months, there is little evidence available on the impact of PPI and no gold standard or comprehensive guidelines for researchers to follow[29]. Thornton[2] suggests that in order for PPI to develop it is important to record its social and cultural history by collecting comprehensive databases and undertaking ongoing reviews of the impact of PPI. This paper along with the study protocol have been written in adherence with the Guidelines for Reporting Involvement of Patients and the Public[23], thus providing templates for involving patients and the public in clinical trial design and development. This study is an important step forwards in documenting the process of conducting PPI as part of a SWAT and evaluating its impact. Future research is needed to further develop PPI in clinical trial settings. As there is currently no gold standard or comprehensive guidelines for researchers to follow when evaluating the impact of PPI, further research is needed. This research should involve PPI partners in the development of core outcome sets for evaluating PPI impact. These would significantly enhance the literature in the area.

Conclusion

Patient and Public Involvement (PPI) is advocated for every step of the trial process. We have demonstrated that it is feasible to involve PPI partners in the development of dissemination materials. Sharing clinical trial results with participants in a format understandable to laypersons will soon be a legal requirement[11]. However, there is a significant lack of evidence as to the most appropriate methods of sharing results with participants. The study identified and developed a patient-based approach to disseminating clinical trial results for trial participants. Although, in this study PPI did not influence patients' final understanding of results, it documents the process of conducting PPI within the clinical trial setting. This process may be useful for other trialists interested in conducting and evaluating the impact of PPI in clinical trials.

Ethics approval and consent to participate

The research was approved in Ireland by the Clinical Research Ethics Committee of the Cork Teaching Hospitals, UCC, Ref ECM 4 (t).

All participants provided signed informed consent to take part in the study.

Data availability

The raw data from this study cannot be sufficiently de-identified, and therefore are not publicly available. However, the data from the current study are available for further (collaborative) research purposes on reasonable request. Available datasets include transcripts from focus groups, field notes from PPI sessions and responses from the patient understanding questionnaire. To access the data, please contact the corresponding author (emmy.racine@ucc.ie) or the Principal Investigator (patricia.kearney@ucc.ie). Researchers must provide a written proposal on how the data will be used in research before access is granted.

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Supplementary material

All supplementary files are contained in one PDF document.

Click here to access the data.

Supplementary File 1: Focus group topic guide

Supplementary File 2: Draft One of Patient-Based Result Letter

Supplementary File 3: Final Draft of Patient-Based Result Letter

Supplementary File 4: Standard Results Letter

Supplementary File 5: Patient Understanding Questionnaire

Supplementary File 6: Costs of Conducting PPI

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