



EUROPE

Patient and public involvement and engagement in research

Enabling meaningful contributions

Sarah Ball, Amelia Harshfield, Asha Carpenter, Adam Bertscher and Sonja Marjanovic



This research was commissioned by The Healthcare Improvement Studies Institute (THIS Institute)

www.thisinstitute.cam.ac.uk

For more information on this publication, visit www.rand.org/t/RR2678

Published by the RAND Corporation, Santa Monica, Calif., and Cambridge, UK

© Copyright 2019 RAND Corporation

RAND® is a registered trademark.

RAND Europe is a not-for-profit research organisation that helps to improve policy and decision making through research and analysis. RAND's publications do not necessarily reflect the opinions of its research clients and sponsors.

Limited Print and Electronic Distribution Rights

This document and trademark(s) contained herein are protected by law. This representation of RAND intellectual property is provided for noncommercial use only. Unauthorized posting of this publication online is prohibited. Permission is given to duplicate this document for personal use only, as long as it is unaltered and complete. Permission is required from RAND to reproduce, or reuse in another form, any of its research documents for commercial use. For information on reprint and linking permissions, please visit www.rand.org/pubs/permissions.

Support RAND

Make a tax-deductible charitable contribution at www.rand.org/giving/contribute

www.rand.org

www.randeurope.org

Preface

In recent years, we have seen an expansion of patient and public involvement (PPI) activities in research. This has been accompanied by a growing interest in understanding how PPI can best be mobilised and enabled, and in how it can contribute positively to the research process and to impacts from it. The body of literature on these issues has expanded rapidly in the last decade. However, the evidence base on what works, how and why remains fragmented and inconclusive.

RAND Europe was commissioned by The Healthcare Improvement Studies (THIS) Institute at the University of Cambridge to conduct a rapid review of the evidence base on PPI in research. This report reflects on what we know and on knowledge gaps. It aims to help inform THIS Institute's efforts to establish and implement an effective PPI strategy. It should also be of relevance to other organisations and initiatives seeking to involve patients and the public in research in a meaningful and effective way.

Section 1 provides background and context to the research and outlines the research aims, study design and methods. Section 2 briefly summarises the profile of the reviewed literature. Section 3 considers what motivates

patients and the public to contribute to research. In Section 4, we discuss how patients and the public contribute to research, reflecting a diverse set of approaches, and in Section 5 we consider how contributions can be enabled. Section 6 overviews key challenges and barriers. In Section 7, we discuss what is known about the impact of PPI in research and how this can be evaluated. Finally, in Section 8 we reflect on the learning from the research and set out some areas for THIS Institute and other organisations to consider when developing PPI strategies. Appendix A provides case-based examples of PPI in research, illustrating key insights from our review in practice. Appendix B provides further detail on the study methodology.

RAND Europe is a not-for-profit policy research organisation which aims to improve policy and decision making through research and analysis. For more information about this document, please contact Dr Sonja Marjanovic (senior and corresponding author):

Dr Sonja Marjanovic
Senior Research Leader
RAND Europe
smarjano@rand.org
+44 (0)1223 353329

Summary

There is a need for better evidence on how to effectively support patient and public involvement and engagement in research.

RAND Europe was commissioned by The Healthcare Improvement Studies (THIS) Institute to conduct a rapid review of the evidence base on patient and public involvement (PPI) in research, in order to find out what is known in this area and where there are gaps. The review aims to help inform THIS Institute's efforts to establish and implement an effective PPI strategy. It should also be of relevance to other organisations and initiatives seeking to involve patients and the public in research in a meaningful and effective way.

Based on a rapid evidence assessment and interviews with experts, the report examines why and how patients and the public get involved with research, what enables meaningful involvement, associated challenges and potential enabling mechanisms, the impact of PPI, and the evaluation of this activity. Based on these insights, we provide a series of recommendations for THIS Institute and other organisations to inform strategies for engaging patients and the public. Throughout our report, we use the words involvement and engagement interchangeably, reflecting the general practice in the literature. However, we recognise the need for the terminology

related to public and patient contributions to be clarified in the wider knowledge base.

This review is primarily concerned with *active* PPI in healthcare research. Active involvement entails, for example, helping shape research questions or priorities, or contributing to data gathering, analysis or interpretation. Considering patients and the public as study participants (e.g. participants in trials or interviewees for a study as part of the research methods) does not qualify as active involvement.

What motivates patients and the public to engage with research?

Patients and the public engage with research for a variety of reasons spanning: (i) interest in a healthcare topic, often driven by personal experience of a disease or of the health service; (ii) altruistic motivations to contribute to a better healthcare system through research; (iii) a desire to influence and reflect patient perspectives in research; and (iv) a more general interest in research activity and in contributing to scientific knowledge.

Why do researchers involve patients and the public in their studies?

Researchers involve patients and the public in their studies for various reasons. Among

these are *pragmatic considerations*, whereby involvement is identified as a condition of research funding, as a response to a policy drive to share power and control in research between researchers and the wider public, or as a practical way of helping with recruitment and retention of study participants. Other reasons for involving patients and the public include drivers *related to values, experiences and perceptions*, such as a belief that it is morally the “right thing to do” or that involvement will improve the quality or relevance of research outputs, or a positive prior experience.

How are patients and the public involved in research?

PPI in health research can take place across different stages of a research cycle, for example through: (i) contribution to priority-setting exercises or specification of research questions; (ii) developing or reviewing funding applications; (iii) helping design studies; (iv) assisting with recruitment of study participants; (v) engaging with data collection and data analysis; (vi) facilitating dissemination and research uptake; or (vii) contribution to evaluation activities. Engagement can take place at the level of an individual project, a portfolio of projects or at the organisational level (for example as a part of advisory or governance structures). The duration, frequency or regularity of patient and public engagement can range from ad hoc, task-based contributions to long-term engagement across the lifetime of a project or research organisation. More detail on the diverse types of patient involvement and the different roles that patients and the public can play in research efforts is provided in Section 4 of the report.

What are the challenges to patient and public involvement in research and how can they be addressed so that contributions are effectively enabled and rewarded?

A range of challenges to effective PPI exist in the research system. However, the growing focus on and increased commitment to PPI within research over the past decade has also revealed some key enabling mechanisms and rewards. These are outlined in Table 1. The rewards for engagement that matter most to patients and the public include feedback on their contributions and on project progress and impacts; acknowledgment and recognition of contributions; learning and personal development opportunities; social rewards such as new social relationships and networks; and financial payments, compensation and rewards for contribution (e.g. vouchers).

What is the impact of engaging patients and the public in healthcare research?

The evidence base on the impact of PPI in research is piecemeal and inconclusive, with many studies reporting hypothesised and perceived impacts, rather than robust evidence of impact. The core categories of potential or realised impact span impacts on individuals, on the quality and relevance of research projects and on the wider research system. *Impacts on individuals* can include the empowerment of patient and public contributors through learning new skills, accessing new knowledge and influencing research activities, or enabling researchers to better understand a research area from a public perspective. PPI can also impact on *the quality of research studies and their relevance for patients and the health service*. This can happen through: influence on research priorities; helping solve ethical

Table 1: Challenges to and enablers of effective patient and public involvement

Driver of effective involvement	Challenges	Enabling mechanisms
Governance, management and infrastructure	<ul style="list-style-type: none"> • Inappropriate financial resourcing for PPI. • Poor reporting on PPI processes and limited monitoring and evaluation. • Little coordination and shared learning between different PPI bodies. • Lack of a common definition of PPI leading to confusion about what it entails. • Limited patient and public awareness about involvement opportunities. • Recruitment and retention strategies based on insufficient understanding of what motivates PPI contributors. • Insufficient clarity on PPI contributor roles. • Lack of in-built feedback mechanisms. • Administrative challenges related to limited support capacity, administration costs and/or system bureaucracy. • Mandating PPI in the absence of evaluation, leading to tokenism. 	<ul style="list-style-type: none"> • Ensuring sufficient resources for PPI activities. • Effective management of financial reimbursement or upfront payment for PPI contributors. • Ensuring clearly specified goals of PPI in projects and clarity in PPI contributor roles. • Establishing mechanisms to involve patients early on in research design to build a sense of shared ownership. • Mechanisms to nurture relationships throughout a project or initiative. • Flexibility in timing of engagement and in how contributions can be made. • User-friendly online platforms (e.g. well-designed, relevant and engaging online websites and portals).
Individual and organisational capacity	<ul style="list-style-type: none"> • Scientific language and jargon posing challenges to effective communication. • Lack of experience, knowledge, skills or confidence among PPI contributors. • Risks of initiatives over-professionalising PPI and losing lay perspectives. • Challenges to representativeness of contributors. • Challenges relating to individual health or wellbeing (e.g. inability to travel). 	<ul style="list-style-type: none"> • Training and support for PPI contributors on how to effectively engage with research (e.g. training on a topic area or the language and process of research). • Training for researchers on how to conduct effective PPI. • Designated leadership for PPI within research organisations. • Individuals feeling well enough to engage through the required mechanisms (and flexible and accommodating engagement mechanisms).
Culture, attitudes and behaviours	<ul style="list-style-type: none"> • Tokenism impeding the implementation of articulated PPI strategies and approaches and their integrity. • Dismissive attitudes of some researchers creating a barrier to effective relationships. • Public and patient reservations about their ability to influence. • Challenges for researchers in managing expectations of PPI partners (e.g. about roles and goals). • Managing group dynamics (e.g. power-dynamics between researchers and PPI contributors, and between individual members of PPI groups). 	<ul style="list-style-type: none"> • Receptive researcher attitudes to PPI. • PPI contributor openness to views other than their own. • Investment in collaboration and co-learning. • A commitment to providing feedback on how an individual has impacted on research, study progress, results and impacts. • Acknowledgment and recognition of contribution and rewards (e.g. vouchers). • A commitment to learning through PPI evaluation. • Mechanisms to enable connected PPI contributor communities.

dilemmas; helping with recruitment strategies for research studies; influencing how data is collected, analysed and interpreted to ensure a patient and public perspective; and ensuring communication and dissemination of outputs in a language and format that is accessible to patients and the public. Finally, there can also be *impacts on the wider research system*, for example through impact on accountability for resource use, access to research funding, and the alignment of research with perceived moral obligations, values and norms.

Although most studies consider desired impacts, there are also some unintended consequences that researchers can try to mitigate against. For example, tokenism and dismissive attitudes can hinder the ability to implement an effective and meaningful PPI strategy. This may have a disenfranchising effect on patients who may not feel listened to or valued. Conversely, where a researcher feels mandated to involve patients and the public even when they do not see the value of involvement, this can demotivate researchers and accentuate tokenistic practice.

Recommendations

As an overarching principle, when designing a patient and public engagement strategy, it is important for organisations and initiatives to ensure that PPI contributions are relevant and meaningful for the research supported and for the stakeholders involved. This means avoiding the risks that mandating PPI or pursuing it in a generic, tick-box fashion could pose to effective engagement. It also means ensuring that an organisation's overarching values and principles are reflected in how it designs and implements the engagement strategy. Stakeholders we consulted for this research emphasised that meaningful involvement does not mean involvement in everything.

To inform strategies for engaging patients and the public in healthcare research, we offer the following recommendations:

1. **Think carefully about who to involve and why.** Finding the right contributors is key for achieving desired impacts from PPI input. The types of individuals and types of engagement required are likely to vary across projects, across different tasks within a project, and across different types of organisational activities. Work towards achieving diversity in the types of individuals involved.
2. **Ensure that the roles of PPI contributors are clear from the outset, communicated in accessible language, and that there is a shared understanding and buy-in.** The mutual management of expectations between researchers and patient and public contributors is key to effective engagement.
3. **Ensure that PPI contributors are well informed and supported to effectively engage.** This includes: (i) providing sufficient background information about a project and contributors' roles in an accessible form; (ii) committing to transparency in the goals and expected outcomes of both the project and of PPI contributors' engagement; (iii) carefully planning for PPI contributors' induction into a project; and (iv) ensuring contributors receive training if needed.
4. **Think about ethical considerations beyond informed consent and formal ethical approval process requirements.** This includes considering realistic timeframes for involvement, how PPI contributions will be acknowledged and recognised, and how research opportunities and outputs will be made accessible.

5. **Build in monitoring and evaluation mechanisms to learn from experience and inform future PPI activities.** Establishing an evaluation plan for PPI activity at the outset of each research programme is important for an adaptive initiative that promotes ongoing learning. Such a plan should highlight the desired impact from PPI activities, contributors' roles and the process for engaging them, and the methodology to be used in the evaluation. The evaluation plan for each project should make clear whether the focus is on the quality of the PPI process, on its outcomes and impacts, or both.
6. **The publications stemming from projects should also report on the methods used to engage patients and the public (who was involved, how) and on the outcomes of involvement.** This can help ensure that that PPI approaches are replicable in future studies and also contribute to the wider evidence base on the impacts of PPI in research.
7. **Design efforts to recruit and retain patient and public contributors in a way that reflects the diverse factors which motivate people to engage with research.** The communication approach needs to explain the work of an organisation and the engagement opportunity in a way that people can relate to and find compelling and exciting, using accessible language.
8. **Consider the mix of approaches that will allow for effective awareness raising and recruitment, within the resources available.** Consider the appropriate mix of online and offline mechanisms for advertising opportunities and targeting individuals or organisations. Also consider the extent to which there is a need to directly engage with individuals, organisations and communities, as well as the extent to which this requires the involvement of intermediary organisations and system stewards.
9. **Enable engagement through a mix of levers.** This mix of levers should: facilitate an appropriate information environment and support required skills and competencies; ensure appropriate management, governance and administrative arrangements; nurture the requisite infrastructure for engagement; and help ensure that organisational values and norms are reflected in the behaviours and attitudes of both researchers and PPI contributors.

Table of contents

Preface	i
Summary	iii
Table of contents	ix
List of tables	xi
List of boxes	xii
Acronyms and abbreviations	xiii
Acknowledgements	xv
1. Background and context	1
1.1. <i>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</i>	1
1.2. <i>This report considers how meaningful and effective patient and public engagement with research can be achieved</i>	2
2. Profile of the reviewed literature	7
3. Why does patient and public involvement in research happen?	9
3.1. <i>Why do patients, carers and the public engage with research?</i>	9
3.2. <i>Why do researchers involve patients and the public in research activities?</i>	10
4. How patients and the public are involved in research: an overview of approaches and methods	11
4.1. <i>Role or level of engagement</i>	12
4.2. <i>Duration, frequency and regularity of engagement</i>	13
4.3. <i>Models of engagement</i>	13
4.4. <i>Tasks and activities engaged in and specific methods used</i>	15
5. How can effective contributions from patient and public involvement be enabled?	21
5.1. <i>How can patient and public involvement with research be enabled and rewarded?</i>	21
6. What are the challenges and barriers to patient and public involvement with research?	25
6.1. <i>Despite a growing awareness of enabling mechanisms, challenges to patient and public involvement persist</i>	25

7. The impacts of patient and public involvement in research: what we know and what we do not know	31
7.1. <i>Better evaluative evidence is needed to understand the impacts of patient and public involvement</i>	31
7.2. <i>Patient and public involvement can lead to numerous potential benefits, but can also have undesired consequences</i>	32
7.3. <i>How to evaluate patient and public engagement in research</i>	34
8. Reflection on key learning points and areas for consideration in future practice	39
8.1. <i>Meaningful involvement does not mean involvement in everything</i>	39
8.2. <i>How to prepare for effective engagement</i>	40
8.3. <i>How to raise awareness about involvement opportunities and recruit patient and public contributors</i>	43
8.4. <i>How to create an enabling environment for contributions throughout the research process</i>	45
References	49
Appendix A. Examples of patient and public involvement in practice	55
<i>Project-specific examples of patient and public involvement in research</i>	55
<i>Examples of initiatives to involve patients and the public across diverse projects and programmes of research</i>	60
Appendix B. Study design and methods	71
<i>Study design</i>	71
<i>Rapid evidence assessment</i>	71
<i>Interviews with experts on public and patient participation</i>	76

List of tables

Table 1:	Challenges to and enablers of effective patient and public involvement	v
Table 2:	Summary of inclusion and exclusion criteria: Round 1 screening	3
Table 3:	Additional inclusion and exclusion criteria: Round 2 screening	4
Table 4:	Models of patient and public involvement	14
Table 5:	PPI tasks, activities and specific methods: research preparation and design	16
Table 6:	PPI tasks, activities and specific methods: implementation	18
Table 7:	PPI tasks, activities and specific methods: dissemination, translation, uptake and evaluation	19
Table 8:	Enablers of patient and public involvement with research	21
Table 9:	Challenges to involving patients and the public in research	26
Table 10:	Potential impacts from patient and public involvement identified in the literature	32
Table 11:	Sources of guidance of relevance for evaluating PPI in research	36
Table 12:	Search terms for rapid evidence assessment – review articles	72
Table 13:	Search terms for rapid evidence assessment – original articles	73
Table 14:	Inclusion and exclusion criteria: Round 1 screening	74
Table 15:	Additional inclusion and exclusion criteria: Round 2 screening	75

List of boxes

Box 1:	Levers for an enabling PPI contribution environment	45
Box 2:	The many roles service users can play in a project: the case of collaboration in research on paramedic decision making	55
Box 3:	A case study in public involvement in mental health research	56
Box 4:	Addressing challenges and trade-offs in community-based participatory research	57
Box 5:	Understanding who engages with priority setting in child health research	58
Box 6:	Involving patients and parents in research – the importance of facilitating communities	59
Box 7:	INVOLVE	60
Box 8:	People in Research	62
Box 9:	The James Lind Alliance	62
Box 10:	An advisory group in mental health nursing research	64
Box 11:	A service user and carer panel for cancer and palliative research	66
Box 12:	Reflexivity as a mechanism for enabling patient contribution to research: using video-reflexive ethnography	67
Box 13:	Enabling patient and public involvement in research through a network approach	68
Box 14:	Enabling patient and public involvement in research through a network approach	69

Acronyms and abbreviations

AHSN	Academic Health Science Network
CLAHRCs	Collaborations for Leadership in Applied Health Research and Care
CRAG	Clinical Research Ambassador Group
CRUK	Cancer Research UK
GRIPP2	Guidance for Reporting the Involvement of Patients and the Public
HTA	Health Technology Assessment
MRC	Medical Research Council
NCRI	The National Cancer Research Institute
NIHR	National Institute for Health Research
NTCRN CRP	North Trent Cancer Research Network Consumer Research Panel
PCORI	Patient-Centered Outcomes Research Institute
PiiAF	Public Involvement Impact Assessment Framework
PLA	Participatory Learning and Action
PPI	Patient and Public Involvement
PSP	Priority-Setting Partnership
RCT	Randomised Controlled Trial
REA	Rapid Evidence Assessment
THIS Institute	The Healthcare Improvement Studies Institute
WE-ENACT	Ways of Engaging-Engagement Activity Tool

Acknowledgements

We would like to thank Joann Leeding, Dr Rebecca Simmons, Andy Paterson and Prof Mary Dixon-Woods from THIS Institute for their continued engagement with our work, and Lucy Hocking from RAND Europe for her support. We are very grateful to RAND research librarian Jody Larkin for all her help with the search protocol and process. We also thank Jon

Sussex and Marlene Altenhofer from RAND Europe for their quality assurance reviews. A particular thanks to all the individuals who shared their expertise with us during the interviews conducted for this research: Prof Roger Barker, Richard Stephens, Dr Jo Ellins, Dr Jane O'Hara, Beccy Maeso, Caroline Whiting, Jeremy Taylor and Katie Scott.

1 Background and context

1.1. 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

The Healthcare Improvement Studies (THIS) Institute is an ambitious new initiative which aims to strengthen the evidence base on how to improve the quality and safety of healthcare, and in doing so to create a large-scale, unique scientific asset for the health and care system. Supported by the Health Foundation, THIS Institute is led by the University of Cambridge (Prof Mary Dixon-Woods) and works with a diverse range of partners spanning universities and research institutes, professional bodies and patient organisations. The Institute is committed to working closely with the healthcare workforce, patients and the wider public to ensure an evidence base on improvement that is relevant to those delivering and receiving care, and to ensure actionable research outputs.

In recent years, we have seen an expansion of patient and public involvement (PPI) activities in research projects and programmes, with many research funders, medical charities, professional associations, research networks and healthcare provider organisations seeking to integrate PPI into their structure, operations, policies, professional roles and working cultures (1, 2).

Unsurprisingly, this has been accompanied by a growing interest in understanding how PPI in research can best be mobilised, enabled and supported across research, funder, policymaking, healthcare provider and voluntary sector contexts. For example, the National Institute for Health Research (NIHR) has invested significant effort to develop and strengthen a national and regional infrastructure to support PPI, including through the INVOLVE national advisory body and the Research Design Service regional centres, as well as via regional and national research networks (2). In March 2018, the Public Involvement Standards Development Partnership – which involves the NIHR, the Public Health Agency (Northern Ireland), the Chief Scientist Office (Scotland), and Health and Care Research Wales – launched a set of standards aiming to support effective PPI practices and accompanying indicators against which practices can be monitored and evaluated (3). These standards focus on inclusive opportunities, respectful and productive ways of working together, support and learning for effective engagement, the use of appropriate and accessible language and timely communications, a commitment to identifying and sharing evidence of impact, and PPI in governance and leadership of research.

An increased commitment to PPI in research is driven by two key factors (1). The first is a recognition of the potential benefits to research quality and relevance that can stem from

involving people with “lived experiences” in research. The second factor relates to moral considerations around what constitutes “the right thing to do”. However, there are also some risks and growing concerns that what is being advocated as good practice in some communities may become too far removed from feasible practices on the ground and thus detract from meaningful engagement.¹

Ensuring meaningful engagement means navigating two societal drivers, namely: (i) the opportunity for tangible benefits to the research process itself, to research outcomes and impacts, and to the experiences of stakeholders involved; and (ii) the moral imperative. Enabling meaningful engagement also calls for mitigating against undesired consequences.²

In this light, efforts to facilitate meaningful engagement of patients and the public in improvement research need to be based on an understanding of the following factors:

- What motivates patients and the public to engage with research;
- What the barriers and challenges to effective engagement might be;
- How effective involvement can be enabled, rewarded and sustained;
- What the intended and undesired impacts of involvement can be; and
- How evidence-based practice can be nurtured.

Despite the accumulation, particularly in the last ten years, of a vast body of literature on PPI (including both qualitative studies of individual initiatives and systematic reviews), there remains no single, consolidated and shared understanding about what works, why, when and how. Developing such insights is important

for informing future efforts and for facilitating impact from patient and public contributions.

1.2. This report considers how meaningful and effective patient and public engagement with research can be achieved

1.2.1. Research aims

The study examines the following questions:

1. What types of approaches and methods of engaging patients and the public in research are used in research practice (with a particular interest in research design, conduct and priority setting)?
2. What motivates patients and the public to engage with research and how can their engagement be enabled and rewarded?
3. What are the challenges to engagement and how might they be overcome?
4. What impacts (intended or unintended) can patient and public engagement have on the research process and outcomes, and how can this be evaluated?
5. Based on the above, what considerations should THIS Institute and other related organisations or initiatives bear in mind when developing an engagement strategy for PPI?

It is important to highlight that this study is primarily concerned with active PPI that helps inform a research project’s design, its implementation (for example through gathering or analysing and interpreting data) or research priority setting. The study is not focused on passive engagement where patients are just study participants (e.g. participating in trials or being interviewed for a study as part of

1 Insights from interviews conducted for this study. More detail is provided in later sections of the report, and specifically Section 8.1.

2 We recognise that not all unintended consequences are negative, as positive but unintended effects can also happen.

the research methods). An exception would be in priority setting, where the boundaries between driving the priority-setting process and participating in it are blurred. In the case of priority setting, we considered any type of patient engagement.

In our report, we use the words involvement and engagement interchangeably, reflecting the general practice in the literature. However, we recognise the need for the terminology related to public and patient contributions to be clarified, and return to this issue in Section 4.4.

1.2.2. Study design and methods

In order to answer the questions on PPI as set out above, we conducted a rapid evidence assessment (REA) of the literature. This was complemented by interviews with key informants with expertise in the field. The REA process consisted of searches of academic

and grey literature, the screening of the titles and abstracts of identified articles against inclusion and exclusion criteria, and full-text review and analysis of articles that met the specified criteria.

Due to the very large number of articles identified during the initial title and abstract screening phase, additional exclusion criteria were applied during a second round of screening, which further specified the topic focus and added some additional date restrictions. This helped ensure clear focus for the review and the coverage of diverse types of research papers (71 in total).

Table 2 and Table 3 summarise the inclusion and exclusion criteria used in the first and second rounds of screening, respectively. For more information on the methods used in this research, please see Appendix B.

Table 2: Summary of inclusion and exclusion criteria: Round 1 screening

Round 1 – Title and abstract screening	Inclusion	Exclusion
Topic	Articles which: <ul style="list-style-type: none"> • Focus on the topic of methods or approaches for the engagement of patients or the public in the prioritisation, design or conduct of health research (or evaluations of healthcare or improvement interventions); • Describe challenges and enablers to patient or public engagement; • Provide insights on the impact of patient or public engagement, including insights on advantages and disadvantages; • Provide insights on the evaluation of patient or public engagement. 	Articles which: <ul style="list-style-type: none"> • Report patient or public involvement only as participants in research (rather than actively engagement in the process of informing design, conduct or priority setting); • Report on research outside the health sphere; • Focus on PPI involvement in priority setting for health services (not research) or service design; • Focus on patient or public involvement in healthcare service decision making.
Language	English	Any language other than English
Country setting	Any country	None
Document type	<ul style="list-style-type: none"> • Any type of publication (including commentaries, editorials or opinion pieces) where the assertions are based on empirical evidence or practical experience. 	<ul style="list-style-type: none"> • Commentaries, editorials or opinion pieces without direct reference to empirical evidence or practical experience. • Conference abstracts.
Date of publication	From 2000	Before 2000

Table 3: Additional inclusion and exclusion criteria: Round 2 screening

Round 2 – Title and abstract screening	Inclusion	Exclusion
Topic	As for Round 1	Any articles which: <ul style="list-style-type: none"> • Focus on community engagement in a public health or health promotion setting; • Focus <i>solely</i> on the history and philosophy of PPI or the ethics of PPI, or on the quality of PPI reporting only (we did include articles which cover these issues in addition to the core topics of our focus). Review articles which: <ul style="list-style-type: none"> • Focus on biomedical research or on drug or medical device development; • Focus on specific conditions (but include case studies where relevant).
Language	English	Any language other than English
Country setting	Any country	None
Document type	<ul style="list-style-type: none"> • Any type of publication (including commentaries, editorials or opinion pieces) where the assertions are based on empirical evidence or practical experience. 	<ul style="list-style-type: none"> • Commentaries, editorials or opinion pieces without direct reference to empirical evidence or practical experience. • Conference abstracts.
Date of publication	<ul style="list-style-type: none"> • From 2008 for review articles (10 years). • From 2013 for primary studies (5 years). 	<ul style="list-style-type: none"> • Before 2008 for review articles (10 years). • Before 2013 for primary studies (5 years).

The research papers we discuss in this report fall into two categories: core papers describing research on the topic of public and patient involvement in health research more generally, and “case” papers that focus on public and patient involvement in the context of a particular project or organisation. The analyses presented in the core body of this report draw on both types of papers. Appendix A highlights individual case-based examples, illustrating key insights from our review in practice.

The insights from the literature review were complemented with interviews with key experts on the topic of PPI in research, who have agreed to be named in this report.³ They are:

- Prof Roger Barker – Professor, University of Cambridge
- Dr Jo Ellins – Health Services Management Centre Senior Fellow, University of Birmingham
- Dr Jane O’Hara – Senior Research Fellow, Bradford Institute for Health Research
- Beccy Maseo – Senior Research Manager, James Lind Alliance
- Katie Scott – Senior Patient Involvement Manager, Cancer Research UK (CRUK)
- Richard Stephens – Consumer Lead, National Cancer Research Institute (NCRI)

- Jeremy Taylor – Chief Executive, National Voices
- Caroline Whiting – James Lind Alliance.

The topic guide for these interviews is presented in Appendix B. Throughout the report, interview evidence is referenced in the form of “Int#”; the numbers do not follow the order list presented above. Individuals interviewed were happy to be named as contributors to the research. They are anonymised in relation to specific points discussed and views expressed, for the purposes of respecting informed consent.

In interpreting the evidence from the wider literature on patient and public engagement with research and from our consultations with experts on the topic, we keep in mind THIS Institute’s interest in understanding diverse models of engagement, including how “micro contributions” of people’s time can best be stimulated – potentially (although not exclusively) via a citizen-science platform. Although citizen science is not the focus of this research report, where possible, we have considered what the evidence from the wider engagement literature implies for potential engagement opportunities via citizen science mechanisms.

1.2.3. Caveats

This research was primarily based on a rapid evidence review of the literature, complemented with expert interviews which added depth and nuance to the findings from the literature. We believe that this combination of methods has ensured a comprehensive assessment of key approaches to involving patients and the public and associated enablers and challenges. However, when interpreting the findings, it is worth bearing in mind the following caveats associated with the nature of the rapid evidence review method

adopted and with the wider evidence base on PPI in research:

- We consulted leading experts on the topic: representatives of organisations engaging patients and the public or providing guidance on how to do so effectively, individuals who are patient representatives or work in organisations representing patient views, and individuals researching the topic of PPI. However, we recognise that further consultation with a greater number of individuals could help refine some of the insights on areas for consideration in the development of PPI strategies by organisations such as THIS Institute.
- Our research reviewed a substantive body of literature from a diverse range of sources, but we cannot claim to have reviewed all documents on the topic of interest. In total, and as elaborated on in Section 2, we reviewed 71 source documents, some of which were systematic reviews. The journal articles alone referred to 3,303 articles that informed their thinking. However, as identified in the literature itself, the quality of studies on PPI in research is variable. For example:
 - Very few studies evaluate the integrity of a PPI intervention being implemented and so it is not always known whether a designed PPI approach is implemented as intended, in practice.
 - With respect to existing systematic review articles, most noted challenges in comprehensively identifying the relevant literature due to the lack of standard terminology used and lack of specific subject headings for PPI in health databases (4-6).
 - In many papers, information that was provided on the nature of PPI activities and their impacts lacked detail. Findings on the impacts of PPI were

also often based on opinion rather than detailed and evidence-based, validated measurement (7).

- The findings and conclusions of review articles can also be limited by “*non-comparative, observational, and/or non-empirical*” literature (6) (page 1,161).

Despite these caveats, we believe that the diversity of issues explored in our review and the stakeholder perspectives considered ensure a valuable resource of practical, well-informed and actionable issues to consider in developing and implementing effective PPI approaches.

2 Profile of the reviewed literature

This research builds on a review of 71 source documents. Of these, 53 were journal articles, 14 of which were reviews (1 overview of systematic reviews, 5 systematic reviews, 2 narrative reviews, 3 literature reviews with unspecified methods, 2 scoping reviews and 1 literature review following principles of critical interpretive synthesis). These covered (i.e. referred to and included in their analysis) 3,303 articles in total. Thirty-five original articles and four commentaries/opinion pieces were also included. This was complemented with 18 grey literature sources that included a range of reports, and policy and guidance documents authored by national organisations that promote and support PPI, identified via the websites of these organisations.

The types of study on which articles reported covered a range of study designs and methods, including qualitative interview studies, mixed-methods evaluations, questionnaire and consensus studies, and comparative studies (including two parallel group trials and one

cluster randomised controlled trial (RCT)). In addition, a number of articles presented conceptual or evaluation frameworks, case studies and method-development papers were also included.

Within the broad field of health research, the literature spanned health, healthcare, and clinical research, relating to a range of patient groups and clinical conditions (e.g. mental health, dementia, cancer and palliative care) and a range of research areas (e.g. patient-centred outcomes research, implementation research, community-based participatory research, clinical practice guideline development and patient safety). The majority of articles focused on the methods, approaches, challenges to and enablers of PPI or its conceptual underpinnings, and many also considered PPI in the design and conduct of research.

The findings from the literature review, along with insights from the expert interviews, are presented in the chapters that follow.

3 Why does patient and public involvement in research happen?

3.1. Why do patients, carers and the public engage with research?

Patients, carers and the wider public engage with research activities for a range of reasons broadly related to personal interests in a topic or in research, altruistic motivations, and a desire to have influence on research and healthcare.

More specifically, key motivations span the following:

- **Interest in a health topic of importance to an individual.** This is often driven by personal experience of a disease and of the health service – either directly as a patient or as a caregiver, relative or friend (8-11) (Int1, Int2, Int3, Int4, Int7). The experiences of the health service may be positive or negative, but both can motivate individuals to contribute to research (Int4).
- **Altruistic motivations.** This includes a desire to give something back to the community and help others (9, 10, 12-14) (Int1, Int2, Int3, Int7, Int8), and to contribute to a better healthcare system (15) (Int3), for example by supporting the work of charities (Int1).
- **A desire to reflect patient perspectives and experiences in the research and to have influence** (8, 10, 11) (Int2). This is related to a belief that PPI can

Patients get involved in research because they see it as a tool for improved services or improved medicines. There is no good for us to get involved if it [the research] results in loads and loads of brilliantly argued academic articles ... and researchers presenting at conferences in Chicago or Lake Lugano ... and getting loads and loads of impact factors logged up for their CV and kudos for their university ... we want to know what happens to the patients ... has that [the research] actually changed clinical practice? (Int2)

improve the relevance of research and the meaningfulness of results for service users (8) (Int2), and facilitate impact on improving health services (16) (Int2, Int4). As highlighted by an interviewee: *“Patients get involved in research because they see it as a tool for improved services or improved medicines. There is no good for us to get involved if it [the research] results in loads and loads of brilliantly argued academic articles ... and researchers presenting at conferences in Chicago or Lake Lugano*

... and getting loads and loads of impact factors logged up for their CV and kudos for their university ... we want to know what happens to the patients ... has that [the research] actually changed clinical practice?" (Int2). Forsythe et al. (2017) emphasise an interest in sharing patients' points of view on a research topic, helping decide on research questions and helping explain study results to patients and caregivers, but less interest in engaging with approaches for data collection (8). Black et al. (2018) and two of the experts we consulted for this research (Int2, Int8) also highlight personal gain from the prospect of improved care as a related motivating factor (15).

- **Interest in research activity more widely and in learning about research and contributing to scientific knowledge** (8, 9, 11, 14). Thompson et al. (2014) highlight the role that PPI in research can play in helping fill the gap created in an individual's life due to loss of work or a career as a result of disease (14).

Different individuals are motivated by a mix of common and unique factors, which will depend on personal life experiences, interests, prior experience with contributing to research and the type of contribution required (Int3). An interviewee consulted for this research also highlighted that some people may have health service or research backgrounds as well as being affected by disease, so may be predisposed and particularly motivated to contribute (Int4).

3.2. Why do researchers involve patients and the public in research activities?

Researchers' motivations for engaging patients and the public share some common elements with what motivates those patients

and members of the public. However, there are also many distinct factors that motivate researchers. The literature highlights the frequently *pragmatic* nature of incentives for researchers to integrate PPI into their projects, including such involvement where such involvement is:

- **A condition of research funding** (1, 9, 11, 17, 18);
- **A response to a policy imperative** (16) to share power and control over research between the research community and the wider public;
- **A practical way of helping with the recruitment and retention of participants for studies and for securing acceptability for the study** (11, 19).

Other drivers for researchers to engage patients and the public relate to *values, experiences and perceptions*, although these are discussed somewhat less frequently in the literature. Examples include:

- **Values**, i.e. believing it is the right thing to do and related to a moral dimension (9, 19, 20);
- **A belief that PPI will improve research outputs**, for example to optimise clinical tools developed through research (16);
- **A positive prior experience**, for example researchers' feeling that PPI improved the quality or relevance of a prior research study they were engaged in (9).

4 How patients and the public are involved in research: an overview of approaches and methods

PPI in health research takes place throughout the course of the research cycle, drawing on a variety of approaches to involvement, and diverse methods of contributing across different stages of the research process (6, 21).

The literature on PPI in research sets out a variety of frameworks identifying key phases or stages in the research process in which PPI takes place. For example, in a key systematic review on PPI in biomedical and health services research, including 202 articles, Shippee et al. (2015) highlight PPI 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 a number of 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 (6).

Among the other frameworks presented in the literature, some cover similar stages and phases to those overviewed in 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 (6). For example, Ray and Miller (2017) categorise involvement 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) (22).

Across a variety of frameworks, there is agreement that PPI can contribute across a diverse range of aspects of research, from identifying research topics to implementing a research design and supporting the uptake of research findings (16-18, 23-25). However, a number of studies report on variations in the degree to which PPI is conducted across the different stages of the research cycle. Based on findings of surveys with chief investigators involved in health research, higher levels of PPI involvement tended to be reported in the set-up and conduct of research (designing methodology and providing input to research materials) than in data collection and data analysis (9, 18) or in the dissemination and translation phase (6, 9, 17). Van Bekkum and Hilton (2014) and Shippee et al. (2015) also highlight the scarcity of evidence relating to PPI

involvement in funding decisions, and Shippee et al. identify very limited PPI in evaluation of research (6, 19).

As elaborated on in the contents below, approaches to PPI in health research vary widely. There are significant differences with respect to the role or level of PPI contributors' engagement and the duration, frequency or regularity of their engagement, as well as the model of engagement employed across different phases of research, the tasks or activities in which contributors are engaged, and the specific methods used during the course of these activities.

4.1. Role or level of engagement

At any stage of research, PPI covers a range of potential forms of engagement, from the most passive role (that of study participants) to more engaged roles (4, 6). Manafo et al. (2018a) set out a "spectrum of engagement" consisting of six levels at which patients and the public can contribute (21). These are:

- Learning/informing (i.e. questions and learning about how to get involved);
- Contributing as participants in research studies;
- Consultation (i.e. providing feedback or advice on specific research activities);
- Involvement (working directly with research teams);
- Collaboration (partnering on equal footing with researchers); and
- Leadership (leading research activities).

Recent guidance published by INVOLVE aims to distinguish between more passive contributions and active patient engagement. It focuses on the concept of co-production, emphasising that this is not just a consultation and collaboration, but rather a relationship based on a broader set of principles. These principles call for the sharing of power and joint

ownership of research, inclusion of diverse perspectives and skills, respecting and valuing the knowledge of all those involved with equal importance, reciprocity, and a commitment to relationship building (26). The types of roles held by patient and public contributors also vary and span 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, with involvement guided by researchers (9, 11).

Crocker et al. (2017) also identify a range of distinct roles that may be played by individual PPI contributors at different stages in a research study (27):

- 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); and
- The passive presence (where just the presence of a PPI contributor has an influence on how researchers think).

However, it has been highlighted that the roles played by patients and the public may vary depending on the field of enquiry. Gray-Burrows et al. (2018), for example, explored the potential roles of PPI contributors in both clinical research (focusing on patients) and implementation research (focusing on health professional behaviour) in a consensus study. They concluded that while there was

strong support for PPI roles throughout the research process in clinical research, the role for PPI in the design and management of implementation research was less well supported (24).

4.2. Duration, frequency and regularity of engagement

The duration, frequency and regularity of patient and public engagement can vary across research projects and programmes (11, 21). For example, drawing on insights from ten case studies of PPI in research by the Medical Research Council (MRC) clinical trials unit, South et al. (2016) classify involvement as long-term (spanning the duration of the project), ad hoc (drawing on PPI at intervals as required), or one-off (for the purposes of a specific activity) (11). In a recent scoping review of patient engagement in health research, Manafo et al. (2018b) note that long-term PPI involvement across the research lifecycle is only rarely reported on in the literature. For example, they cite a recent review in which the authors identified nearly 200 studies involving Patient Reported Outcome Measures for chronic disease and quality of life impact, of which only 30 per cent involved patients in any part of the research process, and only 10 per cent throughout the duration of the research (21).

4.3. Models of engagement

A wide range of models for engaging patients and the public in health research are described in the literature and overviewed in Table 4 below. They range from models for involvement in a particular project or research study to involvement in the governance structure of a research institute or initiative. Different models for involvement are not mutually exclusive – the same initiative, project or programme can pursue multiple models

and an individual patient or member of the public can be involved in multiple ways. For example, in a survey of chief investigators of health technology assessment (HTA) trials, while almost half of respondents indicated use of a single approach (the majority of which were restricted to membership on the trial steering committee), many employed a multi-model approach (9, 18). Similarly, individual PPI contributors engaged by PPI organisations, research networks or charities may respond to ad hoc requests from project teams, alongside longer-term collaborative roles (7, 25).

Since the focus of this review is on active patient and public involvement in health research, as opposed to contributing purely as participants in research studies (e.g. as participants in clinical trials, or being interviewed on a research topic) the models outlined in Table 4 do not cover the roles that patients and the public play as research participants. However, it is worth noting that the boundaries between active involvement and contribution as research participants are often blurred in the literature and that many articles on PPI cover both active involvement and participant approaches (7, 10, 15, 20, 27-29). This points to a need for a more unified framework for distinguishing between different levels of active engagement.

The NIHR and INVOLVE have tried to clarify the plurality of terms used. INVOLVE uses the terminology of *involvement* to describe contributions “where members of the public are actively involved in research projects and research organisations”; *engagement* to describe “where information and knowledge about research is provided and disseminated” and *participation* “where people take part in a research study” (2, 26). However, in much of the literature the terms involvement and engagement are used interchangeably to cover a broad range of contribution types. In our report, we use the word involvement

and engagement interchangeably, reflecting general practice in the literature and our view of involvement and engagement as being terms that both provide scope for contributions

beyond one-way receipt of information. We recognise the need for further research on terminology in this space.

Table 4: Models of patient and public involvement

Level of engagement	Engagement model
Consultation: providing feedback and advice on specific research activities	<ul style="list-style-type: none"> • Respondents to open consultation on research issues. Examples include consultation on research priorities or patient-relevant outcomes through online surveys, social media and crowdsourcing platforms (31, 32). • Attendees of ad hoc consultation sessions, e.g. expert seminars, focus groups, workshops or consensus meetings (13, 23). • Members of PPI organisations, charities, research networks, clinical studies groups or local PPI panels, responding to ad hoc requests from project teams (7, 11, 14, 25, 28).
Involvement: working directly with a research team throughout the project	<ul style="list-style-type: none"> • Standing members of project advisory groups, together with other stakeholders (8, 17, 18, 23, 27, 29). • Standing members of patient or public advisory groups or PPI panels for a project or programme (7, 24, 28). • Engaged in collaborative research priority-setting activities, e.g. through priority-setting partnerships (4).
Collaboration: partnering on equal footing with researchers in all aspects of research	<ul style="list-style-type: none"> • Engaged in the governance structure of a research institute or funding body as reviewers of proposals and members of funding panels. Patient-Centered Outcomes Research Institute, NIHR and other UK research funding bodies use this model (19, 25, 33). • Engaged in roles within organisations conducting systematic reviews: members of editorial teams, consumer co-ordinator, review author and peer reviewer (34). • Members of project steering committees, management groups or oversight groups for specific elements of a project (7, 8, 11, 18, 20, 35). • Co-researchers embedded as members of a research team and contributing to various aspects of research design, implementation and dissemination (7, 10, 15-17, 28, 29, 36). • Co-applicants on research grants or contracts (29, 35).
Patient or public leadership: leading research decisions and activities	<ul style="list-style-type: none"> • Principal investigators on research studies or leads of patient steering committees (21, 37). • Promoting patient-led research projects, e.g. through organisations such as PatientsLikeMe and 23andMe (33).

In addition, the boundaries between active involvement and more passive participation may be more blurred in applied health research and health services research (especially when experiences or views are being analysed) than in biomedical research or in clinical trials, where the distinction between a study participant and

an active contributor to shaping or conducting the research may be more distinct.

Approaches to recruiting patients and members of the public into active PPI roles are diverse. For example, in a UK survey of chief investigators of health research studies, 40 per cent of PPI contributors were recruited

via voluntary organisations, 35 per cent were recruited via an established service user group, 25 per cent replied to an open invitation, and a small number were recruited via their local NHS comprehensive local research network or research design service (18). Around half of those recruited were previously known to a member of the research team. Vat et al. (2017) identify further potential recruitment approaches, including recruitment via the healthcare system, community outreach, social marketing to enable self-identification, and partnering with marketing companies. They suggest approaches to recruitment need to be tailored to the types of individuals that are being targeted for recruitment (30).

Recently, increased attention is being placed on coordinating involvement across the health research landscape. For example, national bodies (such as INVOLVE) as well as organisations such as National Voices and others, aim to facilitate a co-ordinated approach and to support this with the development and promotion of appropriate guidance and standards of practice.

4.4. Tasks and activities engaged in and specific methods used

Patients and the public undertake a wide range of tasks and activities in fulfilling their roles. Drawing on our analysis of the literature, Table 5, Table 6 and Table 7 below set out the range of tasks and activities in which PPI has been reported, organised by stage of the research process, along with details on specific methods used (based on available evidence).

Table 5 shows the diversity of contributions made at the research preparation and design phase, for example through involvement with

agenda setting, research funding or research design. Table 6 discusses involvement at the study implementation phase in recruitment of study participants, data collection and data analysis. Table 7 discusses PPI in research dissemination and translation, in facilitating research uptake and in evaluation activities.

Contribution roles can be achieved through diverse methods of engagement. Some examples highlighted in the literature include: helping draft funding applications, informing the questions in surveys or interviews, helping organise and design focus groups, piloting surveys, implementing interviews and patient panels, contributing to advisory meetings and expert workshops, conducting literature reviews, contributing to ethnographic research tasks, helping interpret research findings from a patient or public perspective, conference participation, engagement with consensus-building exercises on research priorities, peer-to-peer awareness raising about research opportunities and recruitment, and reviewing funding applications.

While it has not been possible to estimate an overall number of patients or members of the public involved in health research on the basis of this review,⁴ a trend towards increasing involvement has been widely reported in the literature (6, 21). The different models of PPI described in the literature vary considerably with regard to the number of individuals involved. For example, while an advisory board usually has between one and five service users, priority-setting partnerships can involve hundreds or even thousands of service users (4). It is important to flag that the numbers of people who can be involved in specific activities can vary depending on both need and ability to attract contributors.

4 This is due to the varied timeframes of the studies reviewed and a lack of consistency with regard to whether the number of PPI contributors is reported for a given study.

Table 5: PPI tasks, activities and specific methods: research preparation and design

Tasks and activities	Methods of involvement (from least to most engaged)	
Research preparation and design phase		
Agenda setting	<ul style="list-style-type: none"> Identifying or generating research topics or questions (18, 23).⁵ Prioritising topics for research (4, 19, 23, 25, 34). Developing patient-relevant commissioning briefs (23). Providing a patient perspective on outcomes that are important to them and their families, e.g. through participating in patient-centred outcomes research (31, 33, 34). 	<ul style="list-style-type: none"> Surveys, interviews, focus groups and patient panels (4). Use of facilitation tools (e.g. World Café⁶ and Dotmocracy⁷ for collecting data) (4). Use of social media for online survey referral and as online discussion forums (31). Crowdsourcing and recruitment from patient registries to facilitate survey delivery (32). Structured planning processes such as the James Lind Alliance Priority Setting Partnerships (UK), The Dialogue Method (Netherlands), Global Evidence Mapping (Australia), and the Deep Inclusion Method/Choosing All Together (US) (4, 25).
Funding	<ul style="list-style-type: none"> Providing input to funding decisions (19, 25). Contributing to the development of research proposals/funding bids (25, 29, 34, 36). Reviewing research proposals (25, 34). 	<ul style="list-style-type: none"> Ad hoc advice on proposal development via PPI panels etc. (7). Sitting on funding panels/grant review committees (19, 25). Input into funding decisions via patient panel (23). Collaborating on proposal development e.g. discussion via teleconference calls (36).

5 It is important to note that the sources of evidence on patient and public perspectives about research priorities are diverse, although fragmented. For example, some are articulated in priority-setting partnerships, some are known to funders to whom patients can contribute research ideas (e.g. NIHR, Int6), some are known to charities or networks such as Academic Health Science Networks (AHSNs) and Collaborations for Leadership in Applied Health Research and Care (CLAHRCs) via their patient and public consultation and engagement activities, and some are articulated in research organisation reports (Int7). There is no one consolidated source of information, but these types of information sources can contribute active patient involvement in priority setting.

6 World Café is a simple and flexible format for hosting large group dialogue, involving creating a café environment for multiple rounds of small-group discussion. Participants move to a new table after each round and insights from conversations are shared with the larger group. See: <http://www.theworldcafe.com/key-concepts-resources/world-cafe-method/>

7 Dotmocracy or voting with dots is a facilitation method in which participants vote on chosen options using a limited number of dot stickers. The approach is a form of cumulative voting. See: <https://dotmocracy.org/>

Tasks and activities	Methods of involvement (from least to most engaged)
Research preparation and design phase	
Design and procedures <ul style="list-style-type: none"> • Advising on development of surveys and interview guides with respect to accessibility of language, question relevance and appropriateness, or acceptability of format and timings (7-9, 16, 23, 27-29). • Advising on scope and/or search strategy for reviews (20, 23, 36-38). • Advising on the feasibility of conducting research in real-world settings in relation to type/timing of interventions (25) or identifying cultural issues that need to be considered (23). • Advising on variables/outcomes that matter to patients/public (23, 25, 33, 37). • Advising on sampling (8, 25, 27). • Advising on ethical issues such as consent processes (5, 23, 33). • Developing patient information materials (7, 9, 16, 18, 23, 33) and study websites (25). 	<ul style="list-style-type: none"> • Ad hoc input via PPI panels (7). • Discussion fora/conferences (5, 37). • Advisory group meetings and correspondence (8). • Involvement in expert workshop debates e.g. as part of a Delphi study (28). • Piloting survey questions e.g. Delphi study (28).

Table 6: PPI tasks, activities and specific methods: implementation

Tasks and activities	Methods of involvement (from least to most engaged)	
Study implementation		
Recruitment and participation	<ul style="list-style-type: none"> • Advising on recruitment/troubleshooting recruitment difficulties (7, 9, 23, 25, 27). • Identifying or assisting with access to potential research participants (20, 23). • Developing participant recruitment materials (35). • Actively engaging in participant recruitment activities (5, 9, 16, 17). • Advising on how to maintain adherence/ continued participation (25). 	<ul style="list-style-type: none"> • Ad hoc/impromptu consultation (7). • One-off expert workshops (35). • Advisory group meetings and correspondence (27). • Working closely with research team to develop materials (35). • Peer-to-peer recruitment (17). • Research team staff training on how to interact with specific patient groups (17).
Data collection	<ul style="list-style-type: none"> • Contributing to the conduct of literature reviews: locating relevant literature, screening and extracting or coding articles (5, 36, 37, 39). • Collecting data from research participants by conducting interviews, administering surveys, co-facilitating focus groups (17, 23, 29). • Co-generating data with researchers on topics of interest through participatory methods (16). • Contributing to management of data collection (e.g. through tracking participant visits) (17). 	<ul style="list-style-type: none"> • Specific methods for literature review, such as systematic review methodology (5) or qualitative meta-synthesis methodology (36). • Qualitative data-collection methods: (peer-to-peer) interviews and focus groups (17, 23, 29). • Innovative participatory methods, such as participatory learning and action (PLA) using visual representation to aid data generation (16), or video-reflexive ethnography (40).
Data analysis	<ul style="list-style-type: none"> • Actively conducting data analysis tasks (e.g. coding interview transcripts in qualitative studies) (15, 29). • Contributing to data analysis by guiding or identifying themes (23) in both reviews (5, 23, 39) and qualitative studies (7, 10, 27, 29, 36). • Co-analysing with researchers through participatory action learning (16). • Adding patient perspective to the synthesis and interpretation of findings (12, 17, 23, 37). • Highlighting key findings (5, 8, 36). • Contributing to development of practice recommendations (6, 36). 	<ul style="list-style-type: none"> • Conference discussion workshops or expert seminars (23, 37). • Advisory group meetings/discussions (23). • Systematic review methods (5). • Qualitative review methods, following training/guidance (36). • Qualitative data analysis (e.g. thematic networks analytical technique) (10, 15). • Innovative participatory methods for co-analysis of data by patients and researchers (16).

Table 7: PPI tasks, activities and specific methods: dissemination, translation, uptake and evaluation

Tasks and activities	Methods of involvement (from least to most engaged)	
Dissemination and translation		
Dissemination	<ul style="list-style-type: none"> • Contributing to drafting journal articles/ reports/reviews (17, 18, 27, 29, 34, 37). • Critically reviewing articles/reports (5, 7, 10, 15, 18, 28, 34). • Producing or co-creating accessible outputs on study findings, such as plain language summaries (5, 18, 25, 28, 34, 39), participant newsletters/results communication (13, 17), infographics (5) or webinars (8). • Delivering or co-delivering presentations at conferences (17, 23, 29, 34) or project dissemination events (29). • Participating in the release of results or publications (11). • Determining avenues to share findings (17). 	<ul style="list-style-type: none"> • Focus groups involving trial participants to inform preferred option for results (13). • Working collaboratively with research team (5). • Giving media interviews (17).
Uptake	<ul style="list-style-type: none"> • Contributing to clinical guideline development – question development and development of plain English questions (41). • Preparing decision aids for patients (34). 	<ul style="list-style-type: none"> • “Question- development retreat” with prior training on guideline development and topic (41).
Evaluation	<ul style="list-style-type: none"> • Providing feedback on experience as a PPI contributor as part of an evaluation of PPI for a specific project (17, 29, 36). • Participating in research on the topic of PPI (7, 10, 14, 15, 20, 27-29). 	<ul style="list-style-type: none"> • Survey completion (36). • Delphi or modified Delphi surveys (28). • Consensus workshop (20). • Semi-structured interviews (7, 10, 15, 20, 27). • Ethnographic methods, observations and interviews (14).

5 How can effective contributions from patient and public involvement be enabled?

5.1. How can patient and public involvement with research be enabled and rewarded?

Even the most motivated individuals need to be empowered and enabled to engage with research activities. In the literature, a range of practical levers are described. These relate to:

- *The governance and management of PPI*, including having appropriate levels of funding to support PPI contribution, clearly specified roles for PPI contributors and clear processes for initiating contributors to a project;
- *The infrastructure for involvement*, such as user-friendly online platforms for engaging

patients and the public with research tasks and supportive administrative infrastructure;

- *Individual and organisational capacity for involvement*, for example through training and mentorship programmes for both PPI contributors and researchers, and through supportive leadership;
- *Enabling attitudes and behaviours*, such as researchers who value PPI input, a culture of feedback, and a commitment to acknowledging and rewarding contributions.

Table 8 overviews and elaborates on the enablers identified in our literature review and interviews.

Table 8: Enablers of patient and public involvement with research

Type of enabler	Examples
Process governance and management	<ul style="list-style-type: none"> • Financial reimbursement or upfront payments to PPI contributors for expenses and time devoted to contributing (2, 5, 10, 11, 20, 41) (Int1, Int2, Int4, Int5, Int6). Although literature often discusses reimbursement, our interviews highlighted the importance of considering that some individuals may need upfront payments (Int2, Int5, Int6). • Funding for researchers to support effective PPI engagement (28). • Providing appropriate background information on a study to PPI contributors (11), including clarity on what is expected from their role and what they will be doing (10, 15, 36). • Clearly specified and defined goals for PPI and roles of PPI contributors (6, 9, 12, 15, 27, 28, 36, 42). • Engagement early on in a research design process to help inform design and help nurture an ethos of shared ownership (7, 9, 28) (Int1). • Building and nurturing relationships with a core group of people who can act as sounding boards and champions to help attract further engagement (Int1, Int7, Int8).

Type of enabler	Examples
Infrastructure-related enablers	<ul style="list-style-type: none"> • User-friendly involvement platforms and flexible engagement mechanisms. Patients and the public need to be able to contribute in a way that works for them (e.g. remotely, in their own time, online or offline). Crowdsourcing platforms and mobile interfaces can facilitate engagement (20, 32) (Int2, Int7), but offline, face-to-face mechanisms also matter (10, 19) (Int2, Int5, Int6, Int7). As highlighted by an interviewee: <i>"...If you want someone elderly to come to a meeting or appointment, you probably need to time that [in consideration of] free bus transport for elderly kicking in after 10 am ... it is that kind of knowing the context that matters."</i> (Int2). Another interview highlighted that <i>"Anyone under the age of 20 doesn't use email very much – they use snapchat or something like that."</i> (Int5, emphasis inserted). • Realistic timescales for engagement are important for a positive experience, as is supportive administration (Int5, Int6).
Capacity to be involved	<ul style="list-style-type: none"> • Individuals feeling well enough to engage and family support for engagement (8). • Training for researchers on how to do effective PPI (2, 20, 28). • Training and support for PPI contributors on the topic area or on the language and processes of research (2, 10, 19, 43) (Int4, Int7, Int8). • Leadership for PPI activities within initiatives and organisations (2, 37) (Int7).
Behavioural and attitudinal enablers	<ul style="list-style-type: none"> • Receptive researcher attitudes to PPI contributors (8, 10, 19, 28), leading to patient and public representatives feeling <i>welcomed</i> (Black et al. 2018) and <i>respected</i> by the researchers (8, 10), and feeling they <i>can speak freely</i> (10). • PPI contributors' openness to views that may be different from their own (28). • Collaboration: building reciprocal relationships and a commitment to co-learning (6) and working as a team (2, 10, 29) (Int8). • Clear communication lines between researchers and PPI contributors (28) and the use of accessible language (2, 9) (Int2, Int4, Int7, Int8). • A commitment to providing feedback (6, 13, 27) (Int1, Int2, Int3, Int8). <ul style="list-style-type: none"> - Feedback on how an individual has impacted on research facilitates retention and makes it easier for individuals to see the value they add (27) (Int8). - Feedback on study progress (Int1, Int2, Int3, Int8). - Feedback on study results helps individuals feel that they have contributed to a greater good (Int1, Int2, Int3, Int8). • A commitment to evaluation (29). • Acknowledgment and recognition of contribution (2, 28, 32) (Int1, Int2), • Enabling connected PPI contributor communities – including through informal mechanisms and events. Hamilton et al. (2018) note: <i>"Participants valued informal communication with other patient partners and connecting with the research team socially (e.g. research retreats or team lunches with researchers, research trainees or other patient partners)"</i> (10) (page 402). • Advocacy by patient groups (Int8) or public policy bodies.

If you want someone elderly to come to a meeting or appointment, you probably need to time that [in consideration of] free bus transport for elderly kicking in after 10 am ... it is that kind of knowing the context that matters. (Int2)

The boundaries between enablers and rewards can be blurred, as an enabling experience can be rewarding in and of itself. Our analysis of the literature did not identify a specific focus on rewards. However, if we consider rewards to be distinct from enablers in that they are a result of engagement, key types of rewards considered in the literature (and based on our analysis of it) or identified by interviewees highlight the importance of both intrinsic rewards related to the experience of engagement and tangible rewards for contributing (financial, reputational and social). These include:

- **Feedback** which allows PPI contributors to see the value they added and the progress or ultimate impact of the work they were involved in (6, 27) (Int1, Int2, Int3).
- **Acknowledgement and recognition for contributions** (28, 32) (Int1, Int4) to an important health research area. This could range from light-touch and small “thank yous” (Int1) that make people feel special (e.g. for example through a voucher for a special shop or experience), to nominations for internal, regional and national awards and potentially other types of recognition in research outputs (Int1, Int2, Int3). As highlighted by an interviewee: *“Rewards come from the sense that you have made a meaningful contribution.”* (Int4).
- **Financial payments, compensation and rewards** (5, 10, 11, 20, 41) (Int1, Int2, Int4). These need to recognise that

Rewards come from the sense that you have made a meaningful contribution. (Int4)

some individuals do not benefit from reimbursement and need upfront payment in order to be able to engage (e.g. to cover travel or accommodation costs). The scales for honoraria also need to be considered, building on established guidance in the system (for example the practices of INVOLVE, the National Cancer Research Institute (NCRI) and CRUK (Int2)). Two interviewees highlighted that financial payments are more of an enabler, though also a reward – but not the main reward (Int3, Int4).

- **Learning and personal development through the experience**, especially for individuals motivated by an interest in research activity and in learning about research and gaining scientific knowledge (8-11, 14).
- **Social rewards such as new social relationships and networks and feeling like part of a social movement** (Int3), and contributing to something bigger than an individual’s own need and concerns (10, 13).

6 What are the challenges and barriers to patient and public involvement with research?

6.1. Despite a growing awareness of enabling mechanisms, challenges to patient and public involvement persist

The growing focus on and increased commitment to patient and public engagement with research over the past decade has revealed a range of enabling mechanisms and rewards that matter for efficient and effective practice, both to PPI contributors and to researchers. Despite this, challenges to effective involvement are numerous and persistent. These include:

- *Systemic challenges in the research system, related to the governance of PPI in research and to knowledge management.* Examples include 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, and limited patient and public awareness about engagement needs and opportunities.
- *Challenges related to the capacity of individuals to engage.* Examples include lack of experience, knowledge, skills or confidence; lack of access to training; and health and wellbeing related challenges such as inability to travel to research meetings.
- *Administrative and management challenges.* Examples include limited administrative support for implementing PPI processes such as organising meetings and timely payment of contributors, and lack of in-built mechanisms for giving feedback to PPI contributors.
- *Challenges related to culture, values and attitudes.* Examples include tokenism, dismissive attitudes of some researchers, challenges to managing expectations of PPI contributors about the nature and scale of engagement, and managing power dynamics in teams.

Table 9 provides an overview and further detail on the wide range of issues raised in the literature and in our consultations with experts.

Table 9: Challenges to involving patients and the public in research

Type of challenge	Examples
Systemic challenges to the governance of PPI in research and knowledge management of PPI activities	<ul style="list-style-type: none"> • A lack of appropriate funding for researchers to implement effective PPI (2, 5, 15, 18, 20, 23, 28, 30, 37). • A fragmented PPI contributor landscape with little coordination and shared learning between PPI bodies (21), high variability and inconsistencies in practice (2). This impedes the establishment of clear and transparent good practice standards (2). • The absence of a common definition for PPI in research comprising shared understandings of what it entails and how to go about it. The plurality of terms used (e.g. patient involvement, patient engagement, patient participation, service user involvement, citizen engagement, etc.) can contribute to misunderstandings and misinterpretation of goals, expectations and outcomes of PPI involvement in research and challenge effective partnership working between researchers and PPI contributors (16, 44). • A lack of a validated framework(s) to guide researchers on how to conduct effective PPI (5, 21). In relation to this, Brett et al. (2014a) highlight that integrating user views into the research agenda might result in divergence from scientific methods and cause ethical dilemmas during the protocol design. Compromises may have to be reached to ensure user views are incorporated in a relevant but feasible and methodologically sound manner (12) (Int7). • Enactment and appraisal challenges, which impede learning about effective practice. These include limited monitoring of PPI implementation, conflation with standard research methods and limited appraisal activity and evidence of impact (16). • Enrolment, recruitment and retention challenges, including a lack of awareness of what motivates individuals to engage and how to respond to those motivators in recruitment and retention efforts (9, 16, 18), (Int7). • A lack of awareness of existing opportunities for engagement (i.e. information brokering in the research system) (2) (Int4). The NIHR has recently established a web portal (People in Research)⁸ – which advertises opportunities for engagement (Int7). • Lack of public awareness about the need for and potential impact of PPI (30) is a barrier to the scale of engagement that can be achieved. • Mandating PPI in the absence of evaluation of its usefulness for research or its relevance and usefulness for contributors. There is a need to evaluate not only impact but also different types of value (e.g. both research value and social value) (Int2).

Type of challenge	Examples
Capacity to effectively engage	<ul style="list-style-type: none"> • The scientific language and jargon used in research can pose challenges to communication between researchers and lay PPI contributors (18, 19, 28, 36) (Int1, Int2, Int5, Int6, Int7, Int8). • A lack of prior experience with research, which can limit opportunities for some types of engagement (28), especially when “expert patients” are required (e.g. for some funding panels and committees). This will depend on the nature of engagement as in some cases a lay perspective is needed and in some cases a mix of lay and expert perspectives is called for (Int5, Int6). • There are also risks to over-professionalising PPI contributors and losing a lay patient perspective which is sometimes needed (9). This can happen in some models of engagement and with repeated use of the same individuals (Int1, Int3, Int4, Int7). • Health and wellbeing or practical challenges preventing longer-term and continuous engagement related to illness and an inability to travel, (9, 17, 18) (Int4). • Challenges to reaching and recruiting under-represented groups to reflect the true diversity of a society. Many engaged individuals are educated, with professional backgrounds, sometimes with a prior career connection to healthcare services or research (10, 12, 14, 15, 17, 18, 20, 30) (Int1, Int2, Int7, Int8). This can lead to unintended consequences of PPI contribution being seen as the preserve of the “<i>well-educated and reasonably well off middle-classes</i>” (Int2). Citizen science and crowdsourcing models may be able to partially mitigate this.
Administrative and management challenges	<ul style="list-style-type: none"> • Lack of clarity on roles of PPI in projects (5, 10, 15) can lead to a poor PPI contributor experience and misalignment of expectations, and thus limit success. • Poor reporting on PPI processes (their design, implementation and impact) can limit potential for learning and informing future efforts (5). • Limited administrative infrastructure and bureaucracy, including in relation to ethics approval processes, scheduling and logistics of involvement, and timely payment to participants (2, 5, 17, 18, 30). • Time demands or time pressures on PPI contributors or researchers can limit engagement (18, 30) and effective working relationships (23). This includes the potential for PPI contributors to feel that they cannot do the job properly within the time provisions they have (37). • Administrative time and costs of managing PPI contribution processes (12). • Poorly considered recruitment strategies and processes (9, 18, 31), including those rooted in a one-size-fits-all approach. • Lack of in-built feedback mechanisms and practices (36) can impede motivations to engage, retention and learning from past experience.

Type of challenge	Examples
Challenges related to culture, values and attitudes	<ul style="list-style-type: none"> • Tokenism, which impedes the robust implementation of articulated PPI strategies and approaches and their integrity (1, 5, 16, 18, 28). According to Tierney et al. (2016) <i>“There is a sense that those involved in research and healthcare delivery projects believe it is right that they engage with stakeholders to follow policy imperatives, but less evidence that they believe it is worthwhile and valuable as a way of working”</i> (16). • Dismissive attitudes of some researchers are a barrier to effective relationships and contributions. These can be related to a lack of willingness to relinquish power and control, related to perceptions that members of the public bring biased views (2, 5, 9, 15, 20, 28) (Int8). • Some patients are less likely to contribute to research if they do not consider public involvement to be influential (28). • Challenges for researchers in managing expectations of PPI partners (18), for example about the nature of engagement, goals and outcomes. • Managing group dynamics such as issues related to some PPI contributors being dominant and exerting power over others (or perceptions of that being the case) (Int3), and the need to build trust between services users and clinical researchers (6, 37).

Addressing some of the deeply rooted challenges outlined above and nurturing effective practices at scale will call for further change in research cultures and in the attitudes and expectations of researchers, healthcare providers, and patients and the public. Cultures can take time to change and evolve. For example, this applies to addressing instances of tokenistic PPI practices but also to reaching a shared understanding about the purposes of PPI in research and about what constitutes meaningful engagement between academic, healthcare professional and patient and public contributors (Int2, Int7).

There is a need for innovative approaches that balance the ideal solution with what is feasible in the real world. One example of this pertains to representativeness (Int2, Int7). Advances in internet and mobile technology may help to partially address this challenge, including through citizen science and crowdsourcing models which hold promise in enabling the engagement of a large and diverse set of participants (but can themselves exclude those individuals who are not connected to the internet or do not engage with digital platforms). However, securing contributions

that are representative in “statistically valid” terms – however much this should be aimed for – would require a substantial commitment of resources, staff and time to attract and retain participants through a variety of online and offline mechanisms. Moreover, it is likely to exceed what is currently feasible in the health research system, particularly at scale and consistently across projects, programmes and initiatives. However, learning from diversity in itself holds value, provided that the information is considered and interpreted with due caution and caveats in mind (Int2, Int7).

Tackling some other challenges (such as those related to clarity on PPI roles or to accessible language) calls for changes which are comparatively straightforward to address through practical mechanisms. Such mechanisms include clear and documented agreements on roles, the use of practical tools such as language glossaries, and the development of information infrastructures to enable feedback to PPI contributors.

Interventions at the level of the health research system will also be needed. For example, interventions will be needed to ensure

that the funding available in the research funding system can support the scale and nature of PPI that stakeholders may wish to pursue. They will also be required to foster a knowledge management infrastructure in the health research system that can bring

together fragmented information about diverse opportunities for engagement (for example through identifying coordinating and information-broker organisations in combination with online platforms).

7 The impacts of patient and public involvement in research: what we know and what we do not know

7.1. Better evaluative evidence is needed to understand the impacts of patient and public involvement

Patient and public engagement in research has the potential to positively impact on the relevance and quality of research, on the efficiency of the research process and the experience of those involved, and on the uptake of research findings in practice.

However, the evidence base on the nature of PPI contributors' impacts on research is piecemeal and inconclusive, with many studies reporting assumptions, along with hypothesised and perceived impacts, over evidence from evaluations of impact (1, 2, 4, 6, 27, 35). This challenge applies also to evaluations of the involvement of other stakeholder groups in research, and is hence not specific to patient and public involvement only. Manafo et al. (2018a) also highlight the variable quality of studies about the impact of PPI contributions on research (4). Snape et al. (2014) stress that although the absence of evidence on impact does not mean the absence of impact, there is a lack of sufficient evidence on what the actual (as opposed to potential) impacts are, what some of the unintended and undesired consequences might be, how desired impacts are enabled and in what contexts (35). Manafo

et al. (2018a) also highlight a time dimension, with immediate impacts on individuals being easier to identify than medium- and longer-term impacts on organisations or the wider healthcare system (4).

Hughes-Morley et al. (2016) highlight that the likelihood of desired impacts unfolding is likely to be both context- and mechanism-dependent (35), while Staley et al. (2015) stress the need for better understanding of the conditions under which desired impacts are most likely to occur (35). The context- and mechanism-dependence helps in part to explain one of the underlying reasons⁹ for inconclusive findings on the impacts of PPI on specific aspects of research. These aspects include patient recruitment, with some studies reporting positive associations and others finding no significant link (4, 5, 11, 27, 35, 45).

A practical implication of this is the importance of clarity about what an initiative or programme is expecting to achieve from PPI contributions and how. Well-defined expectations and roles can facilitate bespoke evaluations of achievement against goals of specific initiatives. This implies a need to make clear whether an evaluation aims to assess the quality of the PPI engagement process itself, or the outcomes and impacts of that process.

9

Others relate to factors such as variable study quality, lack of robust evaluations and limited shared learning between initiatives.

7.2. Patient and public involvement can lead to numerous potential benefits, but can also have undesired consequences

The core categories of potential or realised impact discussed in the literature include impacts on:

- *Individuals.* Examples include personal benefits, such as those related to individual empowerment of PPI contributors, learning new skills and accessing new knowledge; and enabling researchers to better understand a research area from a public perspective.
- *The quality of research studies and their relevance for patients and the health service.* This can occur through impact on research priorities,¹⁰ helping solve ethical dilemmas, helping with recruitment strategies and their implementation, influencing how data is collected and analysed and on the interpretation of research from a patient and public perspective, and ensuring communication and dissemination of outputs in a language and format that is accessible to patients and the public.
- *The wider research system.* This can occur through impacts on accountability for

resource use, access to research funding, and alignment of research with perceived moral obligations, values and norms.

Tierney et al. (2016) argue that most studies tend to report on real or potential positive impacts from PPI and rarely on negative ones or on unintended consequences, and this is reflected in the overview of impacts considered in the literature (summarised in Table 10) (16). However, a number of factors can influence the directionality of a potential impact (i.e. whether it is positive and enhancing or negative and diminishing in nature). For example, tokenism and dismissive attitudes can hinder the ability to implement a PPI approach in practice in the same way as it is described on paper, such as on a funding application form. This compromised integrity in implementing PPI can in turn influence the nature of impact as well as having a disenfranchising effect on patients who may feel that they are not listened to or valued (2, 12). This may demotivate participants and make their retention in a study or participation in future research less likely (21). Moreover, from a researcher's perspective, feeling mandated to involve patients and the public, even when the researcher does not see the value in their involvement, can demotivate researchers and accentuate tokenistic practice (2).

Table 10: Potential impacts from patient and public involvement identified in the literature

Category of potential impacts	Examples of potential benefits
Personal impact on individuals	<ul style="list-style-type: none"> • Individual benefits for PPI contributors such as individual empowerment to engage in research and collaborate with other stakeholders, confidence building, influence about a health topic and about research, feeling valued, or improved access to information about how to manage a condition (4, 5, 10, 15, 16, 23, 38). • Impact on researcher understanding of a health research area from a public/community perspective, including in relation to understanding the ultimate beneficiary population and making researchers feel more purposeful and better connected to the potential beneficiary (2, 4, 8, 16).

10

We have not come across systematic evaluation evidence of impact from the priorities identified (for example through key approaches such as James Lind Alliance (JLA) priority-setting partnerships) on the uptake and translation of those priorities into research funding calls.

Category of potential impacts	Examples of potential benefits
Impact on research studies	<p><i>Priority setting</i></p> <ul style="list-style-type: none"> • Helping prioritise research topics in a way that better reflects what matters to patients (2, 4, 12, 17, 29, 32). <p><i>Study design</i></p> <ul style="list-style-type: none"> • Contributions to solving ethical dilemmas (27) and informing consent processes efficiently and effectively (5, 12, 46). • Contributing to more patient-focused study designs to enhance relevance for patients (8, 12, 17, 18). • Helping inform various elements of a study design and methods, including research questions and objectives, research methods and protocols, research schedules, and the planning of data analysis (2, 5, 11, 12, 17, 18, 22, 29, 46). • Facilitating better-quality research conduct, informed by a more nuanced understanding of patient and public perspectives and needs (2, 8, 18, 38), with better quality often being defined in the context of more useful and more relevant evidence. • Expanding the potential applicability of research, for example through PPI enabling the inclusion of hard-to-reach populations due to a greater sense of public ownership and acceptability of a study (38). <p><i>Participant recruitment</i></p> <ul style="list-style-type: none"> • Helping inform effective participant recruitment strategies and assistance with recruitment during research implementation, through providing access to participants and increasing public acceptability and understanding of a study and enabling more efficient and effective recruitment processes (2, 4, 5, 7, 11, 12, 16, 17, 22, 27, 29). <p><i>Study implementation</i></p> <ul style="list-style-type: none"> • Impact on how data is collected or analysed (5, 17, 27, 46), including through contributing practical problem-solving skills and adding additional perspectives (5). • Impact on convincing researchers to persist with a study when recruitment or other challenges raise questions about termination (11). • Enabling a better experience for study participants, for example through ensuring that participants are better informed about a study and better prepared to contribute (10, 11, 15, 36). • Helping ensure that a user perspective is reflected in how data is interpreted and meaning constructed (in addition to an academic and clinical perspective) (12). • Contributing to ideas for follow-on work (11). • Tokenism and dismissive attitudes can compromise the ability to implement a PPI approach in practice as it is described on paper in a funding application form. This can demotivate contributors and negatively impact on retention (2, 12). <p><i>Communication and dissemination</i></p> <ul style="list-style-type: none"> • Improved communication with patients and the public, for example through PPI contributors making the information about a study, or the interpretation of results from it, more understandable and accessible for patients and the public (11, 27), or through their active engagement in dissemination (4, 27, 46). • Increased likelihood of translation and uptake of research findings in practice (2, 8, 38). • Acceptability of a study to the public (7, 38).
On the wider research system	<ul style="list-style-type: none"> • Greater accountability regarding the use of public funds by researchers (38). • Impact on access to research funding (5). • Alignment of research practice with the perceived moral obligations of researchers (38), including values and norms in terms of reducing power imbalances and increasing mutual respect and trust between the research and patient community (5, 23).

7.3. How to evaluate patient and public engagement in research

7.3.1. Theoretical debates and challenges for the evaluation of patient and public involvement

The relatively fragmented and inconclusive evidence base on the impact of public and patient involvement in research has been attributed in part to the challenges associated with its evaluation. There is general consensus that evaluating the impact of PPI is methodologically challenging (28) and it has been suggested that the complexity and cost of such evaluation activities may act as an additional practical barrier to their conduct (35). Variability in PPI activities and the context in which they occur poses both methodological and practical challenges. For example, Staley (2015) suggests that statistical evidence of impact is weakened by the failure to sufficiently account for the context in which involvement takes place and the way it is carried out (46). Efforts have also been hampered by a scarcity of established and validated frameworks to support and evaluate patient engagement in research (21) and poor or inconsistent reporting of PPI, which may obscure the real impact of PPI, (16, 23).

There is also debate with respect to whether PPI should be evaluated as a complex intervention, which would call for a particular methodological approach. For example, there is disagreement regarding the appropriateness of using a realist evaluation approach as a means to explore context, mechanisms and outcomes associated with PPI (29, 46). Edelman and Barron (2016) argue that such an approach is inappropriate and that the tendency to evaluate public involvement in research as if it were a complex intervention has in fact impeded the development of an evidence base (1). Instead they suggest that PPI is a part of the research

process (i.e. the activities of research funding, design, conduct, analysis and dissemination), and not a complex intervention. They argue that its misconstruction as such has led to difficulties with evaluation.

There is also significant theoretical debate in the literature regarding the purpose and focus of the evaluation of PPI. There is a general consensus about the intrinsic value of PPI in research (i.e. the idea that there is some type of value regardless of the outcome) and that it should be scrutinised and evaluated. However, there is less agreement when it comes to whether the impact should be quantitatively measured at all (27). Tensions between different rationales for conducting PPI are apparent in the literature. The consequentialist view suggests that PPI is valuable because of beneficial effects on research quality and relevance, while the deontological view expounds a moral duty to give rightful voice and power to those in receipt of healthcare. This has led to contention about what types of impact are worth investigating (and for whom, when and why) and disagreement over appropriate methods and study designs (1).

What is clear from the literature is the need for the selected approach to evaluation to be based on a clear purpose for evaluating involvement in the first place. Two broad categories of evaluation approach are identified in the literature. The first is related to evaluations that seek to assess the *quality* of the PPI engagement process itself, focusing on how well public and patient engagement in research is being conducted and whether it is in line with agreed standards or best practice guidance. These tend to be underpinned by a belief in PPI in research as a moral duty. The second approach relates to evaluations that are particularly concerned with understanding the practical *impacts* of PPI on research or on the wider health system (i.e. whether PPI has an effect with respect to a range of intended

outcomes). In the section below we present examples of identified guidance materials relating to the assessment of both the quality and the impact of PPI. These are summarised in Table 11.

A core message from our analysis of the literature is that evaluations should be clear on the extent to which they are seeking to address the quality of the engagement process, the outcomes and impacts from engagement, or both.

7.3.2. Evaluating the quality and impact of PPI engagement: existing frameworks and guidance

A number of recently published articles and reports set out frameworks or reporting guidelines to assist researchers in planning and conducting PPI and evaluating its impact. Key examples include: the **Public Involvement in Research Standards (3)**, **Patient-Centered Outcomes Research Institute Evaluation framework (17)**, the **Public Involvement Impact Assessment Framework (PiiAF) (47)**, and **Guidance for Reporting the Involvement of Patients and the Public (GRIPP2) (48)**. These are summarised in Table 11. It is important to highlight that these frameworks do not focus exclusively on evaluation of PPI (for example they may inform general standards or approaches for PPI in research) but provide overarching guidance on how it can be evaluated.

The literature highlights the importance of establishing clear definitions of PPI roles and activities and making explicit the expectations

of contributors. This is seen as important for a meaningful and fair evaluation (16). In order to overcome the challenge associated with variability in PPI activities and context, Staley (2015) suggests a need to precisely define the form of PPI (who is engaged and how), the context in which it is undertaken and detailed mechanisms of action (rather than using a loose definition that describes many different types of activity) (46).

There is limited description in the literature of specific data-collection methods or tools used in the evaluation of PPI. In outlining the evaluation approach used by the Patient-Centered Outcomes Research Institute (PCORI) (US), Forsythe et al. (2018) described the use of structured annual investigator progress reports. These are completed by chief investigators for PCORI funded studies, and require investigators to answer closed- and open-ended questions about their experiences with patient and other stakeholder engagement in their projects. In addition, feedback was elicited from stakeholders (including patients) using the Ways of Engaging-Engagement Activity Tool (WE-ENACT) delivered by web-survey or telephone interview (17). However, a range of data-collection methods were also identified that were not specific to PPI *evaluation* but to PPI in research more broadly. These included surveys, focus groups and interviews. In addition, the frameworks and guidelines outlined in Table 11 below provide a structure within which researchers are able to consider the data sources and data-collection methods appropriate to the anticipated outcomes of their PPI activity.

Table 11: Sources of guidance of relevance for evaluating PPI in research

Frameworks/reporting guidelines	Structure
<p>Public Involvement in Research Standards (3):</p> <ul style="list-style-type: none"> Standards providing a framework within which to consider the purpose of public involvement in research and to improve its quality and consistency, outlining the building blocks for “good” public involvement and setting out expectations. Provide clear, concise benchmarks along with indicators against which improvement in PPI practice can be evaluated. 	<p>These standards include:</p> <ul style="list-style-type: none"> The provision of inclusive opportunities (public involvement opportunities that are accessible and that reach people and groups according to research needs). Working together (in a way that values all contributions, and that builds and sustains mutually respectful and productive relationships). Offering and promoting support and learning (building confidence and skills for public involvement in research). Effective communications (using plain language for timely, two-way and targeted communications, as part of involvement plans and activities). Impact (capturing and sharing information on the difference that public involvement makes to research). Involving the public in governance and leadership (so that decisions protect and promote the interests of the public). Within this framework, establishing the impact of public involvement is conceptualised as a single component in evaluating the quality of public involvement.
<p>Patient-Centered Outcomes Research Institute Evaluation framework for stakeholder involvement in PCORI-funded projects (17):</p> <ul style="list-style-type: none"> Framework and conceptual model developed to guide the evaluation of PPI in PCORI-funded research projects. Applicable to research (beyond PCORI) in which healthcare stakeholders (including but not limited to patients) are actively engaged. 	<p>The framework and conceptual model have been used by PCORI as a source of evaluation questions, organised into four areas:</p> <ul style="list-style-type: none"> Predictors: description of engagement approaches (who, when, how, etc.). Intermediate outcomes: effect of engagement on research processes and intermediate outcomes (e.g. research questions, outcomes selected, study design, dissemination of results). Goals: longer-term effects of engagement on achievement of strategic goals (e.g. to increase the quantity and quality of useful information for health decision making). Impact: impact of engagement in research on better health (health decisions, healthcare, health outcomes).
<p>The Public Involvement Impact Assessment Framework (PiiAF) (47):</p> <ul style="list-style-type: none"> Developed by The PiiAF Study Group (including academics, public involvement facilitators from NIHR Research Networks and members of the public). Designed for use at the proposal development stage to help to prompt discussion and consideration of how to assess the impacts of public involvement. It may also be used in the context of ongoing research projects. 	<p>The guidance is presented in two parts:</p> <ul style="list-style-type: none"> Part 1 – Public Involvement Impact Assessment Framework. Provides guidance on five elements: values about public involvement; approaches to public involvement; research focus and study design; practical issues shaping public involvement in research; and the impacts of public involvement. Part 2 – designed to support researchers to develop a plan to assess the impact of public involvement in their research. It takes people through four phases: laying the foundations (who should be involved and why); developing an intervention theory; identifying possible effects of context on impacts of public involvement in research; and formulating assessment questions and study design.

Frameworks/reporting guidelines	Structure
<p>Guidance for Reporting the Involvement of Patients and the Public (GRIPP2) (48):</p> <ul style="list-style-type: none"> • International evidence-based, consensus-informed guidance for reporting PPI. • Aims to improve the quality, content, detail, consistency, transparency and completeness of reporting on engagement activities, which in turn facilitates the evaluation of PPI impact in research studies. 	<p>The GRIPP2 checklists provide key PPI concepts that guide authors on what they should report in papers:</p> <ul style="list-style-type: none"> • GRIPP2-SF includes five items on aims, methods, results, outcomes and critical perspective, and is suitable for studies where PPI is a secondary focus. • GRIPP2-LF includes 34 items on aims, definitions, concepts and theory, methods, stages and nature of involvement, context, capture or measurement of impact, outcomes, economic assessment, and reflections, and is suitable for studies where the main focus is PPI.

8 Reflection on key learning points and areas for consideration in future practice

8.1. Meaningful involvement does not mean involvement in everything

In reflection of the lessons learnt from this rapid evidence assessment, the contents below offer some recommendations for organisations to consider when developing their strategies for engaging with patients and the public. We hope that these will be helpful in informing both THIS Institute's evolving work and the PPI-related efforts of wider stakeholders in the health research system.

The areas for consideration that we highlight are based on the research team's analysis of the literature and interview evidence, and are not intended as prescriptive in nature. Rather, they serve to open discussion and encourage further constructive dialogue and exchange of ideas about how best to engage patients and the public with the Institute's vision and work (and with the work of other organisations interested in establishing effective PPI strategies).

As an overarching principle, when designing a patient and public engagement strategy, it is important for organisations and initiatives to ensure that the PPI contribution is relevant and *meaningful*, both for the research supported and for the stakeholders involved (Int2, Int5, Int7). This means avoiding the risks that mandating PPI or pursuing it in a generic, tick-box fashion could pose to effective

The term meaningful is often used in a meaningless way...what it means needs to be considered.... [Is it about research being] meaningful emotionally for the patient? Or meaningful in terms of influence on research? PPI is not about therapy ... it should mean relevant and effective for research. (Int2)

engagement. It also means ensuring that an organisation's overarching values and principles are reflected in how it designs and implements the engagement strategy.

All of the individuals we interviewed for this study highlighted the importance of a positive, enabled and rewarding experience for PPI contributors. This notwithstanding, one interviewee emphasised the need for more clarity and transparency as to what constitutes meaningful engagement and cautioned about some emerging PPI practices in the research system more widely (Int2). In this person's opinion: *"The term meaningful is often used in a meaningless way...what it means needs to be considered.... [Is it about research being] meaningful emotionally for the patient? Or meaningful in terms of influence on research?"*

It is easy to fall into trap of having to engage everyone in everything. There is a need to prioritise ... you don't want to tie up lots of people's time in activity that might not be high value. Be thoughtful about purpose and priorities. (Int4)

PPI is not about therapy ... it should mean relevant and effective for research. (Int2).

Another individual advised: *"It is easy to fall into trap of having to engage everyone in everything. There is a need to prioritise ... you don't want to tie up lots of people's time in activity that might not be high value. Be thoughtful about purpose and priorities."* (Int4). This includes making pragmatic and carefully considered choices within available resources (Int7). Similarly, another individual commented: *"INVOLVE are very clear that it's not like a ladder. They say really good consultation although quite a one way activity is meaningful and important. Sometimes I think I can't do co-research but if I just do consultation thoughtfully and carefully and I listen and I show people I listened it's still building a relationship. So it might be a bit limited but if you do it and do it well then people trust you and then they want to come back and work with you again."* (Int7).

The core areas for THIS Institute and other organisations to consider when establishing a patient and public engagement approach are summarised below, and relate to three overarching pillars:

- How to prepare for effective engagement;
- How to raise awareness about opportunities and recruit contributors; and
- How to enable engagement and create a supportive and rewarding environment.

8.2. How to prepare for effective engagement

1. **Think carefully about who to involve and why.** Finding the right contributors is key for achieving desired impacts from PPI input. The types of individuals and types of engagement required are likely to vary across projects, across different tasks within a project, and across different types of organisational activities (9, 11).

Identifying who to engage and why calls for reflection on the relative need for lay input versus input from "expert patients" (Int1, Int3, Int5, Int6). It is also necessary to consider whether the nature of participants required varies between different stages or aspects of a project, and how the nature of contributors needed may vary between different models of engagement and aspects of an organisation's or initiative's activities (e.g. project level, or organisation-wide, peer review panels or committees, engagement via a citizen science platform). As emphasised by an interviewee: *"A strategic partner won't be same person as someone who has an interesting story to tell but both will have contribution to make"* (Int4). At a project

Sometimes I think I can't do co-research but if I just do consultation thoughtfully and carefully and I listen and I show people I listened it's still building a relationship. So it might be a bit limited but if you do it and do it well then people trust you and then they want to come back and work with you again. (Int7)

Recruiting people – this is about being clear about who you are, what you are doing, being clear what you’re looking for and why you’re looking for people. It’s about starting where you can and building your networks and confidence and ability over time. It’s accepting that you can’t do all of it straight off but at least pushing yourself to have some commitments to diversity from the outset ... (Int7)

level, identifying appropriate individuals and organisations to involve should be accompanied by consideration of whether specific relationships need to be cultivated beyond individual projects (42).

The theme of representativeness receives a lot of attention in the literature, and is an important factor to consider and aim towards. However, achieving ideal representativeness is often not possible in the real world, and the literature also recognises benefits from involving carefully selected and diverse individuals, even if they do not – in statistical terms – represent the full profile of a community. As highlighted by an interviewee commenting on recruitment more widely: *“Recruiting people – this is about being clear about who you are, what you are doing, being clear what you’re looking for and why you’re looking for people. It’s about starting where you can and building your networks and confidence and ability over time. It’s accepting that you can’t do all of it straight off but at least pushing yourself to have some commitments to diversity from the outset ... It’s about what’s the best we can do right now ... We probably*

won’t be able to get all of it perfectly, won’t have the best involvement in the world straight off...” (Int7).

2. **Ensure that the roles of PPI contributors are clear from the outset, communicated in accessible language and based on shared understanding** and buy in for the roles (including in terms of boundaries to the scope of involvement and reasons for them). The mutual management of expectations between researchers and patient and public contributors is key to effective engagement. In relation to this, role descriptions should be linked to desired outputs and impacts from engagement.

Proactively agreeing on how PPI contributors will engage and managing expectations (both ways, between researchers and PPI contributors) at the outset of a relationship (or as early as possible if the roles are co-created) is critical for efficient and effective working throughout a project or initiative, and for a positive experience. Investing in relationships early on matters (9, 15, 23, 27, 28, 36, 42) (Int2, Int4, Int7).

It’s the initial engagement that is so vital-the bit where you first make contact with people ... If you don’t get it right then you have to try and unpick it and redo it again ... This doesn’t necessarily mean that you have to be clear before you go out to them. It can be that you seek clarity with them so they can co-define their role and shape it. That’s the more empowering relationship. (Int7)

Some people will need training and support to participate. If it's not [in place], then a lot of people will just self-select out of it as they will think that they don't have the right skills and that no one will support them. (Int7)

As highlighted by one interviewee: "It's the initial engagement that is so vital-the bit where you first make contact with people ... If you don't get it right then you have to try and unpick it and redo it again ... This doesn't necessarily mean that you have to be clear before you go out to them. It can be that you seek clarity with them so they can co-define their role and shape it. That's the more empowering relationship. I have had meetings where I've said I don't really know what we're doing but in 2-3 meetings we will all be clear. I'm not certain at the moment and am honest with you, never done this before. I need you to help me shape this but we will be clear in 2-3 meetings time" (Int7).

3. Ensure that PPI contributors are well informed and supported to effectively engage.

This includes: (i) providing sufficient background information about a project and contributor's roles in an accessible form (e.g. avoiding jargon, providing glossaries of terms); (ii) committing to transparency in the goals and expected outcomes both of the project and of PPI contributors' engagement; (iii) thinking and planning carefully about the time required to ensure that individuals understand and

feel comfortable in their roles; and (iv) that they are trained to deliver on the roles (5, 8, 9, 11, 15, 28, 36).

As emphasised by an individual consulted for this research: "Some people will need training and support to participate. If it's not [in place], then a lot of people will just self-select out of it as they will think that they don't have the right skills and that no one will support them. Use the principles as a way of self-checking that you are focusing on the things that help to make it more of a success. This issue is about doing it [PPI] small scale or doing it simple but doing it really well and building a positivity around it and an enthusiasm and trust around it for the people who have participated.[One] should think about the aims of it first-what is the contribution we are looking for from patients and the public? Is it advice, to comment on a patient information leaflet,

This issue is about doing it [PPI] small scale or doing it simple but doing it really well and building a positivity around it and an enthusiasm and trust around it for the people who have participated.[One] should think about the aims of it first-what is the contribution we are looking for from patients and the public? Is it advice, to comment on a patient information leaflet, helping us to carry out interviews as we think they can access a more diverse group of people or have a different kind of conversation? Do this rather than starting with the method [for involvement]. (Int7)

helping us to carry out interviews as we think they can access a more diverse group of people or have a different kind of conversation? Do this rather than starting with the method [for involvement].” (Int7).

4. Think about ethical considerations beyond formal ethical approval process requirements and informed consent. This includes considering:

- Realistic timeframes for PPI contributor engagement (Int5, Int6);
- How contributions will be acknowledged and recognised;
- How research opportunities and outputs will be made accessible to PPI contributors and the wider public (4).

5. Build in monitoring and evaluation mechanisms to learn from experience and inform future actions in terms of PPI contribution (2, 4, 16, 20, 36). Establishing an evaluation plan for PPI activity at the outset of each research programme that an organisation will support is important for fostering an adaptive initiative that promotes continuous learning and can eventually contribute to the wider evidence base on the impacts of PPI in research:

- For each project, the evaluation and learning plan should highlight the potential and desired impact from PPI activities, the expected roles of contributors, the process of engaging them and the methodology to be used in evaluating both the process and impacts of engagement.
- Organisations should also establish clear criteria for what constitutes effective PPI engagement, considering both costs and benefits over time. These criteria may have common as well as unique elements between different projects or types of contribution.

- The evaluation plan for each project should make clear to what extent the focus of the evaluation is on the quality of the PPI process itself, or on the *outcomes and impacts* from PPI, or *both*.

6. Ensure that publications stemming from projects also report on the methods used to engage patients and the public (who was involved, how) and on the outcomes of involvement, so that approaches are replicable in future studies and can contribute to the evidence base on impact.

8.3. How to raise awareness about involvement opportunities and recruit patient and public contributors

7. Design efforts to recruit and retain patient and public contributors in a way that reflects the multitude of factors which motivate people to engage with research.

The communication approach needs to explain the work of an organisation and the engagement opportunity in a way that people can relate to and find compelling, using accessible language. It also needs to make clear why and how individuals can contribute, as some individuals or groups may not immediately understand how they can add value. The key reasons why patients and the public engage with research span altruistic motivations to help others and improve the health system, personal interest in a health topic area and a desire to influence the relevance of research and meaningfulness of research results for service users and the health service, and a general interest in research and in contributing to scientific knowledge. This need for a multipronged “value offer” may be particularly pronounced for projects where large-scale contribution from diverse individuals and groups is required,

as different individuals by a different mix of common and unique factors. Within a multipronged communication approach, there may be a need for bespoke messages for different “cohorts” of people (Int4). One expert recommended avoiding long documents (Int8).

8. Consider the mix of approaches that will allow for effective awareness raising and recruitment, within the resources available. This requires consideration of:

- **The appropriate mix of online and offline mechanisms** for advertising opportunities and targeting individuals or organisations (Int1, Int2, Int3, Int4, Int5, Int6, Int7). For illustrative purposes only, online mechanisms could potentially include organisational websites, websites of partner and collaborating organisations, and social media. Offline mechanisms could span a broad range of activities such as advertising through provider organisations and charities (Int2) via leaflets, pamphlets and newsletters, as well as raising awareness at conferences, community events and various meetings.
- **The extent to which there is a need to directly engage with individuals, organisations and communities** and the extent to which raising awareness about opportunities requires the involvement of intermediary organisations and system stewards (e.g. partner institutions for whom the work is relevant (Int4), patient organisations, PPI networks of healthcare providers and professional associations, and individual brokers and champions (Int2, Int4, Int7, Int8)).

It is important to not always rely on the same people (Int3, Int4), although a core group of individuals can be a useful sounding board for an organisation and

can also assist with accessing wider communities of contributors (Int1). Gillard, Foster and Papoulia (2016) highlight that people who have lived experience and research skills can be particularly valuable in acting as knowledge brokers with legitimacy and credibility across research, practice and service user communities (49). For some types of engagement, it can be important to mitigate against lay individuals becoming “expert PPI contributors” and being influenced by an organisation’s or researchers’ ways of thinking (Int1). This may not matter in all instances, but for some types of engagement lay contributions may be essential. Refreshing committees and creating a safe space so people can openly challenge and critique thinking are ways of mitigating against both inbreeding of ideas and over-professionalisation (Int1, Int7).

While there is value in coordinating with the existing PPI infrastructure at national and regional or local levels, it is equally important to not only rely on the “usual suspects” and to consider how people who have not been reached before can be reached (Int4). Citizen science and web-based platforms (such as the one THIS Institute is developing) may help in this regard but should not be the only mechanism for raising awareness about opportunities and reaching members of the patient and public communities. For THIS Institute, it will be important to be seen as both national and local (Int3).

8.4. How to create an enabling environment for contributions throughout the research process

9. Enable engagement through a mix of levers. These levers should aim to:

- Facilitate an appropriate information environment and support required skills and competencies;
- Ensure appropriate management, governance and administrative arrangements;
- Nurture the requisite infrastructure for engagement;
- Help ensure that organisational values and norms are reflected in the behaviours and attitudes of both researchers and PPI contributors.

Box 1 below provides further detail on how this can be achieved.

Box 1: Levers for an enabling PPI contribution environment

Levers related to the information environment, skills and competencies

- Make sure that information about opportunities for engagement is advertised and communicated clearly, in a timely manner and with appropriate scale and reach. The information communicated should cover the goals of a research initiative and why it matters in a way that is compelling, understandable and motivating for individuals. It should also explain what value patient and public engagement is expected to bring, what the envisaged roles and tasks for PPI contributors are and how their contributions will be used, what the avenues for engagement will be, the approximate timeframes involved and any other project-specific factors of relevance. This could be done through a combination of online mechanisms (e.g. website, email, citizen science platform) and offline interaction (written and verbal), including via system stewards (e.g. charities, research networks with PPI structures, voluntary organisations, professional associations) and individual champions.
- Ensure that PPI contributors receive the requisite training for tasks and that there is troubleshooting support for tasks (e.g. clarifications on tasks or on guidance provided and contact points for any IT-related queries for engagement via a citizen science platform).
- To the extent possible, ensure that researchers have an understanding of the motivations for engagement and of the support needs of the PPI contributor group, and that they communicate in accessible and understandable language. As highlighted by one

...It [communicating about PPI opportunities] must be simple, engaging, in language they [people] understand. It's not about patronising people. It has to be as engaging and as easy to do as humanly possible. [It is about] challenging yourself with what is it we are asking people, does it make sense, does it look interesting, does it look like something they can work out what's being asked of them with one read... If any of those are no it's a stop point and you need to go back... (Int7)

interviewee: *"...It [communicating about PPI opportunities] must be simple, engaging, in language they [people] understand. It has to be as engaging and as easy to do as humanly possible. [It is about] challenging yourself with what is it we are asking people, does it make sense, does it look interesting, does it look like something they can work out what's being asked of them with one read... If any of those are no it's a stop point and you need to go back..."* (Int7).

Levers related to governance, management and administration

- Build in mechanisms for regular feedback on individual contributions, on study progress, and when possible on outputs and impacts (e.g. via online platforms, newsletters and other mechanisms). This is critical for motivating individuals and helping them feel valued, as well as for retention of contributors (15, 16, 27, 36). It is important both to provide some immediate feedback and to be clear on what longer-term feedback is yet to come (Int2). Consider how feedback can also be given on individual performance and progress (e.g. success with tasks, through gamification approaches).
- Recognise that some individuals will be interested in feedback about an organisation's overall progress and feedback across a portfolio of projects. In relation to THIS Institute, the idea of a "citizen science club" option was identified as one way of providing regular update bulletins across a portfolio of work to those interested (Int2). This could be administered on the website, by email or in paper format.
- Acknowledge and reward contributions (e.g. vouchers that feel like a treat, nominations for awards, recognition of contribution in research outputs).
- Consider the likely budget needed to support the diverse PPI engagement activities in annual budgeting cycles and establish a payment and reimbursement process which is not overly bureaucratic and makes it straightforward for individuals to receive payment.
- Identify clear leads for PPI activities in projects and organisational level (37). These individuals should not only have oversight roles, but also be involved with managing and facilitating engagement.

Levers related to infrastructure

- Design engagement platforms and mechanisms (online or offline) that – to the extent possible – support flexibility in terms of when people might contribute and how. Structured methods for involvement (especially when accompanied by clear guidance) are seen to assist with clarity in terms of how to engage and clarity of expectations (37). However, within those methods there is a need for flexibility to accommodate engagement without interfering unduly with people's wider lives (Int2). For example, interviewees highlighted that "Engagement cannot be a 9–5 only ... offering involvement from the comfort of a person's home online is attractive, especially if it can be made quick and easy" (Int1).
- Mechanisms for engagement need to support both online and offline interactions, face-to-face and remote. This means that the knowledge management infrastructure needs to accommodate multiple mechanisms of gathering and analysing information. This is important to allow diverse groups to engage.

Behavioural and attitudinal levers

- Ensure that PPI practices align with organisational values and principles (2).

- Ensure that members of the research team and PPI contributors have a shared understanding of the goals, expectations, reasons for and scope of PPI contribution.
- Ensure researchers know what it means to engage effectively in their style of working and behaviours (e.g. avoiding jargon, showing welcoming attitudes, transparency, self-reflexiveness, respect).
- Ensure that PPI contributors are clear on the scope of their role, and support PPI contributors (when needed and to the extent possible given the nature of a task) to be open to, value and constructively engage with different views.
- Build relationships between researchers and PPI contributors around clarity in shared and unique goals and drivers.
- Aim to engage contributors early on, including in the scoping and design of projects. This is seen to enable a sense of shared ownership and coproduction (7, 9, 28) (Int1, Int4, Int8).
- Nurture relationships (online and offline, formally and informally – potentially through feedback, supporting communities of contributors).

References

1. Edelman N, Barron D. Evaluation of public involvement in research: time for a major re-think? *Journal of health services research & policy*. 2016;21(3):209-11.
2. NIHR. Going the Extra Mile 2015 [Available from: <https://www.nihr.ac.uk/patients-and-public/documents/Going-the-Extra-Mile.pdf>]
3. The Standards Development Partnership. National Standards for Public Involvement 2018 [Available from: http://www.invo.org.uk/wp-content/uploads/2018/03/71110_A4_Public_Involvement_Standards_v4_WEB.pdf]
4. Manafo E, Petermann L, Vandall-Walker V, Mason-Lai P. Patient and public engagement in priority setting: A systematic rapid review of the literature. *PloS one*. 2018;13(3):e0193579.
5. Price A, Albarqouni L, Kirkpatrick J, Clarke M, Liew SM, Roberts N, et al. Patient and public involvement in the design of clinical trials: An overview of systematic reviews. *Journal of evaluation in clinical practice*. 2018;24(1):240-53.
6. Shippee ND, Domecq Garces JP, Prutsky Lopez GJ, Wang Z, Elraiyah TA, Nabhan M, et al. Patient and service user engagement in research: a systematic review and synthesized framework. *Health expectations : an international journal of public participation in health care and health policy*. 2015;18(5):1151-66.
7. Dudley L, Gamble C, Preston J, Buck D, Hanley B, Williamson P, et al. What Difference Does Patient and Public Involvement Make and What Are Its Pathways to Impact? Qualitative Study of Patients and Researchers from a Cohort of Randomised Clinical Trials. *PloS one*. 2015;10(6):e0128817.
8. Forsythe LP, Frank LB, Workman TA, Hilliard T, Harwell D, Fayish L. Patient, caregiver and clinician views on engagement in comparative effectiveness research. *Journal of comparative effectiveness research*. 2017;6(3):231-44.
9. Gamble C, Dudley L, Allam A, Bell P, Buck D, Goodare H, et al. Health Services and Delivery Research. An evidence base to optimise methods for involving patient and public contributors in clinical trials: a mixed-methods study. Southampton (UK): NIHR Journals Library; 2015.
10. Hamilton CB, Hoens AM, Backman CL, McKinnon AM, McQuitty S, English K, et al. An empirically based conceptual framework for fostering meaningful patient engagement in research. *Health expectations : an international journal of public participation in health care and health policy*. 2018;21(1):396-406.

11. South A, Hanley B, Gafos M, Cromarty B, Stephens R, Sturgeon K, et al. Models and impact of patient and public involvement in studies carried out by the Medical Research Council Clinical Trials Unit at University College London: findings from ten case studies. *Trials*. 2016;17:376.
12. Brett J, Staniszewska S, Mockford C, Herron-Marx S, Hughes J, Tysall C, et al. A systematic review of the impact of patient and public involvement on service users, researchers and communities. *The patient*. 2014;7(4):387-95.
13. Racine E, Hurley C, Cheung A, Sinnott C, Matvienko-Sikar K, Baumgartner C, et al. Participants' perspectives and preferences on clinical trial result dissemination: The TRUST Thyroid Trial experience. *HRB Open Research*. 2018.
14. Thompson J, Bissell P, Cooper CL, Armitage CJ, Barber R. Exploring the impact of patient and public involvement in a cancer research setting. *Qualitative health research*. 2014;24(1):46-54.
15. Black A, Strain K, Wallsworth C, Charlton SG, Chang W, McNamee K, et al. What constitutes meaningful engagement for patients and families as partners on research teams? *Journal of health services research & policy*. 2018:1355819618762960.
16. Tierney E, McEvoy R, O'Reilly-de Brun M, de Brun T, Okonkwo E, Rooney M, et al. A critical analysis of the implementation of service user involvement in primary care research and health service development using normalization process theory. *Health expectations : an international journal of public participation in health care and health policy*. 2016;19(3):501-15.
17. Forsythe L, Heckert A, Margolis MK, Schrandt S, Frank L. Methods and impact of engagement in research, from theory to practice and back again: early findings from the Patient-Centered Outcomes Research Institute. *Quality of life research : an international journal of quality of life aspects of treatment, care and rehabilitation*. 2018;27(1):17-31.
18. Mathie E, Wilson P, Poland F, McNeilly E, Howe A, Staniszewska S, et al. Consumer involvement in health research: A UK scoping and survey. *Int J Consum Stud*. 2014;38(1):35-44.
19. van Bekkum JE, Hilton S. UK research funding bodies' views towards public participation in health-related research decisions: an exploratory study. *BMC health services research*. 2014;14:318.
20. Miller CL, Mott K, Cousins M, Miller S, Johnson A, Lawson T, et al. Integrating consumer engagement in health and medical research - an Australian framework. *Health research policy and systems*. 2017;15(1):9.
21. Manafo E, Petermann L, Mason-Lai P, Vandall-Walker V. Patient engagement in Canada: A scoping review of the 'how' and 'what' of patient engagement in health research. *Health research policy and systems*. 2018;16(1).
22. Ray KN, Miller E. Strengthening stakeholder-engaged research and research on stakeholder engagement. *Journal of comparative effectiveness research*. 2017;6(4):375-89.
23. Brett J, Staniszewska S, Mockford C, Herron-Marx S, Hughes J, Tysall C, et al. Mapping the impact of patient and public involvement on health and social care research: a systematic review. *Health expectations : an international journal of public participation in health care and health policy*. 2014;17(5):637-50.

24. Gray-Burrows K, Willis T, Foy R, Rathfelder M, Bland P, Chin A, et al. Role of patient and public involvement in implementation research: a consensus study. *BMJ quality & safety*. 2018.
25. Green G. Power to the people: To what extent has public involvement in applied health research achieved this? *Research involvement and engagement*. 2016;2:28.
26. INVOLVE. n.d. [Available from: <http://www.invo.org.uk/about-involve/>]
27. Crocker JC, Boylan AM, Bostock J, Locock L. Is it worth it? Patient and public views on the impact of their involvement in health research and its assessment: a UK-based qualitative interview study. *Health expectations : an international journal of public participation in health care and health policy*. 2017;20(3):519-28.
28. Snape D, Kirkham J, Britten N, Froggatt K, Gradinger F, Lobban F, et al. Exploring perceived barriers, drivers, impacts and the need for evaluation of public involvement in health and social care research: a modified Delphi study. *BMJ open*. 2014;4(6):e004943.
29. Wilson P, Mathie E, Keenan J, McNeilly E, Goodman C, Howe A, et al. Health Services and Delivery Research. *ReseArch with Patient and Public involvement: a RealisT evaluation - the RAPPORT study*. Southampton (UK): NIHR Journals Library 2015.
30. Vat LE, Ryan D, Etchegary H. Recruiting patients as partners in health research: a qualitative descriptive study. *Research involvement and engagement*. 2017;3:15.
31. Dyson MP, Shave K, Fernandes RM, Scott SD, Hartling L. Outcomes in Child Health: Exploring the Use of Social Media to Engage Parents in Patient-Centered Outcomes Research. *Journal of medical Internet research*. 2017;19(3):e78.
32. Truitt AR, Monsell SE, Avins AL, Nerenz DR, Lawrence SO, Bauer Z, et al. Prioritizing research topics: a comparison of crowdsourcing and patient registry. *Quality of life research : an international journal of quality of life aspects of treatment, care and rehabilitation*. 2018;27(1):41-50.
33. Sacristan JA, Aguaron A, Avendano-Sola C, Garrido P, Carrion J, Gutierrez A, et al. Patient involvement in clinical research: why, when, and how. *Patient preference and adherence*. 2016;10:631-40.
34. Morley RF, Norman G, Golder S, Griffith P. A systematic scoping review of the evidence for consumer involvement in organisations undertaking systematic reviews: focus on Cochrane. *Research involvement and engagement*. 2016;2:36.
35. Hughes-Morley A, Hann M, Fraser C, Meade O, Lovell K, Young B, et al. The impact of advertising patient and public involvement on trial recruitment: embedded cluster randomised recruitment trial. *Trials*. 2016;17(1):586.
36. Bayliss K, Starling B, Raza K, Johansson EC, Zabalán C, Moore S, et al. Patient involvement in a qualitative meta-synthesis: lessons learnt. *Research involvement and engagement*. 2016;2:18.
37. Boote J, Baird W, Sutton A. Public involvement in the systematic review process in health and social care: a narrative review of case examples. *Health policy (Amsterdam, Netherlands)*. 2011;102(2-3):105-16.
38. Esmail L, Moore E, Rein A. Evaluating patient and stakeholder engagement in research: moving from theory to practice. *Journal of comparative effectiveness research*. 2015;4(2):133-45.

39. Gradinger F, Britten N, Wyatt K, Froggatt K, Gibson A, Jacoby A, et al. Values associated with public involvement in health and social care research: a narrative review. *Health expectations : an international journal of public participation in health care and health policy*. 2015;18(5):661-75.
40. Collier A, Wyer M. Researching Reflexively With Patients and Families: Two Studies Using Video-Reflexive Ethnography to Collaborate With Patients and Families in Patient Safety Research. *Qualitative health research*. 2016;26(7):979-93.
41. Armstrong MJ, Mullins CD, Gronseth GS, Gagliardi AR. Impact of patient involvement on clinical practice guideline development: a parallel group study. *Implementation science : IS*. 2018;13(1):55.
42. INVOLVE & NIHR. Guidance on co-producing a research project 2018 [Available from: <http://www.invo.org.uk/posttypepublication/guidance-on-co-producing-a-research-project/>]
43. Parkes JH, Pyer M, Wray P, Taylor J. Partners in projects: preparing for public involvement in health and social care research. *Health policy (Amsterdam, Netherlands)*. 2014;117(3):399-408.
44. Gallivan J, Kovacs Burns K, Bellows M, Eigenseher C. The many faces of patient engagement. *The Journal of Participatory Medicine Research*. 2012;4:(e32).
45. Johnson M, O'Hara R, Hirst E, Weyman A, Turner J, Mason S, et al. Multiple triangulation and collaborative research using qualitative methods to explore decision making in pre-hospital emergency care. *BMC medical research methodology*. 2017;17(1):11.
46. Staley K. 'Is it worth doing?' Measuring the impact of patient and public involvement in research. *Research involvement and engagement*. 2015;1:6.
47. Popay C, and with the PiiAF Study Group. The public involvement impact assessment framework guidance 2014 [Available from: <http://piiaf.org.uk/documents/piiaf-guidance-jan14.pdf>]
48. Staniszewska S, Brett J, Simera I, Seers K, Mockford C, Goodlad S, et al. GRIPP2 reporting checklists: tools to improve reporting of patient and public involvement in research. *Research involvement and engagement*. 2017;3:13.
49. Gillard S, Foster R, Papoulias C. Patient and public involvement and the implementation of research into practice. *J Ment Health Train Educ Pract*. 2016;11(4):256-67.
50. Moule P, Davies R. A devolved model for public involvement in the field of mental health research: case study learning. *Health expectations : an international journal of public participation in health care and health policy*. 2016;19(6):1302-11.
51. Mayan M, Lo S, Richter S, Dastjerdi M, Drummond J. Community-Based Participatory Research: Ameliorating Conflict When Community and Research Practices Meet. *Progress in community health partnerships : research, education, and action*. 2016;10(2):259-64.
52. Ridders W, Boterhoven de Haan K, Lawrence D, McKenzie A, Hancock K, Haines H, et al. Two methods for engaging with the community in setting priorities for child health research: who engages? *PloS one*. 2015;10(5):e0125969.
53. D'Udekem Y, Forsdick V, Du Plessis K. Involvement of patients and parents in research undertaken by the Australian

- and New Zealand Fontan Registry. *Cardiol Young*. 2018;28(4):517-21.
54. INVOLVE. Examples of projects using social media for public involvement n.d. [Available from: <http://www.invo.org.uk/resource-centre/libraries/examples/examples-of-projects-using-social-media-for-public-involvement/>]
 55. INVOLVE & NIHR. Exploring public involvement in research funding applications 2013 [Available from: http://www.invo.org.uk/wp-content/uploads/2014/10/INVOLVEExploringPublicInvolvementInResearchFundingApplications_linksSept14FINAL.pdf]
 56. INVOLVE & NIHR. Exploring the impact of public involvement on the quality of research: examples 2013 [Available from: http://www.invo.org.uk/wp-content/uploads/2014/11/INVOLVEExploringImpactQualityResearchExamples_updated-July-2014.pdf]
 57. INVOLVE & NIHR. Examples of projects using social media for public involvement 2014 [Available from: <http://www.invo.org.uk/wp-content/uploads/2014/12/Social-media-guidance-FULL-examples.pdf>]
 58. NIHR. People in Research n.d. [Available from: <https://www.nihr.ac.uk/patients-and-public/how-to-join-in/people-in-research.htm>]
 59. Minervation. People in research: a new website connecting members of the public with researchers 2010 [Available from: <http://www.minervation.com/tag/people-in-research/>]
 60. People in Research. About this site 2014 [Available from: <https://www.peopleinresearch.org/about/>]
 61. Madden M, Morley R. Exploring the challenge of health research priority setting in partnership: reflections on the methodology used by the James Lind Alliance Pressure Ulcer Priority Setting Partnership. *Research involvement and engagement*. 2016;2:12.
 62. The James Lind Alliance. The James Lind Alliance Guidebook 2018 [Available from: <http://www.jla.nihr.ac.uk/jla-guidebook/downloads/Print-JLA-guidebook-version-7-March-2018.pdf>]
 63. The James Lind Alliance. Reporting on results and impact 2018 [Available from: <http://www.jla.nihr.ac.uk/jla-guidebook/downloads/Print-JLA-guidebook-version-7-March-2018.pdf>]
 64. The James Lind Alliance. Making a difference n.d. [Available from: <http://www.jla.nihr.ac.uk/making-a-difference/>]
 65. The James Lind Alliance. Funded research n.d. [Available from: <http://www.jla.nihr.ac.uk/jla-guidebook/downloads/Print-JLA-guidebook-version-7-March-2018.pdf>]
 66. The James Lind Alliance. Funded research n.d. [Available from: <http://www.jla.nihr.ac.uk/making-a-difference/funded-research.htm>]
 67. The James Lind Alliance. Working with the priorities 2018 [Available from: <http://www.jla.nihr.ac.uk/making-a-difference/working-with-the-priorities.htm>]
 68. Simpson A, Jones J, Barlow S, Cox L. Adding SUGAR: service user and carer collaboration in mental health nursing research. *Journal of psychosocial nursing and mental health services*. 2014;52(1):22-30.
 69. Collins K, Boote J, Ardron D, Gath J, Green T, Ahmedzai SH. Making patient and public involvement in cancer and palliative research a reality: academic support is vital

- for success. *BMJ supportive & palliative care*. 2015;5(2):203-6.
70. Sheffield Hallam University. North Trent Cancer Research Network Consumer Research Panel 2018 [Available from: <https://www.shu.ac.uk/research/specialisms/health-and-social-care-research/what-we-do/our-expertise/health-care-and-service-delivery-research/case-studies/north-trent-cancer-research-network-consumer-research-panel>]
71. Arain M, Pyne S, Thornton N, Palmer S, Sharma RA. Consumer involvement in cancer research: example from a Cancer Network. *Health expectations : an international journal of public participation in health care and health policy*. 2015;18(5):1530-42.
72. Skilton E, Aslam M, Yeung J, Gao F, Melody T. Embedding patient and public involvement within research - How to set up a research patient ambassador group within a NHS trust. *J Intensive Care Soc*. 2016;17(3):234-7.
73. Varker T, Forbes D, Dell L, Weston A, Merlin T, Hodson S, et al. Rapid evidence assessment: increasing the transparency of an emerging methodology. *Journal of evaluation in clinical practice*. 2015;21(6):1199-204.

Appendix A. Examples of patient and public involvement in practice

Throughout this report, we have discussed a diverse range of approaches to involving patients and the public in research, as well as associated enablers and challenges, and potential impacts. The examples below illustrate this learning in practice. They span

examples of both project-level Patient and Public Involvement (PPI) and initiatives seeking to facilitate PPI across a diversity of project and programmes. They are based on examples from papers we reviewed for this research and selected in consultation with THIS Institute.

Project-specific examples of patient and public involvement in research

Box 2: The many roles service users can play in a project: the case of collaboration in research on paramedic decision making

Collaboration with service users in research to understand influences on paramedic decisions

What was the research project or initiative about?

Johnson et al. (2017) detail the methodology used in a research project that explored organisational, community and individual influences on paramedic decision making in the pre-hospital, ambulance service emergency care system, at three NHS Ambulance Trusts in England. Their study describes a multi-stakeholder, multi-method approach (including document reviews, focus groups, workshops, interviews, audio recordings representing digital diaries, and observations) that aimed to enable a more thorough understanding of the influences on decision making.

How were patients and the public involved?

Service users were recruited through existing PPI networks of the participating ambulance trusts and from wider community networks. Service users contributed in diverse ways, for example by participating in focus groups and engaging with the thematic analysis of data emerging from the focus groups. They also contributed to verifying findings and providing a user-based interpretation of the research results. In addition, service users helped in recruiting participants for the research. Some members of a PPI panel provided inputs into reviewing research ethics documentation and producing leaflets about the study findings in a language that would be accessible to the wider public. A member of the local PPI panel was also a study co-applicant and participated in the design of the research, recruitment, data collection, data analysis and disseminating findings.

What enabled engagement?

The effort invested by the research team in building relationships with service users that were contributing to the research, together with the regular discussion and feedback on emerging themes and findings, helped nurture effective engagement. Establishing contacts within existing PPI networks enabled the recruitment of additional service users.

What were the challenges?

The nature and scale of engagement had time implications for the study's progress and the complexity of the study design.

Impacts

Johnson et al. (2017) suggest that any challenges were offset by enhanced insights on the decision-making process and enhanced confidence in the research findings associated with the diversity of stakeholders that contributed. Service user engagement contributed to the accessibility and reach of the findings, allowing for outputs to be understandable to a lay audience and disseminated via local PPI and service provider networks.

Source: Johnson, M., et al. (2017). "Multiple triangulation and collaborative research using qualitative methods to explore decision making in pre-hospital emergency care." *BMC Medical Research Methodology* (45).

Box 3: A case study in public involvement in mental health research**A devolved model for public involvement in the field of mental health research****What was the research project or initiative about?**

Moule and Davies (2015) examine the impact of involving the public in a specific case study that was part of a wider research project conducted by a mental health charity. The case study aimed to evaluate the impact and experience of personalisation and using personal budgets for people with mental illnesses. Personalisation in social care allows individuals to identify what support they need and enables support packages to be tailored specifically for them. The process includes providing payments to service users to use for their care needs.

How were patients and the public involved?

Service users were interviewed by researchers for the case study of personalisation, with a focus on the service users' roles in research, the support and training made available to them to support involvement with research, the impact of involvement, the feedback they received from researchers, what had worked well and whether they experienced any challenges.

What enabled engagement?

Key enablers identified by Moule and Davies included: (i) ongoing support provided by researchers to individual service users, such as availability to answer emerging questions; and (ii) effective relationships enabled by patient and public contributors feeling that they were valued and listened to.

What were the challenges?

Some challenges related to the alignment of expectations between researchers and PPI contributors were reported despite a written agreement on roles. For example, some contributors

wanted to be more involved with activities such as interviewing, while others felt they would have benefited from more background information on the research. Tensions also manifested themselves in efforts to involve service users heavily and to support a user-led involvement model, while also managing the practical aspects of carrying out complex research. Turnover of team members affected continuity in the research, accentuating challenges to ensuring ongoing clarity in service user roles.

Impacts

PPI impacted on the research protocols used (for example, members of the user-led organisations influenced adaptations in interview questions to make the language used more relevant and accessible) and contributed to the nature of data-collection methods and dissemination plans.

Source: Moule, P. and Davies, R. (2015). "A devolved model for public involvement in the field of mental health research: case study learning." *Health Expectations*, 19, pp.1302–1311 (50).

Box 4 : Addressing challenges and trade-offs in community-based participatory research

Families First Edmonton: participatory research on improving health outcomes for low income families

What was the research project or initiative about?

The paper specifically focuses on the real-world challenges of collaborative working between researchers and community members, based on the experiences of Families First Edmonton – a community-based participatory research project. Families First Edmonton was a longitudinal randomised controlled trial and aimed to advance knowledge about the types of interventions that could improve health outcomes of low-income families.

How were patients and the public involved?

The idea for the project originated in response to a community request for evidence on improved (cost-effective and efficient) services for low-income families. Members of the community were involved in selecting the research design together with researchers, and in developing the logic model for the intervention. The logic model outlined the conditions that the intervention would seek to address, how it would address them, and the expected outcomes. They also offered suggestions on how to select a partner to deliver the intervention. Community members suggested this be done through a request for proposals.

What enabled engagement?

Mayan et al. (2016) focus explicitly on challenges to implementing community-based participatory research. However, a commitment to addressing challenges rather than abandoning a project sheds light on some enablers of progress. In the Families First Edmonton project, this was done by revisiting the original logic model for the intervention and confronting differences (but working to find commonalities rather than dwelling on differences). Finding a concrete task around which to constructively engage in dialogue and debate facilitated conflict resolution and enabled a feeling of collective ownership.

What were the challenges?

Different perspectives and thought processes – between community members and researchers, and between community members and a chosen service provider for the intervention – led to relational tensions and clashes, stifling timely progress.

Impacts

Community-based participatory research approaches in this project had unintended consequences, most notably in the form of project delays and strained collaborator relationships. The timely delivery of the trial was impeded and the methodological approach for the project had to be adapted. However, Mayan et al. (2016) also highlight the impacts that the participatory approach had on establishing a shared understanding and appreciation of the realities and constraints within which the trial would need to be delivered, allowing for a more context-sensitive, adaptive approach.

Source: Mayan, M., et al. (2016). "Community-Based Participatory Research: Ameliorating Conflict When Community and Research Practices Meet." *Prog Community Health Partnersh* 10(2): 259–264 (51).

Box 5: Understanding who engages with priority setting in child health research

Two Methods for Engaging with the Community in Setting Priorities for Child Health Research: Who Engages?

What was the research project or initiative about?

This study aimed to evaluate participatory methods used to obtain views from the community in relation to research in child health. Ridders et al. (2015) compared a public forum approach (termed Community Conversations) that was used by the Western Australian Telethon Kids Institute Participation Program, with an approach based on telephone surveys. The researchers wanted to investigate how representative the Community Conversations approach was of the wider population.

How were patients and the public involved?

The public were involved through telephone surveys and two Community Conversations (public discussion forums). The telephone survey consisted of questions about people's lived experience and what their opinions were about participation of the community in research, as well as on research priorities. Attendees of the two Community Conversations were shown a presentation on research in childhood education. Individuals were recruited to the first Community Conversation forum through the participation network of the Western Australian Telethon Kids Institute Participation Program (11 participants attended). The second Conversation consisted of individuals who were invited during the telephone survey (only 3 participants attended). The telephone survey received 816 responses with nearly 26 per cent of participants identifying children's mental health as the most essential area to research out of five topics: children's nutrition, childhood obesity, childhood education, language development and children's mental health. The telephone survey allowed information to be collected across a wider range of individuals and the authors highlight that this enabled analysis of a more quantitative nature. In contrast, the Conversations enabled the collection of more in-depth information about a specific subject.

What enabled engagement?

The Western Australian Telethon Kids Institute Participation Program advertised information about the Community Conversations through a website (www.involvingpeopleinresearch.org.au) and through community and consumer organisations that were relevant to the specific topic that

was to be discussed. This combination of online awareness raising and community engagement through intermediary organisations facilitated recruitment of research participants.

What were the challenges?

Only two (plus a partner of one of those individuals) of the 816 people telephoned as part of the survey attended a Conversation, indicating that this type of cold-calling approach is not a successful method for recruiting participants to discussion fora. The authors identified challenges to ensuring a representative sample.

Impacts

The authors highlight that a telephone survey can yield information that is useful for identifying priority areas for research in child health and how the community could be involved in their research.

Source: Ridders, W., et al. (2015). "Two Methods for Engaging with the Community in Setting Priorities for Child Health Research: Who Engages?" PLoS ONE 10 (5) (52).

Box 6: Involving patients and parents in research – the importance of facilitating communities

Involvement of patients and parents in research undertaken by the Australian and New Zealand Fontan Registry

What was the research project or initiative about?

The paper discusses a model for PPI used by the Australian and New Zealand Fontan Registry to involve patients with a Fontan circulation (which results from an operative procedure to correct a heart defect) and their parents, with a desire to achieve more relevant, service user-focused research.

How were patients and the public involved?

The model used by the Australian and New Zealand Fontan Registry involved the participation of patients and their parents on the Steering Committee, together with individuals in paediatric and adult care for coronary heart disease. The role of the patients and their parents on the Steering Committee was to be representatives of the community and review the information that was communicated to patients and their families. The decision to include patients in this committee was as a result of a recommendation by a professor who was a leading figure in registry research in Australia. This professor felt that PPI could help improve the research being conducted. The patient and public representatives on the project's Steering Committee reviewed information that was intended to be provided to patients and their parents. For example, they provided feedback on information about the expected outcomes after a Fontan surgery and particularly the way it was worded, giving advice to ensure appropriate, understandable and accessible content. Patients were also involved in an annual Fontan education day, the first of which was held in 2014. The aim of this day was to inform the patient community about recent research results. After this event in 2014, patients and parents created an advocacy group, the Australian and New Zealand Fontan Advisory Committee. The aims of the group have been to improve the support offered to individuals with a Fontan circulation. This group identified the major concerns and needs of patients and their families by conducting a survey.

What enabled engagement?

Enablers were not discussed in the paper.

What were the challenges?

D'Udekem et al. (2018) do not discuss any challenges related to the Australian and New Zealand Fontan Registry study specifically.

Impacts

Patient engagement enabled the production of research documentation that used language that was more accessible, understandable and acceptable to patients and the public. Their engagement also helped assess study feasibility and the required timeframes for research delivery. The formation of the patient community has helped identify new research topics for projects conducted by the Australian and New Zealand Fontan Registry.

Source: d'Udekem, Y., et al. (2018). "Involvement of patients and parents in research undertaken by the Australian and New Zealand Fontan Registry." *Cardiology in the Young*, 28, 517–521 (53).

Examples of initiatives to involve patients and the public across diverse projects and programmes of research

Box 7: INVOLVE

INVOLVE

What is INVOLVE and what does it aim to achieve?

INVOLVE is a national advisory group aiming to enable and support active involvement of members of the public in NHS, public health and social care research. It was established in 1996 and is funded by the National Institute for Health Research (NIHR). The advisory group's collaboration with researchers, research commissioners and the public aims to enable stakeholders in the health research system to support PPI in research. INVOLVE has 12 advisory group members, who are recruited every five years using publicly announced advertisements and interviews. They are selected based on their personal and professional expertise related to INVOLVE's main objectives and work (INVOLVE n.d.-b).

What does INVOLVE do?

INVOLVE actively encourages public involvement in research activities. Specific activities include the development of guidelines and strategies to support effective PPI; PPI support for research programmes, Research Design Services and Collaborations for Leadership in Applied Health Research and Care (CLAHRCs); and support for the development and review of plain English summaries for NIHR grant applications, as well as the development of criteria to evaluate the quality of the summaries. INVOLVE gathers and provides information on publications, completed research projects with PPI elements, and good practice examples (including information on impact from PPI) on its website in an Evidence Library, a Library of Research Projects and a Library of Examples (INVOLVE n.d.-b).

What enables INVOLVE'S activities?

INVOLVE'S guidance on co-producing research projects (INVOLVE & NIHR 2018) highlights general enablers of effective PPI, but not enablers of the INVOLVE advisory group's activities specifically. More general enablers include: establishing ground rules for all involved individuals at the start of the project, which should ensure that individuals' expectations, roles and

responsibilities are defined; open dialogue between all individuals involved; joint ownership of key decisions; a commitment to open and trusting relationships, and continuous reflection; support for opportunities for personal growth and development; flexibility throughout the project (e.g. changing the initial project plan); and valuing and evaluating the impacts emerging from research with PPI.

What are the challenges?

We did not identify literature on challenges specifically related to INVOLVE's activities. INVOLVE guidance documents (e.g. INVOLVE & NIHR, 2013a, 2018) highlight challenges related to public involvement more generally, including: managing power dynamics between researchers and patient and public contributors; allowing for the required flexibility in the design and implementation of research projects involving the public, given the way projects are usually funded and governed; finding ways of assessing and evaluating co-produced research and developing criteria for assessment and evaluation; and providing appropriate practical arrangements to facilitate involvement (INVOLVE & NIHR 2013a, 2018).

What is the evidence on impacts from INVOLVE's activities?

INVOLVE's Library of Examples includes a wide range of cases about PPI and its impact. Examples of reported impacts include: impact on the design of research funding applications (e.g. on the methods and research questions) to make them more robust and relevant to patient, public and healthcare service improvement needs; impact on research quality (with members of the public doing interviews in a way that gains participant trust and effective engagement with answering questions); impact on the number of people being reached (e.g. when recruiting participants for a survey); and impact on effective dissemination strategies and on the relevance and accessibility of the research (INVOLVE n.d.-a, n.d.-b; INVOLVE & NIHR 2013a, 2013b, 2014).

Sources: INVOLVE. n.d.-a. 'Examples of projects using social media for public involvement'. As of 11 June 2018:

<http://www.invo.org.uk/resource-centre/libraries/examples/examples-of-projects-using-social-media-for-public-involvement/> (54).

INVOLVE. n.d.-b. [Website]. As of 11 June 2018: <http://www.invo.org.uk/about-involve/>(26).

INVOLVE & NIHR. 2013a. *Exploring public involvement in research funding applications*. As of 11 June 2018:

http://www.invo.org.uk/wp-content/uploads/2014/10/INVOLVEExploringPublicInvolvementInResearchFundingApplications_linksSept14FINAL.pdf (55).

INVOLVE & NIHR. 2013b. *Exploring the impact of public involvement on the quality of research: examples*. As of 11 June 2018:

http://www.invo.org.uk/wp-content/uploads/2014/11/INVOLVEExploringImpactQualityResearchExamples_updated-July-2014.pdf (56).

INVOLVE & NIHR. 2014. *Examples of projects using social media for public involvement*. As of 11 June 2018:

<http://www.invo.org.uk/wp-content/uploads/2014/12/Social-media-guidance-FULL-examples.pdf> (57).

INVOLVE & NIHR. 2018. *Guidance on co-producing a research project*. As of 11 June 2018:

<http://www.invo.org.uk/posttypepublication/guidance-on-co-producing-a-research-project/> (42).

Box 8: People in Research**People in Research****What is People in Research?**

The website People in Research “helps research organisations and researchers find members of the public to get actively involved in their work by enabling them to advertise their opportunities” (NIHR n.d.). Opportunities for patient involvement can be published on the website by researchers which are then accessible to patients who can choose what they would like to be involved with. According to Minervation (2010), to ensure the opportunities are of high quality, they are checked by INVOLVE staff before becoming public.

What enables People in Research’s activities?

Patients can sort published opportunities by a particular subject, geographical location (or whether they are able to get involved from home) and if it is suitable for a beginner (NIHR, n.d.). This flexibility can encourage patients to be engaged with research as they are able to get involved to a degree they are able and willing to. New opportunities are emailed to patients and advertised on Twitter (NIHR n.d.).

The website is intended to be user-friendly as it was co-designed with the public (People in Research 2014).

What are the challenges?

No information could be found on the challenges associated with People in Research. However, although not mentioned in the literature, there is a little information about People in Research available online and our conversations with members of the patient and public community for other research we have been involved with suggest low but growing awareness.

What are the impacts?

Collecting the perspectives of a diverse set of patients enriches the research and allows researchers to look at questions from a different angle (Minervation 2010).

No specific impacts of People in Research could be found online.

Sources: People in Research. NIHR.

<https://www.nihr.ac.uk/patients-and-public/how-to-join-in/people-in-research.htm> (58)

Minervation. 2010. People in research: a new website connecting members of the public with researchers. <http://www.minervation.com/tag/people-in-research/> (59).

About this site. 2014. People in Research. <https://www.peopleinresearch.org/about/> (60).

Box 9: The James Lind Alliance**The James Lind Alliance****What is the James Lind Alliance?**

The James Lind Alliance (JLA), established in 2004, aims to give voice to patients, carers and clinicians to help set research priorities for a range of health conditions and health service areas (61, 62). It also aims to promote alignment between what is being studied in the research system and what patients and clinicians think should be researched (62).

How does the JLA engage patients and the public?

The JLA convenes priority-setting partnerships (PSPs) for different conditions. These bring together patients, carers and clinicians to identify areas of priority for research (62). These partnerships undertake large-scale data gathering, often through surveys, in which the JLA asks patients (both “experts” and lay patients) and clinicians what questions they have about research for a specific condition). The surveys most commonly include open-ended questions to allow respondents to have flexibility in their answers. Prioritisation can also occur through focus groups, Delphi techniques, expert panels, nominal group techniques, consensus-development conferences, electronic nominal groups, online voting and research agenda setting (61).

Patients are often contacted through social media or websites, frequently via Twitter (61) but also through established organisational networks, such as charities (The JLA Guidebook, 2018). Social media is particularly helpful in the early stages of engagement to contact a broader audience of individuals and groups (61). To ensure the most appropriate patient representatives are engaged, the JLA often reaches out to communities with specific conditions. One example of this is reaching out to individuals in care homes when investigating what research priorities should be in relation to those living with multiple conditions in older age. Clinicians are contacted through organisations such as charities and professional networks (62).

What enables patient engagement?

Motivations for engagement often include the personal experience of a health condition by patients, or the indirect experience of families and carers). Clinicians are often motivated by a clinical uncertainty that they wish to address.

The JLA is committed to a bespoke approach to enabling patient involvement. For example, the surveys and information sent to patients do not use technical language, and paper surveys can be used instead of online versions if patients do not have internet access (62). Patients attending meetings or events can be financially compensated for travel, accommodation and their time (62). Additionally, JLA advisors may have a call with patients before a meeting to answer any questions they have and ensure the venue is appropriate and accessible for those attending (62). Patients who are more severely ill can be involved with meetings via Skype.

What are the challenges?

- Accessing specific communities: It can be difficult to access particular patient communities, for example those from vulnerable groups (62). In addition, reaching out to these communities can increase the cost and time spent on a PSP (61, 62). It can also be difficult to communicate via email to engage certain groups, such as older patients.
- Sustained engagement: Competing demands on individuals’ time can make it difficult for them to be actively engaged in the process for a long period. For example, patients cannot always attend meetings during working hours.
- The dynamics of group discussions: Group discussions can face issues such as a dominant member persuading other individuals that their opinion is the right one, the dominance of conventionalism and communicating in a mixture of technical, lay and professional language (61).
- Difficulty in sustaining impact after the PSP has finished: The PSP is finished once there is no longer any funding and the groups no longer meet. This leads to uncertainty as to whether the priorities identified are being acted on in research and whether the priorities are suitable for research at all (61).

What are the impacts?

Once PSPs are completed, the JLA disseminates the work done and methods used in journal articles (63). Some PSPs have reported on the impact of the work at a later stage as measuring impact can be a challenge and the effects of PSPs may need a longer period to emerge (63, 64). In general, patients report that they feel empowered after they have been involved in PSPs, and feel fulfilled due to having contributed to something worthwhile). There is also some evidence to suggest that clinicians have in some instances changed the way they practice medicine after collaborating with patients in a PSP context).

More specifically, the JLA has impacted funded research. For example, asthma patients wanted more non-drug treatments to be made available to them, particularly breathing exercises. This resulted in research exploring the ability to provide physiotherapy through digital methods, such as videos, which were found to be just as beneficial as face-to-face physiotherapy. Digital provision of asthma physiotherapy is now being implemented (65). After priorities have been identified, some organisations provide additional funding to support research into the specific areas, such as Coeliac UK who launched a call for research in May 2018 stating that they would favour applications related to the PSP priorities (62).

Sources: The James Lind Alliance Guidebook. 2018.

<http://www.jla.nihr.ac.uk/jla-guidebook/downloads/Print-JLA-guidebook-version-7-March-2018.pdf> (62).

Reporting on results and impact. The James Lind Alliance (a).

<http://www.jla.nihr.ac.uk/making-a-difference/reporting-on-results-and-impact.html> (63)

Making a difference. The James Lind Alliance (b).

<http://www.jla.nihr.ac.uk/making-a-difference/> (64).

Funded research. The James Lind Alliance (c).

<http://www.jla.nihr.ac.uk/making-a-difference/funded-research.html> (66)

Working with the priorities. 2018. The James Lind Alliance.

<http://www.jla.nihr.ac.uk/making-a-difference/working-with-the-priorities.htm> (67).

Madden & Morley. 2016. Exploring the challenge of health research priority setting in partnership: reflections on the methodology used by the James Lind Alliance Pressure Ulcer Priority Setting Partnership. *Research Involvement and Engagement*, 2(12). (61).

Box 10: An advisory group in mental health nursing research

SUGAR: A collaborative group of service users and carers involved with mental health nursing research

What was the research project or initiative about?

Researchers at City University in London, mental health service users, carers and healthcare practitioners developed a group in 2009 – SUGAR: Service Users and Carers Group Advising on Research – to enable collaborative working in mental health research. It seeks to increase the involvement of those with lived experiences of using mental health services in research and to build long-term relationships between researchers and local community members.

How were patients and the public involved?

The group consists of 13 service users and carers who were recruited through the local mental health service in East London and includes members of different ages, genders, sexualities and ethnic profiles. The SUGAR group is still active and meets monthly. Meetings are facilitated by researchers at City University. Service users are consulted on diverse issues spanning identification of research topics and questions to explore, assistance with the design of patient information leaflets for projects, assistance with research ethics issues and participant recruitment, assistance with the design of studies and protocol piloting, data collection, contribution to writing reports and dissemination, and engagement with the implementation of results. According to Simpson et al. (2014), at least 11 SUGAR group members and at least 3 researchers attended each meeting during and prior to 2014. SUGAR group members also wrote reflexive pieces about their involvement for a conference in 2012 and these insights were analysed to understand the diversity of service user experiences, impacts and opportunities for improvement.

What enabled engagement?

A job description and person specification was used to facilitate recruitment. The functioning of the group was supported through NIHR funding for five years, allowing for continual engagement with multiple mental health nursing research projects. Since October 2014, SUGAR has been jointly supported by the School of Health Sciences, City University, and the East London NHS Foundation Trust. Service users were given honorary university contracts and access to the library and other university resources (such as computing systems and facilities), as well as being remunerated for their involvement. They were also provided with training and ongoing support for delivery on their roles (e.g. on how to do literature reviews, ethics and governance, research roles and responsibilities, information technology, presenting and writing results). In general, a friendly and welcoming environment was seen to facilitate effective collaboration between service users and researchers.

What were the challenges?

According to Simpson et al. (2014), service users occasionally found it difficult to understand complex research ideas. Ill health also impeded engagement. Challenges related to interpersonal relationships were sometimes experienced, such as carers occasionally feeling that their views were given less weight than the views of patients, or differences of opinion between individuals with strong viewpoints. Practical challenges related to timekeeping also occurred from time to time.

Impacts

The researchers and members of the SUGAR group identified a range of benefits from collaboration. These include impacts in terms of improved research relevance and quality, facilitated by the inclusion of patient and carer perspectives and lived experiences in research. They also include personal learning and development gains for service users (e.g. about mental health-related issues, research skills, communication skills and effective influencing skills). Simpson et al. highlight that the impacts of some engagements are not yet observable (at the time the paper was written), but do not report on any negative impacts.

Source: Simpson, A., et al. (2014). "Adding SUGAR: service user and carer collaboration in mental health nursing research." *J Psychosoc Nurs Ment Health Serv* 52(1): 22–30 (68).

Box 11: A service user and carer panel for cancer and palliative research

The North Trent Cancer Research Network Consumer Research Panel

What was the research project or initiative about?

The North Trent Cancer Research Network Consumer Research Panel (NTCRN CRP) was established in 2001 to encourage and enable cancer and palliative care service users and carers to engage with research.

How were patients and the public involved?

Collins et al. (2015) reported that the panel consisted of 38 current or former cancer and palliative care service users and carers of diverse ages, with experience of diverse types of cancers and stages of disease, and from different socioeconomic and demographic backgrounds. Service users and carers are involved in influencing research agendas and contributing to the research process – in generating research questions, assisting with protocol development and advising on ethics and participant recruitment, as well as actively engaging as co-researchers in data collection, analysis or interpretation, presenting at conferences and co-authoring papers. Further detail on the operational model for engagement is not discussed.

What enabled engagement?

According to Collins et al. (2015), formal support for NTCRN CRP from established academic organisations (the Academic Unit of Supportive Care at the University of Sheffield Medical School and the Centre for Health and Social Care Research at Sheffield Hallam University) has helped nurture researcher engagement with the network, from both biomedical and health services research staff. A policy environment promoting engagement, as seen in the establishment and evolution of organisations and networks like INVOLVE, the NIHR's commitment to patient and public engagement in research, has helped put the spotlight on the potential value of patient and public engagement more generally and supported scale-up and growth of the model pursued by the NTCRN CRP.

What were the challenges?

At the time of this publication, the group faced the following challenges:

- A lack of sufficient funding levels to support engagement activities;
- Competing demands on the time of service users, carers and researchers who facilitated engagement;
- Tensions between different stakeholder groups, relating to: (i) researcher concerns about the service users' and carers' understanding of biomedical and pre-clinical research, and the impact this could have on their ability to engage effectively; (ii) service user concerns about the use of unfamiliar language and jargon within research meetings; (iii) difficulties around service users integrating effectively due to a lack of clarity on roles; and (iv) researcher reservations about the objectivity and representativeness of the inputs that service users and carers can provide.

Impacts

Despite the challenges, Collins et al. (2015) report positive impacts on the identification and prioritisation of research topics that matter to patients, on the relevance and accessibility of research questions and research protocols, and on fostering links with communities. Although the authors identify a lack of critical evaluation of PPI engagement with research in the wider knowledge base, they do not identify any negative impacts from the NTCRN CRP case.

Sources: Collins, K., et al. (2015). "Making patient and public involvement in cancer and palliative research a reality: academic support is vital for success." *BMJ Support Palliat Care* 5(2): 203–206 (69).

North Trent Cancer Research Network Consumer Research Panel. 2018. Sheffield Hallam University. As of 7 June 2018:

<https://www.shu.ac.uk/research/specialisms/health-and-social-care-research/what-we-do/our-expertise/health-care-and-service-delivery-research/case-studies/north-trent-cancer-research-network-consumer-research-panel> (70).

Box 12: Reflexivity as a mechanism for enabling patient contribution to research: using video-reflexive ethnography

Video-reflexive ethnography as an enabler of high-quality and relevant research through collaboration between those delivering and those receiving care

What was the research project or initiative about?

Video-reflexive ethnography is the reflexive viewing of filmed work practices or behaviours and relationships (Idema et al. 2013, cf. Collier and Wyer 2016). Collier and Wyer (2016) share their experiences of using video-reflexive ethnography as a research method that enables those delivering care and those receiving it to collaborate in research. For example, the technique can enable service users, healthcare professionals and academic researchers to jointly address the complexity of their relationships and practices. It can allow for more nuanced insights to surface, facilitate analytical rigour and integrate richer evidence into identifying opportunities for service improvement. The authors discuss learning from the application of this method to two studies: one considering the relationships between the end-of-life care setting and the quality and safety of care patients receive, and the other investigating the role that patients can play in infection prevention and control in surgical units.

How were patients and the public involved?

Each study included engagement of patients, family members and healthcare professionals. Patients could participate in diverse ways such as being filmed as part of a situation or interaction, participating in an audio or video-recorded interview, collecting video footage, participating in video-reflexive sessions to watch the footage and discuss it, and feeding back insights to clinicians in focus groups.

What enabled engagement?

A number of enablers were identified by the authors, such as (i) a welcoming attitude to patient engagement, recognising their expertise and the value they can add; (ii) giving patients choice and autonomy in deciding whether and how they would like to engage; (iii) getting to know the patients prior to the research and building trusting relationships; and (iv) being flexible and adaptable (for example to patient requests about how the data could be used, to whether they wanted to view their own footage or not, about the extent of their engagement as active collaborators rather than participants in the research).

What were the challenges?

Four main challenges were identified. Firstly, the technique can make it challenging to manage the boundaries between research and clinical practice and can introduce tensions in the

patient-healthcare professional relationship. For example, such tensions can arise if patients (based on their engagement with the research and reflection on video footage) request specific changes in practice which cannot be delivered (either for practical reasons or if health professionals do not see them as the appropriate course of action), or if health professionals take offense at feedback rather than seeing it as a learning opportunity. Secondly, video-reflexive ethnography can expose individuals to aspects of themselves that they might not otherwise be aware of and can introduce vulnerabilities. Thirdly, power imbalances or potential for bias can be introduced by those who control the camera or footage. Finally, although not explicitly identified as a challenge in the paper, there is reference to the potential for individuals to consciously or unconsciously alter their behaviour if they know they are being filmed.

Impacts

The research collaboration between service users and their families and healthcare professionals enabled by this research technique enriched research insights and provided novel learning about more patient-centred opportunities for improving the quality and safety of care.

Source: Collier, A. and M. Wyer (2016). "Researching Reflexively With Patients and Families: Two Studies Using Video-Reflexive Ethnography to Collaborate With Patients and Families in Patient Safety Research." *Qual Health Res* 26(7): 979–993 (40).

Box 13: Enabling patient and public involvement in research through a network approach

Involving patients and the public in cancer research – the Thames Valley Cancer Network Consumer Research Partnership

What was the research project or initiative about?

The aim of this study was to investigate different methods that could be used to involve patients, carers, relatives or members of the public who have been affected by cancer in cancer research in the Thames Valley. The study explores the role of the Thames Valley Cancer Network Consumer Research Partnership, which aims to promote the involvement in cancer research of patients and those affected by cancer. The group was established in 2009 and had a membership of around 25, including consumers and professionals. The study evaluates the group's activity and consumer involvement in 15 projects. The 15 projects consisted of 8 clinical trials, 3 qualitative research projects, 2 patient survey projects and 2 non-randomised interventional studies.

How were patients and the public involved?

Consumers had diverse roles across the 15 projects. These varied from being consulted on key documents such as patient information sheets to collaborative roles in jointly undertaking research with other stakeholders. Some projects involved consumers very early on, including in proposal development and grant applications.

Other types of involvement included designing questionnaires, advising on how researchers should conduct patient interviews, advising on the content and language of patient information leaflets, facilitating recruitment, and being involved in research management groups.

What enabled engagement?

Although Arain et al. (2015) do not specifically identify enablers, the study notes that nine of the projects had funds to cover the cost of travel or other expenses incurred by the consumers.

Members of the Research Partnership group were given basic training in research via a Getting Involved and Influencing Cancer Research workshop. In addition to this, four projects also offered project-specific training which provided participants with more information about the specific research topic.

What were the challenges?

Arain et al. (2015) do not discuss any challenges related to the Thames Valley Cancer Network Consumer Research Partnership specifically.

Impacts

The impacts of consumer involvement on the individual projects studied are not discussed and Arain et al. (2015) note that many of the impacts can only become observable in the longer term. However, the authors highlight more general benefits of the network, such as enabling research that is more relevant to the public.

Source: Arain, M., et al. (2015). "Consumer involvement in cancer research: example from a Cancer Network." *Health Expectations*, 18, pp.1530–1542.(71).

Box 14: Enabling patient and public involvement in research through a network approach

Embedding patient and public involvement within research – how to set up a research patient ambassador group within an NHS trust

What was the research project or initiative about?

The authors focus on how a specific patient group, the Clinical Research Ambassador Group (CRAG), was established. This group is within the Heart of England NHS Foundation Trust and aims to provide guidance for other researchers intending to create their own group.

How were patients and the public involved?

The CRAG was designed to make sure that PPI was not a token effort to fulfil funding criteria, and to ensure that patients and the public who were members felt that they benefited from participation in the group. The main role of the group, however, was to assist specifically with the recruitment of individuals from the public that are served by the Heart of England NHS Foundation Trust for clinical research. CRAG members were involved in deciding the name of the group and members have roles ranging from providing feedback on documentation to being more involved with the research team.

What enabled engagement?

The authors highlight that members of the CRAG preferred being identified as ambassadors and part of an ambassador group rather than a PPI group. The role of an "ambassador" was seen to be more meaningful. Recruitment of participants was assisted through a launch event in the community. Forty-nine people attended the open evening for the CRAG, with 28 of those becoming members of the group. Members were then able to choose how involved they wanted to be in research projects. This could range from making comments on documents to being involved with the research team. Activities of the group were communicated via diverse channels, allowing for participants to be kept up to date with developments and facilitating retention of motivated participants. There is communication with members via email and Twitter and the

profile of the group has been raised by publication of information booklets and articles, as well as newsletters, on the INVOLVE website and in Anaesthesia News. The group also had coffee mornings every three months which enabled informal opportunities for discussions between researchers and members of the group. Education sessions were also run during these meetings with the aim of increasing knowledge about clinical research among members. CRAG members were involved in developing patient information documents, leading to more understandable and coherent information and facilitating more effective patient engagement in research. The authors highlighted that involving members of the CRAG led to more successful interviews with patients.

What were the challenges?

One challenge was to ensure representativeness. Although the CRAG has successfully retained membership and increased the number of members, it was highlighted that the group must continue to aim to represent the diverse nature of the patients that the Heart of England NHS Foundation Trust serves. In particular, the authors suggest that the group should recruit more ethnic minority members in order to better reflect the population of Birmingham. At the time of the study, the group also wanted to recruit more people from younger generations.

Impacts

Researchers highlighted that involving patients made their research projects more credible (but did not specify who perceived this greater credibility). The involvement of patients in reviewing documents contributed to making patient information more understandable to a lay audience.

Source: Skilton, E., et al. (2016). "Embedding patient and public involvement within research – How to set up a research patient ambassador group within a NHS trust." *Journal of the Intensive Care Society*; 17(3) 234–237 (72).

Appendix B. Study design and methods

Study design

To address the questions set out above, we conducted a review of relevant literature alongside interviews with a range of individuals with expertise in the field. Our literature review approach followed the principles of rapid evidence assessment (REA) (73). This approach is consistent with the principles underlying systematic review methodology (having clearly defined research questions, systematic and replicable search strategies, and explicit inclusion and exclusion criteria), but makes some allowances for the rapid delivery of information as required, by restricting the scope and coverage of the review to focus on the most relevant literature.

The ultimate aim of the review was to provide practical insights to inform THIS Institute's evolving engagement strategy. Consequently, the review was inclusive with respect to article type; we did not exclude articles based on methodology and we undertook only a limited assessment of the quality of the articles reviewed (noting limitations of the studies reviewed and considering these in the synthesis of review findings). We sought to undertake a narrative synthesis based on the research questions addressed through the REA, incorporating insights based on interview findings. We also aimed to provide real-world examples of how different approaches to Patient and Public Involvement (PPI) have been

implemented, through the presentation of case studies sourced from the literature.

Rapid evidence assessment

Search strategy

An initial trial of search terms was conducted by two researchers to refine search terms and scope based on the quantity of relevant literature identified. This was an iterative process whereby searches were run and the results obtained were reviewed to ensure relevant articles were being captured without superfluous material being included. After several rounds of testing, a final set of search terms was established and then applied consistently to enable reproducibility of the search approach. The refined search terms were applied to the PubMed and Scopus databases on 5 April 2018, covering the period 2000–present for review articles and 2013–present for original articles. The search terms used are presented in Table 12 (for review articles) and Table 13 (for original articles). Due to the high number of articles identified through initial iterations of the search, restrictions were added to the search with respect to date and geographical location (for original articles only, limiting the search to literature focusing on the UK, US, Canada and Australia) in order to reduce the number of articles for screening to a manageable number.

Table 12: Search terms for rapid evidence assessment – review articles

PubMed English; 2000–present; reviews
Health services[MeSH] OR Healthcare[MeSH] OR "health research"[title/abstract] OR "clinical research"[title/abstract] AND
(patient*[Title] OR carer*[Title] OR caretaker*[Title] OR user*[Title] OR citizen*[Title] OR consumer*[Title] OR community*[Title] OR public*[Title]) AND
((Research*[title] OR Study[title] OR studies[title] OR review*[title] OR "evidence synthesis"[title] OR evaluat*[title] OR trial[title] OR "randomised controlled trial"[title] OR "randomized controlled trial"[title] OR RCT[title]) AND
(participat*[title] OR involve*[title] OR engage*[title] OR contribut*[title] OR design*[title] OR codesign*[title] OR co-design*[title] OR articulat*[title] OR specification*[title] OR priorit*[title] OR conduct*[title] OR develop*[title] OR co-produc*[title] OR "idea generation"[title] OR implement*[title] OR activit*[title] OR collab*[title] OR partner*[title])) (OR "citizen science" OR "citizen-science" OR (crowd[title/abstract] AND source[title/abstract]) OR crowdsource*)
Scopus English; 2000–present; reviews
(TITLE-ABS ("health services" OR healthcare)) AND
(TITLE (patient* OR carer* OR caretaker* OR user* OR citizen* OR consumer* OR community* OR public*)) AND AND
((TITLE (research* OR study OR studies OR review* OR "evidence synthesis" OR evaluat* OR trial OR "randomised controlled trial" OR "randomized controlled trial" OR RCT)) AND
(TITLE (participat* OR involve* OR engage* OR contribut* OR design* OR codesign* OR co-design* OR articulat* OR specification* OR priorit* OR conduct* OR develop* OR co-produc* OR "idea generation" OR implement* OR activit* OR collab* OR partner*)) OR TITLE-ABS ("citizen science" OR "citizen-science" OR "crowd source" OR crowdsource*)

Table 13: Search terms for rapid evidence assessment – original articles

PubMed English; Last 5 years (+ geographic limitations) (removed review, comment, letter, editorial)
Health services[MeSH] OR Healthcare[MeSH] OR "health research"[title/abstract] OR "clinical research"[title/abstract] AND
(patient*[Title] OR carer*[Title] OR caretaker*[Title] OR user*[Title] OR citizen*[Title] OR consumer*[Title] OR community*[Title] OR public*[Title]) AND
Research*[title] OR Study[title] OR studies[title] OR review*[title] OR "evidence synthesis*" [title] OR evaluat*[title] OR trial[title] OR "randomised controlled trial"[title] OR "randomized controlled trial"[title] OR RCT[title] AND
(participat*[title] OR involve*[title] OR engage*[title] OR contribut*[title] OR design*[title] OR codesign*[title] OR co-design*[title] OR articulat*[title] OR specification*[title] OR priorit*[title] OR conduct*[title] OR develop*[title] OR co-produc*[title] OR "idea generation"[title] OR implement*[title] OR activit*[title] OR collab*[title] OR partner*[title])) (OR "citizen science" OR "citizen-science" OR (crowd[title/abstract] AND source[title/abstract]) OR crowdsource*)
Scopus English; 2013–present; Article (+ geographic limitations)
((TITLE-ABS ("health services" OR healthcare)) OR (TITLE-ABS ("health research" OR "Clinical research"))) AND
(TITLE (patient* OR carer* OR caretaker* OR user* OR citizen* OR consumer* OR community* OR public*)) AND
((TITLE (research* OR study OR studies OR review* OR "evidence synthesis" OR evaluat* OR trail OR "randomised controlled trial" OR "randomized controlled trial" OR RCT)) AND
(TITLE (participat* OR involve* OR engage* OR contribut* OR design* OR codesign* OR co-design* OR articulat* OR specification* OR priorit* OR conduct* OR develop* OR co-produc* OR "idea generation" OR implement* OR activit* OR collab* OR partner*)) OR TITLE-ABS ("citizen science" OR "citizen-science" OR "crowd source" OR crowdsource*)

In addition to academic literature, we also searched for grey literature using terms relating to PPI (using Google) between 5 April 2018 and 13 June 2018, and conducted a targeted search of the websites of organisations involved in patient and public engagement activities.

Study selection

Records identified by the searches were assessed for inclusion by screening titles and abstracts against a set of inclusion and exclusion criteria (Section B.2.3). At this stage, studies were deliberately retained if there was any uncertainty as to their relevance. Screening was conducted by one researcher in the first instance; cases of uncertainty were set aside

and screened by a second reviewer. Full-text screening of potentially eligible articles was undertaken as part of the data extraction stage (see below), during which studies were screened against the same inclusion and exclusion criteria, based on the more detailed information available through full-text review.

In addition, a “snowballing” approach was used to identify additional studies for screening and potential inclusion. In this approach, the reference lists of relevant studies were reviewed for further potentially relevant studies, which were then screened in the same way. For key articles we also tracked citations to identify recent related publications.

Inclusion and exclusion criteria

Initially, any type of article published in English that examined the topics described in Section

B.2 above was eligible for inclusion (from 2000 onwards for review articles and from 2013 for original articles). We did not exclude literature based on methodology or country setting. Articles captured by the search included two distinct types of relevant article: those that focused explicitly on PPI as the topic (referred to as “core articles”) and those that described a specific example of PPI or PPI-related activities in action (e.g. within a particular research project or organisation). Due to the large number of articles identified in both categories during initial title and abstract screening, additional exclusion criteria were applied during a second round of screening; these further refined the topic focus and added date restrictions. Table 14 and Table 15 summarise the inclusion and exclusion criteria used in the first and second rounds of screening, respectively.

Table 14: Inclusion and exclusion criteria: Round 1 screening

Round 1 – Title and abstract screening	Inclusion	Exclusion
Topic	Articles which: <ul style="list-style-type: none"> • Focus on the topic of methods or approaches for the engagement of patients or the public in the prioritisation, design or conduct of health research (or evaluations of healthcare or improvement interventions); • Describe challenges and enablers to patient or public engagement; • Provide insights on the impact of patient or public engagement, including insights on advantages and disadvantages; • Provide insights on the evaluation of patient or public engagement. 	Articles which: <ul style="list-style-type: none"> • Report patient or public involvement only as participants in research (rather than actively engaging in the process of informing design, conduct or priority setting); • Report on research outside the health sphere; • Focus on PPI involvement in priority setting for health services (not research) or service design; • Focus on patient or public involvement in healthcare service decision making.
Language	English	Any language other than English
Country setting	Any country	None
Document type	• Any type of publication (including commentaries, editorials or opinion pieces) where the assertions are based on empirical evidence or practical experience.	<ul style="list-style-type: none"> • Commentaries, editorials or opinion pieces without direct reference to empirical evidence or practical experience. • Conference abstracts.
Date of publication	From 2000	Before 2000

Table 15: Additional inclusion and exclusion criteria: Round 2 screening

Round 2 – Title and abstract screening	Inclusion	Exclusion
Topic	As for Round 1	Any articles which: <ul style="list-style-type: none"> • Focus on community engagement in a public health or health promotion setting; • Focus <i>solely</i> on the history and philosophy of PPI or the ethics of PPI, or on the quality of PPI reporting only (we did include articles which cover these issues in addition to the core topics of our focus); • Review articles which focus on biomedical research, or drug or medical device development. • Review articles focused on specific conditions (but included case studies where relevant).
Language	English	Any language other than English
Country setting	Any country	None
Document type	<ul style="list-style-type: none"> • Any type of publication (including commentaries, editorials or opinion pieces) where the assertions are based on empirical evidence or practical experience. 	<ul style="list-style-type: none"> • Commentaries, editorials or opinion pieces without direct reference to empirical evidence or practical experience. • Conference abstracts.
Date of publication	<ul style="list-style-type: none"> • From 2008 for review articles (10 years). • From 2013 for primary studies (5 years). 	<ul style="list-style-type: none"> • Before 2008 for review articles (10 years). • Before 2013 for primary studies (5 years).

Articles were not excluded on the basis of quality and no formal assessment of quality was undertaken as part of the review. However, the included studies reported on a range of limitations which were considered as part of the review process.

Extraction and synthesis

During this stage, data were extracted from studies identified as eligible using an Excel template. References were managed using Endnote (version 8). Guided by our research questions, data were extracted on the following: article type; group discussed in the context of PPI engagement; study type (including summary study aims, objectives, design, headline findings); stage of the research process in which PPI took place; how PPI

contributors were engaged and what they did; motivations for engagement; impacts of PPI engagement on the research/study/evaluation; enablers/facilitators of PPI engagement; challenges/barriers to PPI engagement; how engagement of PPI could be improved in the future; evaluating PPI engagement; limitations of the study.

Data extraction was undertaken by four researchers (AH, AC, SB and SM). We synthesised the evidence available in relation to each of our research questions and identified additional themes arising from the literature that we considered potentially important in the context of informing the development of strategy for THIS Institute.

Overview of identified literature

Our searches of the academic literature identified 1,825 articles, reports and commentaries (1,697 identified through database searches, and a further 128 from other sources, including a separate search of the journal *Research Involvement and Engagement*, and articles identified through a parallel review on NHS staff engagement in research). A total of 127 articles were selected for further review on the basis of two rounds of title and abstract screening using the inclusion and exclusion criteria set out above. These articles fell into two categories: “core” articles describing research on the topic of public and patient involvement in health research (75 articles) and “case” articles that described public and patient involvement in the context of a particular project or organisation (52 articles).

In light of the extent of the relevant literature, and given the available resources and timescale allocated for the review, further strategies were employed to limit the number of articles included for full-text review. With respect to the “core articles”, one researcher first examined the reference lists of the review articles identified, beginning with the most recent reviews. Earlier reviews and original articles were then selected for full-text review if they fell outside the time period covered by the review or were relevant but fell outside their scope. Thirty-one of the 75 “core” articles identified were included on this basis. In addition, abstracts of the 52 “case” articles were reviewed to assess relevance with respect to THIS Institute’s particular areas of interest. The most relevant articles (10) were selected for inclusion. In addition further articles were identified for full-text review through snowballing from reference lists (10 articles: 8 core articles and 2 case study papers) and through direct recommendations from THIS Institute (2 core articles). In total there were 41 “core” articles and 12 “case” articles included based on the review of academic literature.

A search of grey literature was also conducted, which identified a further 18 sources. In total, 71 sources were included in the review.

Interviews with experts on public and patient participation

In order to add depth and nuance to the findings from the literature review, semi-structured telephone interviews were undertaken with a range of experts identified via the professional networks of those commissioning and conducting the review. Interviews lasted between 30 and 45 minutes and took the form of a guided conversation using a semi-structured interview guide.

The topics covered included:

- Views and experiences on the most meaningful and most feasible approaches and methods for engaging patients and the public in research;
- Views on key enablers and challenges (including ways of overcoming challenges);
- Key motivations and incentives for engagement (and associated practical means through which these motivations and incentives can be deployed), including variation across different types of patient and public profiles;
- Examples of particularly relevant or successful programmes or initiatives;
- Views on how THIS Institute might take its engagement strategy forward.

With the participant’s consent, interviews were audio-recorded for the purpose of writing up accurate notes on the interview. Interviews were analysed thematically according to the questions explored by the researcher conducting the interview. Findings were discussed between team members in terms of their relationship to insights from the literature review.