STUDY PROTOCOL

Stakeholder's perspectives and experiences of patient and public involvement (PPI) in clinical trials in maternal and neonatal healthcare: protocol for a qualitative evidence synthesis

[version 1; peer review: 1 approved, 1 approved with reservations]

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Abstract

Background: Patient and public involvement (PPI) has the potential to improve the relevance of trial outcomes and improve participant recruitment within clinical trials. However, the literature on PPI approaches, outcomes, and attitudes towards PPI in specific clinical research areas is limited. We are interested to know the current approaches to and views of PPI within maternal and neonatal clinical trials, from the perspective and experience of involved stakeholders.
Methods: A qualitative evidence synthesis (QES) of stakeholders' perspectives and experiences of PPI will be conducted. Stakeholders will include any individual involved in maternal or neonatal clinical trials with experience of PPI in the area or who expresses their views on PPI. The electronic bibliographic databases CINAHL, MEDLINE, PsycINFO, EMBASE, Web of Science and the Maternity and Infant Care (OVID) will be searched from inception. Qualitative studies, mixed-methods studies where the qualitative data can be extracted independently, and surveys with open-ended qualitative questions, will be included.
Aims: The QES seeks to explore stakeholders', including PPI contributors, trial participants and guardians, and trial researchers, perspectives and experiences of PPI in maternal and neonatal clinical trials.
Discussion: THE QES will provide an understanding of how PPI is understood, operationalised and experienced by stakeholders in maternal and neonatal clinical trials, with the aim of identifying good practice and areas for improvement.

Open Peer Review

Approval Status ✓ ✓

	1	2
version 2 (revision) 14 Sep 2023		✓ view
version 1 12 May 2023	✓ view	? view

1. **julie abayomi**, Edge Hill University, Ormskirk, UK

2. **Sandy Oliver**, University College London, London, UK

Any reports and responses or comments on the article can be found at the end of the article.

PROSPERO registration: CRD42023383878 (2nd March 2023)

Keywords
patient and public involvement, trial methodology, neonatal care, maternal care, clinical trials, GRIPP2

This article is included in the HRB-TMRN gateway.

Corresponding author: Kathleen Hannon (kahannon@tcd.ie)

Author roles: Hannon K: Conceptualization, Methodology, Project Administration, Writing – Original Draft Preparation, Writing – Review & Editing; **Eustace-Cook J**: Methodology; **Daly D**: Methodology, Writing – Review & Editing; **Smith V**: Methodology, Writing – Review & Editing

Competing interests: No competing interests were disclosed.

Grant information: Funded by the Health Research Board – Trials Methodology Research Network in Ireland (grant ref: HRB-TMRN-2021-001) and the School of Nursing and Midwifery, Trinity College Dublin.
The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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Abstract

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Methods: A qualitative evidence synthesis (QES) of stakeholders' perspectives and experiences of PPI will be conducted. Stakeholders will include any individual involved in maternal or neonatal clinical trials with experience of PPI in the area or who expresses their views on PPI. The electronic bibliographic databases CINAHL, MEDLINE, PsycINFO, EMBASE, Web of Science and the Maternity and Infant Care (OVID) will be searched from inception. Qualitative studies, mixed-methods studies where the qualitative data can be extracted independently, and surveys with open-ended qualitative questions, will be included.
Aims: The QES seeks to explore stakeholders', including PPI contributors, trial participants and guardians, and trial researchers, perspectives and experiences of PPI in maternal and neonatal clinical trials.
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PROSPERO registration: CRD42023383878 (2nd March 2023)

Open Peer Review

Approval Status *AWAITING PEER REVIEW*

Any reports and responses or comments on the article can be found at the end of the article.

Keywords
patient and public involvement, trial methodology, neonatal care, maternal care, clinical trials, GRIPP2

This article is included in the HRB-TMRN gateway.

Corresponding author: Kathleen Hannon (kahannon@tcd.ie)

Author roles: Hannon K: Conceptualization, Methodology, Project Administration, Writing – Original Draft Preparation, Writing – Review & Editing; **Eustace-Cook J**: Methodology; **Daly D**: Methodology, Writing – Review & Editing; **Smith V**: Methodology, Writing – Review & Editing

Competing interests: No competing interests were disclosed.

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First published: N/A, **N/A**: N/A N/A

Introduction

'Patient and public involvement' (PPI) is defined as research being conducted *with* and *by* patients or members of the public, as opposed to research being solely conducted *on* these groups (Dudley *et al.*, 2015b; Staley, 2009). The terminology of 'PPI' varies across countries, sometimes being referred to, for example, as 'consumer engagement' or 'public participation' (Dudley *et al.*, 2015a; Molloy *et al.*, 2019).

The inclusion of PPI in research is advocated for on pragmatic and ethical grounds (Bagley *et al.*, 2016); pragmatically in that PPI has the potential to produce more relevant research (Bagley *et al.*, 2016) and ethically by the belief that those affected by a condition or disease should be involved in research that impacts them (Crocker *et al.*, 2015). Involving patients, their families, or other members of the public, can help identify critical gaps in the research (EFCNI, 2017), leading to improvements in what type of research is conducted and how.

While PPI is increasingly encouraged by health research funders (Dudley *et al.*, 2015b; HRB, 2022), discrepancies can arise between the PPI activities detailed within funding applications and its actual implementation (Buck *et al.*, 2014). Involvement can be tokenistic, particularly if PPI inclusion is viewed solely as a means to secure grant funding (Dudley *et al.*, 2015b).

This qualitative evidence synthesis (QES) is being conducted as part of a doctoral study on PPI activity within maternal and neonatal clinical trials. While studies have been conducted on public contributors' and researchers' experiences of PPI within clinical trials (Selman *et al.*, 2021; South *et al.*, 2016), to our knowledge, a QES specifically on stakeholders' perspectives of PPI within maternal or neonatal clinical trials has not been conducted. The objective of this QES is to synthesise the evidence of stakeholders' perceptions of PPI in maternal and neonatal clinical trials.

Protocol

Inclusion criteria

The SPIDER tool was used to define the inclusion criteria for this QES (Cooke *et al.*, 2012).

- Sample: Stakeholders in maternal or neonatal clinical trials who have experience of PPI or have expressed their views on PPI. These may include, although not necessarily limited to, trial participants, PPI contributors, and any member of the trial research team (e.g., principal investigators, research midwives/nurses, trial managers, and statisticians).
- Phenomenon of Interest: PPI recruitment and PPI activity at any stage of a maternal or neonatal clinical trial.
- Design: Published and unpublished primary qualitative research of any design.
- Evaluation: Themes representative of stakeholders' perceptions and experiences of PPI in maternal and neonatal clinical trials.
- Research type: Qualitative research and mixed-methods research where qualitative data can be extracted separately for analysis, quantitative surveys with qualitative open-ended questions and data that can be extracted independently and that have been thematically analysed.

For this QES, clinical trials are, as defined by the World Health Organization (WHO), 'any research study that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects on health outcomes' (WHO, 2023). Maternal clinical trials are defined as clinical trials in maternal health, involving trial participants at any stage of pregnancy and up to 6 weeks postpartum. Neonatal trials are defined as clinical trials conducted with neonates during the first 28 days of life. 'PPI contributor' refers to a patient or lay individual who is involved at any stage of the trial process, from trial conception to trial dissemination.

Search strategy

With the assistance of a subject librarian, a search strategy of index terms and keywords was developed from two key concepts; clinical trials in maternal and neonatal health, and patient and public involvement (Table 1). The electronic bibliographic databases CINAHL, MEDLINE, PsycINFO, EMBASE, Web of Science, and Maternity and Infant Care (OVID) will be searched, with the search terms adapted for each database. Grey literature sources, such as GreyNet, will also be searched.

No restrictions on language or date will be applied; that is the databases will be searched from their date of inception to the date of the search. Although studies in any language will be included in the search, only English language studies will be included in the QES. Searching all languages, however, will enable the reviewers to determine how many studies may be excluded based on language, and thus assess the extent of language bias as a potential limitation of the QES.

All retrieved citations will be exported to Endnote (version 20.5) where duplicates will be removed before the citations are transferred to Covidence for screening. Screening will take place in two stages; title and abstract screening, and full text review. First, title and abstracts will be screened against the inclusion criteria by one reviewer (KH). A second reviewer (VS or DD) will screen 20% of the records on title and abstract, and the reviewers' assessments will be cross-checked for congruency. If reviewer congruency is <80%, based on AMSTAR 2's recommendation that at least 80% agreement should be achieved (Shea *et al.*, 2007), all citations will be screened independently by two reviewers. Full text of records will be sourced and each record will be screened for inclusion/exclusion by at least two reviewers working independently. Any disagreements will be resolved through discussion until consensus is achieved. A record will be retained of reasons for exclusion of papers at full text review and reported in the final QES in a PRISMA flow diagram (Page *et al.*, 2021).

Table 1. Search strategy.

Concept	Index terms and keywords
Concept 1: Patient and public involvement	Index Terms: EMBASE: 'patient participation'/exp OR 'stakeholder engagement'/exp OR 'patient and public involvement'/exp CINAHL: (MH "Consumer Participation") MEDLINE (EBSCO): (MH "Patient Participation") OR (MH "Community Participation+") PsycInfo: DE "Community Involvement" OR DE "Client Participation" Keywords: (public* OR citizen* OR patient* OR user* OR "service user*" OR client* OR "care giver*" OR caregiver* OR carer* OR "lay member*" OR stakeholder* OR consumer* OR representative* OR community) N2 (engag* OR involv* OR participa* OR consult* OR activat* OR partner* OR collab* OR contribut* OR "co-develop*" OR "co-research*" OR "co-lead*" OR "co-produc*" OR "decision-making" OR advisor*" OR "shared leadership") OR TI/AB (ppi* OR "patient involvement*" OR "public involvement*" OR "patient engagement*" OR "public engagement*" OR "patient particip*" OR "citizen science*")
Concept 2: Clinical trials in maternal and neonatal health	Index terms: EMBASE: ('clinical trial'/exp OR 'clinical trial (topic)'/exp) AND ('maternal care'/exp OR 'obstetrics'/exp OR 'pregnancy'/exp OR 'pregnant woman'/exp OR 'newborn'/exp OR 'childbirth'/exp OR 'perinatal care'/exp OR 'prenatal care'/exp OR 'puerperium'/exp OR 'infant'/exp) CINAHL: ((MH "Clinical Trials+") AND ((MH "Obstetrics") OR (MH "Pregnancy+") OR (MH "Expectant Mothers") OR (MH "Infant, Newborn+") OR (MH "Childbirth+") OR (MH "Perinatal Care") OR (MH "Prenatal Care") OR (MH "Puerperium") OR (MH "Infant+")) MEDLINE: (MH "Clinical Trials as Topic+") AND ((MH "Maternal Health") OR (MH "Obstetrics") OR (MH "Pregnancy+") OR (MH "Pregnant Women") OR (MH "Infant, Newborn+") OR (MH "Parturition+") OR (MH "Perinatal Care+") OR (MH "Prenatal Care") OR (MH "Postpartum Period+") OR (MH "Infant+")) PsycInfo: DE "Clinical Trials" AND (((((((DE "Obstetrics") OR (DE "Pregnancy")) OR (DE "Prenatal Care")) OR (DE "Intrapartum Period")) OR (DE "Antepartum Period")) OR (DE "Postnatal Period")) OR (DE "Birth")) OR (DE "Neonatal Period")) Keywords: ("clinical trial* OR trial* OR intervention*) AND (matern* OR obstetric* OR labour OR labor OR pregnant* OR "pregnan* wom*n" OR "expect* mother*" OR birth* OR childbirth* OR "child birth" OR antenatal* OR "ante-natal*" OR antepartum OR ante-partum OR intrapartum OR prenatal* OR "pre-natal*" OR perinatal* OR postpartum* OR puerperium OR parturition OR "post-partum*" OR neonatal* OR neonat* OR newborn* OR "new-born*" OR infant*)

Data extraction and analysis

One reviewer will extract data from all included studies, which a second reviewer will corroborate. A purposively designed data extraction form will be developed. Data extracted will include study design, aim of study, location and setting, year study was conducted, participant demographics, information on PPI and impact on trial design and outcomes (as described by stakeholders), and findings of experiences and views of PPI.

A thematic analysis will be undertaken, following the approach described by Thomas and Harden (2008) unless another method is more appropriate once the raw data have been assessed. This approach will involve generating codes from study findings (i.e., authors' interpretations and study participant verbatim quotes) extracted from the included studies. These codes will then be grouped into descriptive categories, followed by the researchers generating new analytical themes from these descriptive categories (Thomas & Harden, 2008). In order to enhance the trustworthiness of the findings (Barrett *et al.*, 2020), a reflexive approach will be undertaken throughout this QES. Research diaries will be used to document the process and formation of themes, as well as regular discussions between all three reviewers.

Methodological quality assessment

The twelve-item assessment criteria checklist by Thomas *et al.* (2003) will be used to assess the methodological

quality of the included studies. The tool is widely used for assessing the methodological quality of qualitative research. Two reviewers will independently assess the quality of each included study. Consensus will be reached through discussion or through assessment by a third reviewer, if required. Studies will not be excluded from the review based on judgements of low methodological quality, rather this information will be reported in the findings and highlighted in the discussion, as appropriate.

In addition to a quality assessment, the GRIPP2 short form checklist (GRIPP2-SF) will be utilised to evaluate the reporting of PPI in included studies (Staniszewska *et al.*, 2017). The GRIPP2 checklists, available in both long- and short-form versions, were developed to strengthen the quality of PPI reporting (Staniszewska *et al.*, 2017).

Assessing the findings

The GRADE-CERQual (Confidence in the Evidence from Review of Qualitative Research) assessment will be used to assess and summarise confidence in the review findings (Lewin *et al.*, 2018).

Study status

The protocol has been registered on PROSPERO, and the search strategy developed (Table 1). The search strategy will be implemented on publication and approval of this protocol.

Discussion

To our knowledge, this is the first QES on stakeholders' experiences and perceptions of PPI within clinical trials in neonatal and maternal healthcare.

Data availability

Underlying data

Open Science Framework: Stakeholder's perspectives and experiences of patient and public involvement (PPI) in clinical trials in maternal and neonatal healthcare: protocol for a qualitative evidence synthesis

https://doi.org/10.17605/OSF.IO/RD7XH (Hannon *et al.*, 2023)

This project contains the following underlying data:

Data Extraction Form.xlsx

Reporting Guidelines

Open Science Framework: PRISMA-P checklist for 'Stakeholder's perspectives and experiences of patient and public involvement (PPI) in clinical trials in maternal and neonatal healthcare: protocol for a qualitative evidence synthesis'. https://doi.org/10.17605/OSF.IO/RD7XH (Hannon *et al.*, 2023)

Software availability

Endnote and Covidence are proprietary software. Alternative software that can be used includes Zotero for reference management include and Rayyan for conducting literature and systematic reviews.

Acknowledgments

We would like to thank Dr Katie Gillies and Dr Linda Biesty for reviewing this protocol before submission for publication.

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Reference Source

Open Peer Review

Current Peer Review Status: ✔ ?

Version 1

Reviewer Report 18 August 2023

https://doi.org/10.21956/hrbopenres.15019.r35025

?

Sandy Oliver
Social Research Institute, University College London, London, England, UK

This planned qualitative evidence synthesis (QES) will address the important topic of stakeholders' perspectives and experiences of PPI. The key concepts (stakeholders and PPI) are clearly defined. The methods proposed are well established. I have a couple of suggestions to strengthen the work.

First, I suggest the authors consider the variety of people who might be part of PPI: individuals who can reflect on their own experience or expectations; and advocates who are well informed by networks of patients and/ or the wider public about a variety of experiences and expectations. I would anticipate the involvement of these different groups suiting different specific purposes and methods for involvement.

Second, I suggest the authors revisit the procedures for screening search outputs. Their current plans for one reviewer to screen titles and abstracts, followed by a second reviewer screening 20% of titles and abstracts suits the purpose of checking accuracy – the usual purpose for this exercise in effectiveness reviews. However, much of this work involves interpretation. I recommend both reviewers screening independently a small set of titles and abstracts then comparing and discussing any discrepancies so they can understand how they have interpreted the text differently. Taking those different interpretations into account, the definitions and screening guidelines can then be refined to more accurate reflect the concepts of interest in the eligibility criteria. This can be repeated with another small set of titles and abstracts until the reviewers are satisfied that they have clearly expressed and consistently applied their eligibility criteria.

At the data extraction stage, I recommend extracting data about the types of individuals involved in the role of patients or the public.

The protocol states that 'The GRADE-CERQual (Confidence in the Evidence from Review of Qualitative Research) assessment will be used to assess and summarise confidence in the review findings'. This statement would be clearer if 'findings' were replaced by 'analytical themes' or 'synthesis statements'.

Generally the authors make good use of standardised tools for research synthesis.

I look forward to reading the resulting synthesis.

Is the rationale for, and objectives of, the study clearly described?
Yes

Is the study design appropriate for the research question?
Yes

Are sufficient details of the methods provided to allow replication by others?
Yes

Are the datasets clearly presented in a useable and accessible format?
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: PPI and research synthesis methodology

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Author Response 11 Sep 2023
Kathleen Hannon

Reviewer comment:
This planned qualitative evidence synthesis (QES) will address the important topic of stakeholders' perspectives and experiences of PPI. The key concepts (stakeholders and PPI) are clearly defined. The methods proposed are well established. I have a couple of suggestions to strengthen the work.
Author response:
Thank you for reviewing this protocol and for your feedback. We hope that the amendments we have made and our responses will address your suggestions to help improve the protocol.

Reviewer comment:
First, I suggest the authors consider the variety of people who might be part of PPI: individuals who can reflect on their own experience or expectations; and advocates who are well informed by networks of patients and/ or the wider public about a variety of experiences and expectations. I would anticipate the involvement of these different groups suiting different specific purposes and methods for involvement.
Author response:
Thank you for highlighting this. We also agree that there is a wide variety of people who are

involved in PPI. For our inclusion criteria, we have updated our definition of 'PPI contributor' to reflect this, using the HSE Executive Research and Development's definition for involvement in research. Our definition now states:

"'PPI contributor' refers to 'patients, service users, carers, families using health and social care services, people with lived experience of health conditions (who may or may not be current patients), patient advocacy organisations, and members of the public' (Health Service Executive Research and Development, 2021, p. 3), who is involved at any stage of the trial process, from trial conception to trial dissemination.

Reference: https://hseresearch.ie/wp-content/uploads/2021/12/Guide-no-8-Patient-and-Public-Involvement-in-HSE-Research.pdf

Reviewer comment:
Second, I suggest the authors revisit the procedures for screening search outputs. Their current plans for one reviewer to screen titles and abstracts, followed by a second reviewer screening 20% of titles and abstracts suits the purpose of checking accuracy – the usual purpose for this exercise in effectiveness reviews. However, much of this work involves interpretation. I recommend both reviewers screening independently a small set of titles and abstracts then comparing and discussing any discrepancies so they can understand how they have interpreted the text differently. Taking those different interpretations into account, the definitions and screening guidelines can then be refined to more accurate reflect the concepts of interest in the eligibility criteria. This can be repeated with another small set of titles and abstracts until the reviewers are satisfied that they have clearly expressed and consistently applied their eligibility criteria.
Author response:
Thank you for this suggestion. We agree that this method would improve the conduct of this review. We have updated the data screening section of the protocol to reflect this:

'Two reviewers will conduct a pilot of the screening process. A sample of titles and abstracts will be reviewed by both reviewers, who will then discuss with each other their interpretation of the inclusion criteria. This process will be conducted in order to ensure a shared understanding of the inclusion criteria when screening. Once the reviewers have reached agreement on interpretation of the inclusion criteria, screening of retrieved citations will commence.'

As stated above, based on your feedback about the variety of people involved in PPI, we have also updated our definition of 'PPI contributor' for the review, which has refined the eligibility criteria.

Reviewer comment:
At the data extraction stage, I recommend extracting data about the types of individuals involved in the role of patients or the public.
Author response:
Thank you. For data extraction, we stated we wished to extract 'participant demographics, [and] information on PPI.' As we anticipate that the information provided will vary in detail across included studies, our intention is to extract any and all information provided about

the individual or individuals involved in PPI within the included studies. All information on PPI and PPI contributors provided will be considered, particularly as we intend to assess the quality of reporting of PPI using the GRIPP2-SF checklist. We have updated the protocol to state:
'The information on PPI that will be extracted will include information such as who was involved as a PPI contributor, the number of PPI contributors involved, stage and types of involvement, and any other information provided.'

Reviewer comment:
The protocol states that 'The GRADE-CERQual (Confidence in the Evidence from Review of Qualitative Research) assessment will be used to assess and summarise confidence in the review findings'. This statement would be clearer if 'findings' were replaced by 'analytical themes' or 'synthesis statements'.
Author response:
We believe that the statement is appropriate. In the Lewin et al.'s 2018 article cited in the protocol, GRADE-CERQual is described as an approach that 'provides guidance for assessing how much confidence to place in findings from systematic reviews of qualitative research (or qualitative evidence syntheses)' (https://doi.org/10.1186/s13012-017-0688-3). On the GRADE-CERQual website, GRADE-CERQual is described as 'an approach for assessing how much confidence to place in the findings of a qualitative evidence synthesis' (https://www.cerqual.org/what-is-the-grade-cerqual-approach2/)

Competing Interests: No competing interests were disclosed.

Reviewer Report 21 July 2023

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julie abayomi
Edge Hill University, Ormskirk, England, UK

Thank you for asking me to review this comprehensive and well-written protocol.

The paper defines and explains PPI clearly, and then goes on to explain the benefits of using this approach in the introduction. It also identifies that a QES regarding stakeholders' perspectives of PPI in a maternal or neonatal setting appears not to exist, and so identifies a gap in knowledge.

For the QES, the SPIDER tool will be employed to define inclusion criteria, ensuring rigour within the process. Furthermore, the search will be supported by a subject librarian, with relevant expertise. The search terms and databases suggested are all highly relevant.

The authors explain how screening will be conducted and intend to use a PRISMA flow diagram in their report to show this. Extracted data will be analysed thematically, using an agreed approach (Thomas and Harden 2008).

The quality of the research will be ensured by using an assessment checklist (Thomas et al 2003) and by referring to GRIPP2 when evaluating the reporting of PPI included in studies - this also ensures rigour and structure to the process. Another tool, GRADE-CERQual will be used to assess and summarise confidence, so rigorous methods are employed throughout the whole study.

Using Endnote, Covidence and Rayyan will all help to streamline the process.

My only suggestion is that the team may find it helpful to consider/refer to UK Health Research Authority (HRA) Guidelines regarding PPI (https://www.hra.nhs.uk/planning-and-improving-research/best-practice/public-involvement/). This covers the 4 principles for meaningful PPI in health research.

Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Yes

Are sufficient details of the methods provided to allow replication by others?

Yes

Are the datasets clearly presented in a useable and accessible format?

Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Maternal nutrition and health, including PPI with service users and midwives.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response 11 Sep 2023

Kathleen Hannon

Thank you very much for reviewing the protocol and for your feedback.
Thank you for drawing attention to the HRA's guidelines, which highlights important considerations for health researchers planning or engaged in PPI. There are several other PPI frameworks and guidelines aimed at researchers, PPI contributors, organizations, and funders that have been made available in recent years. In 2019, Greenhalgh et al. identified a total of 65 frameworks for conducting PPI that were published between 2003 and 2018. These frameworks provided guidance on various aspects of PPI, including advice on involving PPI in specific stages of research or addressing power imbalances between researchers and PPI contributors (Greenhalgh et al. 2019). More guidelines have been

published since, including the PPI Ignite Network Values and Principles Framework in 2022 (https://ppinetwork.ie/resource/ppi-ignite-network-values-and-principles-framework/).

Reference: Greenhalgh T, Hinton L, Finlay T, Macfarlane A, Fahy N, Clyde B, Chant A. Frameworks for supporting patient and public involvement in research: Systematic review and co-design pilot. Health Expect. 2019 Aug;22(4):785-801. doi: 10.1111/hex.12888. Epub 2019 Apr 22. PMID: 31012259; PMCID: PMC6737756.

Competing Interests: No competing interests were disclosed.