Patient and service user engagement in research: a systematic review and synthesized framework

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Abstract

Background There is growing attention towards increasing patient and service user engagement (PSUE) in biomedical and health services research. Existing variations in language and design inhibit reporting and indexing, which are crucial to comparative effectiveness in determining best practices.

Objective This paper utilizes a systematic review and environmental scan to derive an evidence-based framework for PSUE.

Design A metanarrative systematic review and environmental scan/manual search using scientific databases and other search engines, along with feedback from a patient advisory group (PAG).

Eligible sources English-language studies, commentaries, grey literature and other sources (including systematic and non-systematic reviews) pertaining to patient and public involvement in biomedical and health services research.

Data extracted Study description (e.g. participant demographics, research setting) and design, if applicable; frameworks, conceptualizations or planning schemes for PSUE-related endeavours; and methods for PSUE initiation and gathering patients'/service users' input or contributions.

Results Overall, 202 sources were included and met eligibility criteria; 41 of these presented some framework or conceptualization of PSUE. Sources were synthesized into a two-part framework for PSUE: (i) integral PSUE components include patient and service user initiation, reciprocal relationships, colearning and re-assess-

ment and feedback, (ii) sources describe PSUE at several research stages, within three larger phases: preparatory, execution and translational.

Discussion and Conclusions Efforts at developing a solid evidence base on PSUE are limited by the non-standard and non-empirical nature of much of the literature. Our proposed two-part framework provides a standard structure and language for reporting and indexing to support comparative effectiveness and optimize PSUE.

There is a large and growing impetus among patients, politicians, clinicians and researchers towards increasing the engagement of patients and other layperson/non-professional service users in health- and health-care-related research.[1–6] Attempts to expand this engagement of lay (i.e. non-clinician/non-professional researcher) users of health services have emphasized patients and caregivers,[5,7–11] but also broader categorizations and terms such as consumers/users of health-care services, community members or public/citizens.[1,10,12–15] Given the range of these terms, we will use patient and service user engagement (PSUE) in research to emphasize patients while referring to these endeavours broadly.

Increasing PSUE generally follows two aims: a moral/ethical drive to empower lay participants in an otherwise expert-dominated endeavour and ensure civically responsible research[5,16,17]; and 'consequentialist' reasoning for optimizing the validity, design, applicability or dissemination of the research itself and the effectiveness of resulting interventions.[18–21]

However, the promise of PSUE to ensure equitability and robust findings remains unrealized. Despite policy and funding directives (e.g. INVOLVE in the UK[22,23]; PCORI in the US), and some evidence that PSUE can improve research,[12,19,24–29] challenges persist. In particular, there remains some doubt as to whether current approaches represent merely 'tokenistic' efforts[30] or truly incorporate patient and service user contributions. Moreover, some groups are less likely than others to have their voices included.[31] In addition, there is limited evidence on PSUE's tangible benefits.[26,29,31] This is exacerbated by an absence of standard approaches

to conducting and reporting PSUE[32,33] – when it is reported at all[34] – which limits the potential for indexing, knowledge synthesis or comparative effectiveness for determining best practices. In sum, the reasoning for PSUE is compelling and multifaceted, but the extent to which existing approaches actually ensure inclusion, and whether there are consistent benefits in doing so, remains unclear because of a lack of standard framework or language.

Utilizing a systematic review, environmental scan and manual search of peer-reviewed literature and other sources regarding PSUE in biomedical and health services research, this paper synthesizes a standardized, evidence-based framework for understanding, reporting and assessing PSUE to jump-start a reliable and comparative evidence base. An important percentage of previous work on this topic has consisted of individual studies (usually at single sites and with limited resources) or commentaries. Few studies (incorporated and reviewed here as well) have been performed with necessary rigour or an eye towards systematically assessing and organizing existing evidence into a broadly applicable framework. Doing so is vital to assessing PSUE in existing research and building towards an evidence-based understanding of best practices.

Methods

Methods consisted of a systematic review, environmental scan and manual search of peer-reviewed literature and other sources, supplemented with input from a patient advisory group (PAG). The study received Institutional Review Board clearance.

Electronic database search

Eligibility criteria

We included studies of any design, size and patient age or morbidity status, published in English, in which patients, surrogates, caregivers or other service user stakeholders participated in planning or conducting biomedical and health services research (including input on research/funding agendas, outcomes or design, and active engagement in research activities). We also included non-original or summarized literature (systematic or non-systematic literature reviews, commentaries, etc.).

Search strategy

An expert reference librarian (PJE) and systematic review methodologist (MHM) collaborated to develop the search strategy. Medical subject heading (MESH) terms and text words were selected based on common indexing practices. Search terms were compiled and tested repeatedly to produce sensitive searches and capture potentially relevant publications. Our search covered biomedical databases and other sources: PubMed/Ovid MEDLINE, Ovid EMBASE; Ovid PsycInfo, Ovid Cochrane (especially Sys Rev, Methods, HTA), EBSCO CINAHL; SCOPUS (to capture potentially relevant sources in the social sciences); Web of Science (multidisciplinary scientific content); and Business Search Premier, Academic Search Premier and Google Scholar. We also reviewed reference lists from eligible studies, conducted additional MEDLINE searches using the PubMed-'related articles' feature for eligible studies and employed SciSearch for publications that cited eligible studies.

Environmental scan and manual search

The goal of the *environmental scan* was to identify relevant information in sources not published in the biomedical bibliographic databases. We were interested in the actors and stakeholders involved, key events, documentation (white papers, position papers, proceedings) and trends (descriptions of upcoming and on-going activities). We used search engines Scirus and Sciverse, which contain scientific journal content, scientists' homepages, courseware, pre-print server material, patents and institutional repository and Website information, along with Google and Bing. The *manual search* covered Websites recommended by topic experts including the project team, external advisors and the PCORI methodology working group. We extracted data from relevant links including title, source, author, URL, content description and main conclusions.

Study selection

We collated initial references in citation files using Endnote software, removed duplicates and screened titles and abstracts against eligibility criteria using DistillerSR software. Team members reviewed studies in duplicate until adequate agreement (Kappa > 0.80) was achieved. Disagreements during initial screening were automatically included. Potentially eligible studies then received full text review following a similar procedure. Disagreements in full text screening were reconciled by discussion, consensus or arbitration by third reviewer. We exclusively used electronic file formats (Portable Document Format/PDF) to reduce costs and paper use.

Data extraction

Data extraction from included studies followed a standardized form developed from the protocol, created in DistillerSR and tested on a small sample ($n = 10$) of included studies to ensure sufficient quality and performance. Data extracted included study description (e.g. participant demographics, research setting), methods for patient/service user selection or initiation into research and for obtaining their input or contributions, and frameworks or conceptualizations used for the approach.

Patient advisory group

Finally, we presented our project aims, initial findings and recommendations to a PAG[35] and

asked for feedback on terminology, usefulness and applicability (located at the end of the Results). Notably, among the terms for PSUE participants (consumers, representatives, etc.), they felt that 'patient' or 'informant' was most useful, but found most terms generally confusing. For clarity and inclusivity – and to avoid confusion with 'participants' in more conventional researcher-driven models (e.g. participants as data points in a study) while also avoiding non-relevant connotations of 'informant' – we will simply use *patient and service user representatives* (PSURs) to refer to layperson patients, service users or their surrogates who are engaged in the research process itself.

Analysis

Due to our study objectives and heterogeneity of sources, we did not conduct a quantitative meta-analysis. Rather, we followed a metanarrative approach.[11] The present analysis focused on assessing sources, which included frameworks or conceptualizations of PSUE processes, or which described specific stages of PSUE, and then synthesizing a systematically inclusive framework.

Results

Search and selection results: framework for PSUE

Our search identified 5560 possibly relevant citations, of which 202 met eligibility criteria (study selection is described in Fig. 1); see full bibliography in Data S1. Of these 202 sources, 41 described some framework, conceptualization or planning scheme for the parts of the PSUE process. Of these 41 sources, most were
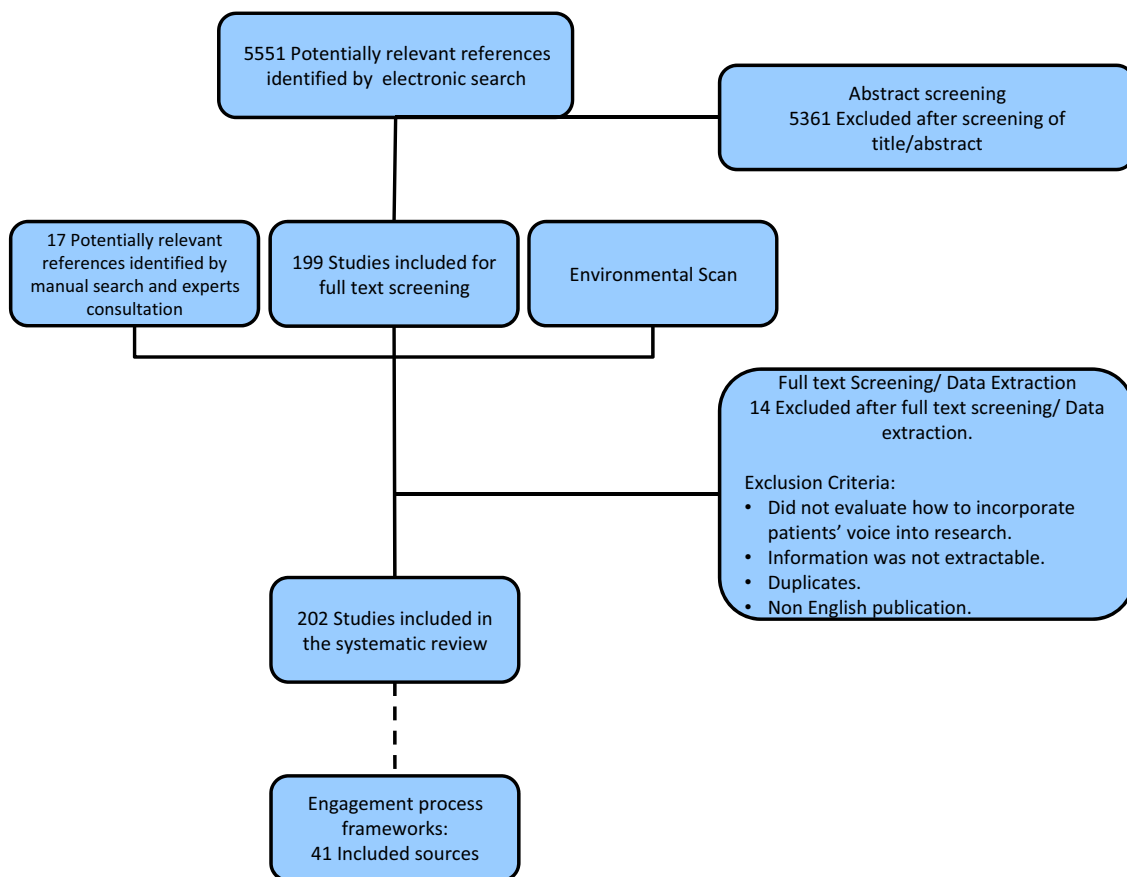

Figure 1 Study selection process.

not accompanied by original evidence, but rather largely consisted of non-systematic literature reviews, commentaries, etc. Instead, the 161 other eligible sources provided a great deal of detail (including research examples). We synthesized the frameworks and supporting sources into a two-part framework, outlined here as an organizing structure, comprised of (i) the integral *components* of PSUE, and (ii) the *phases and stages* of PSUE in research.
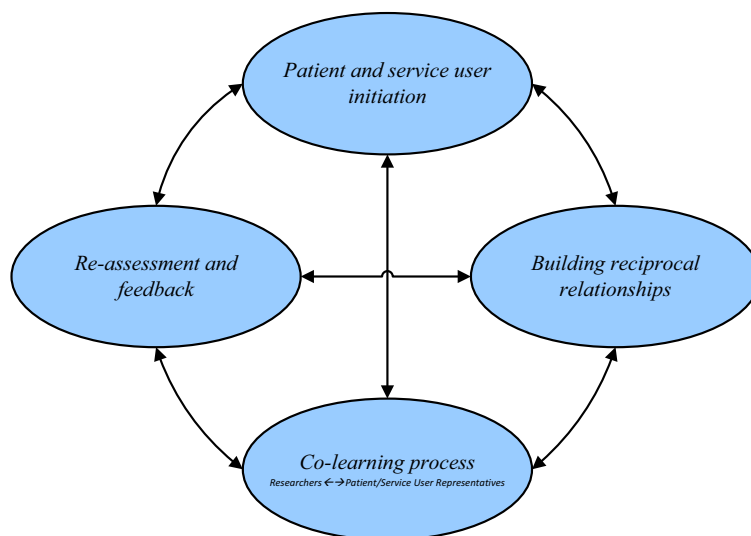
Components of PSUE

Two reviewers coded the components of PSUE described in studies into discrete categories and recorded their inter-relations. We found four essential components: patient and service user initiation, building reciprocal relationships, co-learning and re-assessment and feedback (Fig. 2). Functionally, these components were described as having a circular, bidirectional relationship, comprising consecutive feedback loops in researcher–PSUR collaboration. We were unable to identify one particular component as being the most important, but this proposed framework describes a process in which changes or decisions related to each component may inform and influence the others. Thus, ideally, work on these components might be performed continually throughout projects until saturation – that is, when no additional information is being shared and when both parties agree to move forward – although finite time and resources will likely place limits on groups' abilities to do this. Some studies employed only described one component; others incorporated several. We describe the components below.

Component 1. Patient and service user initiation
Ten sources included information on patient and service user initiation, meaning the entry of lay patients and service users into the research process – whether through researchers' engagement efforts or PSURs' own interests and actions. Three overarching factors are keys to initiation of PSURs' efforts in PSUE processes. First, studies demonstrated the importance of engaging PSURs as early as possible in the process so that they can steer agendas and outcomes and provide a values context, which will improve study design and applicability, ensure a relevant perspective and prevent patients and service users from being relegated to disempowered 'subjects' with no impact.[36–45] Second, PSURs should consist of individuals or communities for whom the outcomes are of interest,[46,47] but must also display at least notable characteristics, particularly participatory behaviour.[45]

Figure 2 Components of patient and public involvement in research.

Third, there must be potential for PSURs to have an active role – including their ability to engage, agreement on expectations and the possibility for a sense of equality between parties.[48] Presumably, these three factors would be best represented by the many instances in which PSURs initiate and lead research themselves.[17,49] However, a review on PSUR-controlled research did note several concerns, including inequality in the interactions between PSURs themselves, added stress for PSURs, and the perception that such research will be less valued than conventional researcher-controlled research.[49]

Component 2. Building reciprocal relationships
Thirty studies highlighted this component. Many stated that, from the very beginning, researchers should see PSURs as equal partners and consider them as a reliable component of the team, rather than simply an additional variable or complication.[36–41,43,44] Both parties have to clearly know their roles and the importance of those roles independent from each other. Thus, reciprocal cultural competence is a vital feature of PSUE,[50] and partnerships should include a mutual understanding of partners' needs, capacities and goals,[51] with conflicts solved promptly and explicitly. After all, PSURs' suboptimal experiences (e.g. abandonment after data collection) could affect future relationships with researchers.[52]

Component 3. Colearning process
Colearning received attention in nine studies. PSURs may need some research expertise to prevent researchers from dominating agendas and opinions.[53] Thus, PSURs may require education/training about content or methodology to carry out a productive dialogue or conduct research themselves.[16] Colearning also may increase PSURs' confidence, promoting more active engagement and reducing the risk of tokenistic PSUE. More broadly, PSUE must provide opportunities to all team members to acquire new knowledge and skills; researchers could begin by training *themselves* so that projects better adhere to participatory principles.[54]

Additionally, researcher education could improve awareness regarding the realities that PSURs face and about relevant social dynamics.[16] This may also improve protocols, phrasing/language in measurement and applicability of results.[45]

Component 4. Re-assessment and feedback
The final component, supported by two studies, entails evaluating the PSUE process, further clarifying PSURs' and researchers' roles and expectations and potentially modifying the other as needed. Doing so continuously not only helps ensure PSUR empowerment, but also can reveal potential facilitators and barriers.[55,56] Thus, the execution of this component will improve the robustness of the research project in question and future attempts at PSUE.

Phases of PSUE in research

Available literature describes PSUE across the research process. Broadly, at any stage of research, PSUE represents a potential spectrum of engagement for PSURs, as organized by existing sources outlining this spectrum,[57–59] ranging from the most passive role of being a study participant (a data point) to more engaged roles including tighter collaboration with researchers, to ultimately initiating, conducting, 'owning' or 'leading' research themselves (Table 1).

Thirty-seven sources describing frameworks/conceptualizations of PSUE converged into a synthesized framework (Fig. 3), comprised of three broad *phases* of research (*preparatory*, *execution* and *translational* phases), which are, in turn, comprised of specific *stages*. Five sources were most complete and useful in providing terminology and structure,[53,60–63] and so we drew from these most heavily in the terms and examples provided for Fig. 3.

Phase I: Preparatory phase
This phase consists of setting research and funding/resource allocation agendas (including identifying and prioritizing key topics and questions), in the service of answering the

Table 1 Levels of patient and service user engagement in research

Level of engagement	Oliver et al.[57]	Hall et al.[58]	Happel et al.[59]
Passive	Minimal	Consultation	Consumer advisory
	Consultation		Consumer consultation
		Collaboration	
	Collaboration		Consumer collaboration
		User control	Consumer led
Engaged	Lay control		
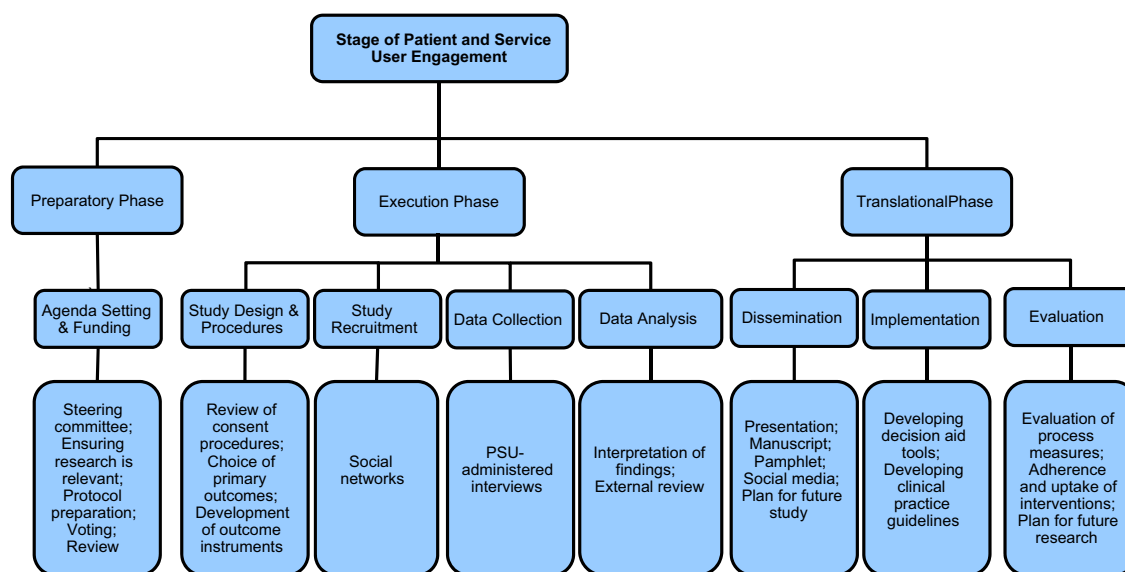
Figure 3 Phases and stages of patient and service user engagement in research.

question: What to research?[63] Thirty-three of 202 studies described PSUE in this phase. Notably, some studies or PSUE efforts have been initiated or primarily conducted by PSURs themselves,[64–66] sometimes due to their dissatisfaction with existing care or weariness of being disempowered research 'subjects'.[67,68] The preparatory phase includes two stages:

1. *Stage 1: Agenda Setting.* Broadly, as revealed in a prior systematic review that summarized studies published through 2008,[69] a sizable literature (156 studies) exists in which PSURs have actively taken part in shaping research priorities; *PSURs specifically identified important research topics or questions in 148 of those studies.*
2. *Stage 2: Funding.* PSUE in funding aspects was much more limited than agenda setting.

One study reported some involvement in developing a bid or a proposal, and another reported PSURs' input leading to improved coverage of all trial expenses.[70,71] We also found a survey by INVOLVE that evaluated PSUE in commissioning and funding processes.[23] The 2007 survey of 32 statutory and voluntary organizations showed that around 80% of surveyed organizations never or hardly ever considered PSUE as a criterion of funding. Also, 50% of organizations never involved PSURs in funding decisions. It is worth noting research conducted by The James Lind Alliance on 'patient and public involvement' among 55 clinical research organizations and funders in the UK along with a review of the literature.[72] This assessment found that current funding decisions are largely based on judgment

about scientific merit, rather relevance and importance of outcomes to PSURs.

Phase II: Execution phase

Study execution includes PSUR feedback or participation in the development of study design and procedures and execution of the protocol (including subject enrolment, delivering the intervention and data collection and analysis).

1. *Stage 1: Study Design and Procedures.* Study design includes the selection of primary outcomes and methods, to answer the question: How to do the research project?[63] Thirty studies described PSUE in study design and procedures. One notable aspect of PSUE here is the issue of ethical considerations raised by PSURs.[37]

2. *Stage 2: Recruitment and participation.* Forty-three sources described PSUE pertaining to subjects/participant recruitment. In particular, numerous systematic reviews and studies examined individuals' perceptions of, or experiences in, trials to address low trial enrolment and participation among different populations.[73–76] One systematic review[15] found evidence that a critical reason for low trial enrolment could be confusion and difficulties with the concept of randomization: without an understanding of the structure and aim of a trial, the decision to participate will be extremely difficult to make.[77] Indeed, there is often a lack of distinction between the goals of research and medical care, and so all parties need assistance to ensure that decisions are consistent with participants' values.[78] Thus, PSUE here can ensure clearer communication and better understanding for prospective subjects/participants.

3. *Stage 3: Data Collection.* Seven studies addressed PSUE in data collection. A clear need for more attention is needed: one survey taken by Cochrane research groups[79] showed that one in three respondents did not include consumers' perspectives in determining the data to be collected in systematic reviews.

There was also no apparent consensus regarding the importance of identifying and collecting information on PSUR-defined outcomes or on integrating such information into their activities. Yet, PSUE in developing self-report questionnaires/indices can be helpful[80–82] and may produce evidence more consistent with PSURs' concerns and minimize bias towards providers' perspectives. In addition, PSUE allowed PSURs who were uncomfortable with questionnaires to participate – for example, engaging key PSURs as trained research assistants in collecting and analysing data.[80]

4. *Stage 4: Data Analysis.* Twelve included studies, with various research designs, presented relevant data regarding PSUE at this stage. Largely, PSUE in data analysis – including presenting findings and conclusions to participants prior to publication – resulted in an improved ability to contextualize conclusions to PSURs' environments and beliefs, added language and cultural insight and highlighted PSURs' priorities for a more focused analysis.[37,44,83–86] Additionally, PSUE at this stage may increase the validity of findings, as the different parties must reach consensus on emerging conclusions, acting as checks and balances on one another's biases.[80] In analysis perhaps more than any other stage, PSUE requires sufficient education and training.[37,44,80] However, PSURs' abilities should not be undervalued: challenges such as serious mental illness[80] or lack of resources in developing countries[37] have not prevented PSURs from understanding research foundations, giving feedback and strengthening research. Thus, information should be understandable to PSURs while avoiding oversimplification.[83]

Phase III: Translational phase

This phase consists of post-analysis activities.

1. *Stage 1: Dissemination.* Twelve studies addressed dissemination. Although peer-reviewed publication is the common dissemination target for academics, it is not the most direct way to

disseminate findings to laypersons.[53,87] A dissemination approach decided jointly by researchers and PSURs is critical to the success of research partnerships, and this process should be personalized and accessible for individuals' different abilities and preferences even within the same population.[40,53,60,88] Such accessibility includes language and terminology according to the target population and purpose of the publication – in most scenarios, jargon and non-applied information should be avoided, which can be facilitated by PSUE.[37,40,53,60,89–91] Regular updates, through newsletters, mailings or other modes can improve confidence and buy-in for research projects among target populations.[37,44] In addition, PSUE may help develop creative dissemination methods, which are more efficient and which may not have been considered by researchers.[16,60,92] This is important, because adequate dissemination has proven to be helpful for future implementation, and making results known within several strata may facilitate obtaining resources and funding for implementation.[53]

2. *Stage 2: Implementation*. Twenty-eight sources described PSUE in implementation of findings. Several studies advocated involving PSURs throughout implementation[44,53,60,89,93,94] and called for adequate PSUR education and support[37,60,89] to maintain their interest and enthusiasm.[95,96] Involvement of authority figures in the community was also helpful for buy-in.[37,97] However, PSURs may create a force for implementation by themselves, without researcher involvement.[86] PSUE was most helpful in prioritizing the sequence of steps and targeting the methodology of implementation.[40,88,98] Flexible and creative plans are needed; problems may arise unexpectedly during implementation and should serve as learning opportunities.[37,94,95]

3. Clinical practice guidelines (CPGs) are a form of PSUE in implementation, which may have the greatest impact on health care and services. One systematic review[99] found that the most frequently cited objective for PSUE in developing guidelines was to incorporate PSURs' values or perspectives in CPG recommendations, an aspect emphasized by most modern guideline development schemes (e.g. the Grading Recommendations, Assessment, Development and Evaluation/GRADE[100]), guideline rigour evaluation tools (e.g. Appraisal of Guidelines for REsearch & Evaluation instrument/ AGREE II[101]) or guideline implementability evaluation tools (GuideLine Implementability Appraisal/GLIA[102]). Boivin *et al.*[6] surveyed 56 guideline developers and highlighted the role of PSUE and the need for training and education. And, of course, PSUE is currently in progress in systematic reviews,[103] which will provide evidence for future CPGs. However, there remains no clear guidance or evidence on processes or outcomes to inform the design of PSUE in CPG development, and one systematic review found that PSUR contributions are frequently not acted upon, raising the spectre of tokenistic engagement.[104]

4. *Stage 3: Evaluation*. Very few studies (only 5) addressed this step and mostly did not provide sufficient details. Authors of these studies advised that the evaluation process should be constant; waiting until the end of the process will make problem-solving more difficult and resource-intensive.[37,94] A continuous flow of information from PSURs also demonstrated great value,[91,92] as did having clear, pre-defined assessment tools.[97] Despite the benefits of constantly evaluating relationships between PSURs and research teams, the extent of PSURs' participation needs to be clarified to avoid conflicts and favour the development of future projects.[53]

Environmental scan

The environmental scan revealed numerous links to relevant Websites, organizations, forums, blogs, videos, associations, workshops, presentations, governmental agencies, abstracts and other unpublished resources spanning both

health-care and non-health-care PSUE. Some focused on shared decision making in the clinical context and were less relevant to this review, whereas others discussed participatory action research or community-based participatory research. The most relevant resources can be roughly categorized into three types:

1. Disease-specific social networks (mostly not-for-profit, PSUR-established). Many offered insight on participation in research and guided patients and service users to on-going trials and investigational treatment. Few provided education; one notable example was The Association of Cancer Online Resources,[105] which provided clinical trial FAQ; access to investigational drugs; and guides to trial terminology and finding clinical trials.
2. Non-disease-specific Websites focused on PSUE (most commonly in Europe, specifically the UK, and in Canada). Examples include the National Institutes for Health Research,[106] the James Lind Alliance,[107] and the PatientPartner project,[108] which aim to help PSURs set priorities, have input on proposals and funding, actively participate in studies and collaborate with clinicians, researchers and other stakeholders to strengthen research, create new partnerships and address challenges.
3. Models for PSUE, which were fairly uncommon. Two examples are 'A Model Framework for Consumer and Community Participation in Health and Medical Research'[63], created by Australia's National Health and Medical Research Council; and the 'National Health Service Patient Involvement Toolkit.'[93] These provided a rationale for PSUE at every step of research, and their findings are incorporated in the frameworks presented in this report.

Patient advisory group feedback

We presented our findings and recommendations to a PAG,[35] a committee of community member PSURs with a long history of research

engagement. Members provided the following feedback:

1. The terms informant/patient/surrogate/consumer/customer/representative were fairly confusing and none seemed satisfactory, although patient or informant seemed most intuitive. Associated explanatory text attached to any terms used was deemed necessary.
2. PAG members understood the purpose of the frameworks presented and valued the need for such frameworks.
3. PAG members equally rated the importance of the four components of the framework (participant selection, building reciprocal provided, colearning and re-assessment and feedback).
4. PAG members rated as most important the overarching recommendation to engage PSURs in all three phases of research, as benefits that would likely outweigh difficulties.
5. In general, PAG members found the extent of possible engagement in research to be surprising.
6. PAG members provided some suggestions for wording, graphics and presentation.

Comparison with other systematic reviews

A systematic review by Oliver *et al.* regarding PSUR ('consumer') involvement in research and development agenda setting for the UK's National Health Service[109] emphasized processes, outcomes and relevant factors of PSUR participation in identifying and prioritizing research topics (similar to part of the 'preparatory phase' in the present framework). Within this area, they presented a multipart framework, focused on (i) the different consumers involved, (ii) who initiated the research, (iii) the degree of PSUR involvement, (iv) forums for communication, (v) decision-making methods, and (vi) practical issues in implementing PSUE in agenda setting. A second review, from a cancer organization in Australia, focused on 'consumer involvement' specifically in 'cancer control' (meaning research, but also prevention, early detection, treatment and other components).[110]

Rather than stages of research, this review emphasized key elements of PSUE, including committed organizations, shared focus and other factors, similar to the first part of our two-part framework. Another review, by Staley for INVOLVE,[111] emphasized 'public' involvement (including patients, service users, organizations, community members and others) and focused on the impact of public involvement by different stages of research, similar to the second part of our current framework. This review called for a greater evidence base, noting the largest gaps in research on PSUE impact on funding and impact on certain kinds of analyses. Finally, the PIRICOM study[17] examined conceptual/theoretical trends, measurement and impact of PSUE on health-care and social-care research. Among other findings, the PIRICOM study concluded that relatively little conceptual or theoretical development exists regarding PSUE, noting that papers that have focused on conceptualization in the past have been based upon reflection or opinion.

These previous reviews have varyingly examined the levels of PSUR involvement, key components necessary for PSUE and/or one or more of the stages at which it occurs. They have also, varyingly, emphasized mainly one phase (e.g. agenda setting) but not others,[109] emphasized research stages over key components[111] or vice versa[110]; described conceptions in other papers without synthesizing a broadly applicable framework of their own[17]; or examined approaches to PSUE alongside its impacts, outcomes or other activities as part of a broader view.[17,111] Our review attempts to capture and synthesize the components and stages presented across previous reviews (along with other studies) with the goal of presenting a comprehensive framework and language that can be directly applied to future studies in reporting PSUE activities.

Limitations and strengths

The main limitation to this systematic review is the non-comparative, observational and/or non-empirical nature of available literature.

Therefore, our two-part framework of the components and stages of PSUE engagement is built from sometimes disconnected, and insufficiently tested or reported, literatures. Additionally, the lack of specific indexing terms in bibliographic databases means that some studies using PSUE may have been missed, and there is a lack of standardized, explicit reporting for PSUE processes. Therefore, standard reporting guidelines for study designs (e.g. the CONSORT statement for randomized trials) can be enhanced by including a template for reporting use of PSUE and at what phases and stages. To overcome these challenges in indexing and reporting, we attempted an environmental scan to supplement the literature search. Heterogeneity of populations, methods and outcomes constitute further limitations to extrapolation of evidence. Publication and reporting biases have also likely affected the conclusions of this report, and their impact could not be estimated.

The strengths of this report include a comprehensive and sensitive search strategy spanning multiple databases and augmented by an environmental scan of unpublished relevant sources and contact with content experts to further capture related studies, Websites and viewpoints. *A priori* protocols for selecting and appraising evidence were implemented to reduce biased selection of studies. Our review and synthesized framework lay the groundwork for a standardized evidence base using guided indexing procedures and following a synthesized model.

Knowledge gaps and recommendations for research

Based on the proposed model, the following areas of research are needed to develop a complete body of evidence:

1. PSUE in the phase of study execution and specifically in the areas of data collection and analysis is needed. Available literature focuses on earlier (agenda setting and participant enrolment) and some later stages (translation) of research.

2. Research comparing the different methods of engagement and obtaining PSUR voice is needed, but this will require standard reporting and measurement.
3. Peer-reviewed studies are needed that incorporate and describe PSUE as outlined in our synthesized framework to assess feasibility; it is notable that of the 41 sources which most informed the structure of the synthesized framework, most provided no original evidence to support their framework or conceptualization.
4. Proper indexing of studies on PSUE will facilitate future synthesis of evidence and advancement of methods and outcomes.

Our review reveals a disconnect in the approaches used by existing studies; the need for a standard framework and language is clear. In synthesizing existing work into such a framework, the present paper also provides such a framework with the broad applicability and cohesive underpinnings necessary to integrate existing knowledge and guide future endeavours.

Funding source

Supporting Information

Additional Supporting Information may be found in the online version of this article:
   Data S1. Included sources.

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