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For peer review only

A scoping review of models and frameworks of patient engagement in health services research

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Abstract

Objective: To count and describe the elements that overlap (i.e., present in 2 or more) and diverge between models and frameworks of patient engagement in health services research. Our specific research question was, "What are the elements that underlie models and frameworks of patient engagement in health services research?"

Design: Scoping review.

Eligibility criteria: We included published and unpublished (i.e., grey) literature that presented (a) models or frameworks (b) of patient engagement (c) in health services research. We excluded articles unavailable as full-text or not written in English.

Data sources: Using a search strategy co-developed with an academic librarian, published literature was identified by searching six electronic databases. Searches for unpublished literature included electronic databases and websites. These searches were supplemented by snowball sampling.

Data extraction and synthesis: Two independent reviewers extracted data from included articles using an a-priori developed standardized form. Data were synthesized using both quantitative (i.e., counts) and qualitative (i.e., mapping) analyses.

Results: We identified a total of 8069 articles and ultimately included 14 models and frameworks in the review. These models and frameworks were comprised of 18 overlapping and 57 diverging elements, that were organized into six conceptual categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and spanned intrapersonal, interpersonal, process, and environment domains.

Conclusions: There is little overlap between the elements that comprise existing models and frameworks of patient engagement in health services research. Those seeking to apply these models and frameworks should consider the “fit” of each element, by conceptual category and domain, within the context of their study.

Trial registration: None.

Keywords

Patient and public involvement, stakeholder engagement, patient involvement, patient-oriented research, patient engagement

Article Summary

Strengths and limitations of this study

- The study's methodological strengths included its published protocol and analysis approach, which supported mapping model and framework elements to generate a toolbox of options for researchers to use in their own patient-engaged research.
- Another strength included the involvement of an interdisciplinary research team comprised of content experts, including patient co-researchers.
- A weakness of this study concerns the grey boundaries between health services research and other forms of health research.
- Greater engagement of patient co-researchers would have likely resulted in other study insights through more direct contact with the study.

Introduction

Patient engagement in research involves the formation of meaningful and active collaborations between academic researchers and patients (i.e., an overarching term that refers to individuals with personal experience of a health issue and informal caregivers) in research governance, priority setting, conduct, and knowledge translation [1]. Globally, it is also commonly referred to as patient and public involvement, patient involvement, and stakeholder engagement in research. This research approach necessitates a shift from the patient's traditional role as a study participant to that of a research collaborator or partner (i.e., patient co-researcher). This shift in roles and power dynamics reflects the approach's roots in participatory research [2] and is founded on the premises that those affected by a problem should be actively involved in the generation of solutions to it [3], and individuals' critical reflections on first-hand experiences are essential to effecting individual and social change [4]. A growing body of evidence supports the benefits of patient engagement in research, including improved enrollment and decreased attrition rates [5], increased relevance of research and accessibility of study materials to study participants [6], improved trial design [7], and increased meaningfulness and understandability of disseminated findings [5 6]. However, despite its underlying rationale and documented benefits, academic researchers report hesitance in adopting this research approach [8 9].

Some of the reported challenges of patient engagement in research include uncertainty about the process (i.e., "how-to" engage patients in research) and the need for a culture shift that supports these collaborations [8 9]. As there is no standard process for engaging patients in research, this first challenge is not surprising [5 6 10]. That is, how patient engagement is operationalized may vary according to the characteristics of a given project (e.g., design, scope, time, and financial resources) and patient and academic co-researchers (e.g., personal and professional backgrounds, interests, skills). Further, the underlying cultural shift needed to support patient engagement in research necessitates the redistribution of power and restructuring of traditional research paradigms to support shared planning and decision-making throughout a study [10]. This requires a mutual understanding and vision for what these research collaborations entail. An exploration of the models and frameworks that underlie patient engagement in research may help clarify the processes and support the culture shift necessary for this approach by shedding light on the universal elements that underlie it.

Three previous reviews have broadly synthesized the literature on models, frameworks and/or other systematic approaches to the engagement of patients (and in the case of Jull et al. - other knowledge users) in research [11-13]. Of these, only one described the elements that comprised identified frameworks and best practice guidelines [12]. Perhaps more importantly, none of these reviews focused on health services research, which entails considerations unique to health research. Specifically, health services research focuses on the impact of social factors, financing systems, organizational structures and processes, health technologies, and personal behaviours on health care access, quality and cost, and population health and well-being [14]. This differs substantially from general health research, where the goal of engagement is often to improve clinical outcomes, the effectiveness of a particular intervention, or uptake of research among patients with a specific condition. Engaging patients in health services research may require added considerations related to partnering with a more diverse group of patients with lived experience of different conditions, groups, interventions, and/or interactions with the health care system. Therefore, we set out to contribute to the existing literature on patient engagement in research by conducting a knowledge synthesis of models and frameworks of patient engagement in health services research. Specifically, we undertook a scoping review, which is "a type of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge [15]."

Objectives

The research question driving our review was, "What are the elements that underlie models and frameworks of patient engagement in health services research?" Our primary objective was to count and describe the elements that overlap (i.e., present in 2 or more models or frameworks) and diverge between identified models and frameworks. This objective intends to support a clearer understanding of similarities in thinking about patient engagement rather than to judge the relevance of elements or prescribe a "one size fits all" approach. Thus, rather than synthesizing the identified elements, we chose to map them according to the categories developed by the original authors and the themes that arose among them, with the vision of presenting a "toolbox" of potential approaches that researchers may choose from for their given research endeavor. Finally, the exploratory nature of our research question and our desire to identify and map key concepts that underlie patient engagement in research led us to adopt a scoping review methodology [16].

Methods

Our scoping review's design and conduct followed the methodological framework proposed by Arksey and O'Malley [17] and enhanced by Levac et al. [18]. Its underlying protocol, including definitions of underlying key concepts, is published elsewhere [19]. Reporting was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analysis: Extension for Scoping Review (PRISMA-ScR) [20] and the revised Guidance for Reporting Involvement of Patients and the Public (GRIPP2) short form [21] checklists. Research ethics approval was not required as this study did not involve participants.

Eligibility criteria: We included published and unpublished (i.e., grey) literature that presented (i) models or frameworks (ii) of patient engagement (iii) in health services research. Both original and adapted models and frameworks were eligible as long as they were developed in or for health services research. We excluded articles unavailable as full-text due to the limited descriptive information they provide (e.g., context, description of development, underlying elements) and not written in English due to feasibility-related considerations.

Information sources: The lead author (A.M.C.) searched six electronic databases CINAHL, Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLine, PsycINFO, and Scopus) and Google Scholar for published literature. Electronic databases (ProQuest Dissertations & Theses and Conference Proceedings Citation Index), Google, and the websites of key agencies (Canadian Institutes of Health Research, INVOLVE, Patient-Centered Outcomes Research Institute) were searched for unpublished literature. These searches were supplemented by snowball sampling, which entailed backwards and forwards reference searches of included articles and contacting experts in patient engagement in research for recommendations about any potentially relevant models or frameworks.

Searches: The search strategy was co-developed by an academic librarian (L.D.) and the lead author (A.M.C.) and finalized through collaboration with the rest of the research team. The search strategy used a combination of search terms related to our underlying concepts (i.e., “models,” “frameworks,” “patient engagement,” and “health services research”), was adapted to the syntax used by each database and website, and used Boolean terms. The search strategy for MEDLine is found in **Appendix 1**. There were no restrictions on publication dates. Searches of the published and unpublished literature were conducted on July 6, 2021 and July 7, 2021, respectively, while backward and forwards searches ended on January 13, 2022.

Selection: Search results for the six electronic databases and Google Scholar were imported into a reference management software (Endnote), and duplicate references were removed. Only the first 10 pages of Google Scholar results (n = 100 citations) were imported as advised by an academic librarian (L.D.). One reviewer (A.M.C.) conducted the level 1 (title) screening. The remaining relevant references were then imported into an online systematic review production and management software (Covidence; Veritas Health Innovation, Melbourne Australia), where two reviewers (A.M.C. and T.H.) independently conducted the level 2 (abstract) and level 3 (full-text) screening. Potentially relevant literature identified through websites and snowball sampling was screened for inclusion by both reviewers (A.M.C. and T.H.).

Data charting process: A standardized data charting (i.e., extraction) form was developed a priori in Microsoft Word by the study team (**Appendix 2**). Items were chosen that described the eligible models and frameworks, as well as provided context, including how and by who the models were developed. Two reviewers (A.M.C. and T.H.) independently extracted information from the final set of included articles using the form. They met with the senior author (A.S) to compare their data after extracting from an initial set of five and ten articles and upon completion of data extraction. Any inconsistencies were resolved through discussion and ultimately referring back to the original article. Further, in two instances [22 23], the lead author (A.M.C.) contacted the corresponding authors of included frameworks to obtain clarifying information. Consistent with methodological guidelines for scoping reviews, we did not appraise the methodological quality of included articles [15 16].

Synthesis of results: Data were compiled into a single Microsoft Excel (2019) spreadsheet for validation and synthesis, which included both quantitative (i.e., counts) and qualitative (i.e., mapping) analyses. In conducting the analyses, the lead author (A.M.C) first immersed herself in the included models and frameworks by thoroughly (and repeatedly) reading the associated literature and reviewing graphical representations (when available) along with element definitions. With increased familiarity, the lead author was able to combine and reframe (where appropriate) similar elements found in the original publications. The revised elements were used to obtain counts of overlapping and diverging elements. During this process, it became evident that the elements were organized into similar over-arching conceptual categories by the original authors, and that they could also be located within multiple domains (**Table 1**). As such, in our analysis, we mapped each element according to conceptual category and domain. The validity (i.e., content and face) of the final set of elements was established through discussion with this study's authors. Visual representations of the data (i.e., concept maps) were created using Mind Manager 2020 software, version 20.1 (Corel Corp., Austin, T.X.). To support applicability, some elements within the concept maps contain clarifying examples in brackets.

Table 1. Explanation of over-arching conceptual categories and domains

Over-arching conceptual category	Explanation
Principles	Values that orient and rule the conduct of a group
Foundational components	Core elements that comprise patient engagement
Contexts	Resources or decisions that are external to but inform the engagement process
Actions	Activities (e.g., behaviors, phases, advisory bodies) involved in the actual conduct of the engagement and associated research
Outcomes	Results of engagement and its associated research
Organizational levels	Different organizational levels at which engagement may occur in a research centre
Domain	**Elements pertain to…**
Intrapersonal	Individual-level knowledge, perceptions, attitudes, and beliefs
Interpersonal	Relationships with other people
Process	Carrying out the engagement or broader research
Environmental	Research or organizational policies, cultures, perceptions
Health systems and outcomes	Health systems and health outcomes

Patient involvement

We engaged two patient co-researchers (R.S. and S.H.), at the level of involve [24], in the design and conduct of this study. They were involved in development and publication of the scoping review protocol [19], provided feedback on the analyses (including element groupings), provided their perspectives on the interpretation of the study findings, and co-authored this manuscript. In helping shape these stages of the research cycle and related outputs, a notable impact of engagement also included identifying gaps in the current literature relevant to patient partners. The primary methods of engagement were 1-on-1 and small group meetings, as guided by a terms of reference co-developed at the outset of the research partnership. The patient co-researchers will continue to be involved in further knowledge translation activities, like synthesizing the findings for our research group's website (www.patientengagementinresearch.ca), co-presenting about the engagement process and study findings, and identifying other appropriate methods of dissemination.

Results

Flow of models and frameworks into the study

Appendix 3 displays the flow of eligible articles into the review (PRISMA flow chart). Of the 10,840 initially identified citations, 2771 duplicates were excluded. After applying inclusion criteria, 7150 articles were excluded at the title screen, 712 at the abstract screen, and 193 at the full-text screen, leaving 13 models and frameworks. One unpublished framework was then identified through snowball sampling, resulting in a total of 14 models and frameworks included in the review.

Characteristics of included articles and models/frameworks

Select descriptive characteristics of the included models and frameworks and the articles they were published in (where applicable) are presented in **Appendix 4**. Included articles were published between 2012 - 2020 (n = 10 were published in or after 2015) and based in the United States (n = 5), United Kingdom (n = 4), Canada (n = 2), Australia (n = 1) and United Kingdom and Wales (n = 1). The included unpublished framework was developed in Canada as part of the CONNECT project (Caroline Jose, Patricia George-Zwicker, Louise Tardif, Aaron Bouma, Darlene Pugsley, Luke Pugsley, Mathieu Bélanger, Jeffrey Gaudet, Marc Robichaud, CONNECT framework). Models and frameworks were developed in a variety of contexts including organizations (i.e., Patient-Centered Outcomes Research Institute, British Columbia Support Unit, Sunnybrook Hospital, PRIME Centre Wales), medical specialties (i.e., pediatric subspecialty care, palliative care), research disciplines (i.e., healthcare operations research), diseases (i.e., chronic or long-term conditions; cancer; dementia; stroke), and other health conditions (e.g., autistic adults, persons with lived experience of long-term physical and/or mental health illness, parents of children with disabilities). They were developed for general use as well in specific health services research contexts like healthcare operational research, practice-based research and innovation, pragmatic trials, patient-centered outcomes research, and comparative effectiveness research. None targeted a specific component of the research process, and patients were involved in the development of slightly over half (n = 8) of the included models/frameworks. Eight of the models/frameworks took into account the public participation spectrum [24] either explicitly or by including considerations related to control of decision-making and/or directionality of information exchange.

Overview of the elements of included models and frameworks

A total of 112 elements of patient engagement were identified across the 14 included models and frameworks. Combining and reframing similar elements reduced the total number to 75. Among these, 18 elements overlapped across the included models and frameworks and 57 diverged (i.e., were unique to individual models and frameworks). We present the elements by conceptual category and identify the domains they are situated in below. In considering these results, it should be noted that we placed select elements from the models by Deverka et al. [25] (i.e., inputs, methods, outputs) and Evans et al. [26] (i.e., opportunities for engagement, research environment that actively supports engagement and its underlying principles, resources to support engagement, and system to enhance the breadth of patient and carer experience brought to research activities) in the "actions" conceptual category despite that not being what the original authors labeled them. This is due to the fact that although these elements were uncategorized by Deverka et al. [25], they closely aligned with the actions category, and Evans et al.'s definition of the conceptual category that encompassed these elements closely aligned with other authors' conceptualization of "actions."

Principles

Principles represent the values that orient and rule the conduct of a group. They form the ethical backdrop of engagement [27] and underpin effective collaborative involvement in research [26]. For example, communication, which included mutual communication and feedback, was identified as a core principle of patient engagement by Evans et al. [26]. Figure 1 displays the 13 elements in this conceptual category, as reported by two articles [26 27]. These elements were situated in interpersonal (n = 7), process (n = 5), and environment (n = 1) domains.

Figure 1 about here

Figure 1. Concept map – principles of patient engagement in health services research

Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements that overlap *across* conceptual categories; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the environment domain.

Foundational components

Foundational components represent the core elements that comprise patient engagement in health services research (Figure 2). According to the eight models and frameworks [27-33] represented here, the 26 underlying elements were primarily situated in the process domain (n = 14 elements), with the remainder situated in intrapersonal (n = 7), interpersonal (n = 3), and environmental (n = 2) domains.

Figure 2 about here

Figure 2. Concept map – foundational components of patient engagement in health services research

Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the environment domain.

Contexts

This conceptual category identifies elements that relate to resources or decisions that are external to but inform the engagement process [34]. Figure 3 presents the four elements in this category, as contributed by one article [34]. Three of the underlying elements resided in the process, and one in the intrapersonal domains.

Figure 3 about here

Figure 3. Concept map – contexts of patient engagement in health services research

Figure legend: **Oval**: elements that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal domain; **Orange**: elements that reside in the process domain.

Actions

The elements within this category pertain to the activities (e.g., behaviors, phases, advisory bodies) involved in the actual engagement of patients in health services research. Figure 4 presents the elements that comprised this category, as reported by six articles [22 25-27 32 34]. These elements were primarily (n = 18) situated in the process domain, with one element in each of intrapersonal and environment domains, and two elements in the interpersonal domain. As displayed in the top half of elements in the process domain (Figure 4), some of the elements located here were conceptualized in terms of phases of research (i.e., preparatory, execution, translational phases and inputs, methods, outputs).

Figure 4 about here

Figure 4. Concept map – actions of patient engagement in health services research

Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the environment domain.

Outcomes

Figure 5 presents the elements that comprise the outcomes of patient engagement in health services research, based upon three articles [25 27 34] and the unpublished CONNECT framework. As reflected in the figure, all but the CONNECT framework further organized the elements by time frame (i.e., immediate/near-term, intermediate, and long-term), with three elements (i.e., health decision-making, research culture, and research outcomes) belonging to two time frames. Overall, this category's elements were primarily situated in the process (n = 11) and health systems and outcomes (n = 8) domains. A further three elements were situated in the intrapersonal, one in the interpersonal, and two in the environment domains.

Figure 5 about here

Figure 5. Concept map – outcomes of patient engagement in health services research

Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the environment domain; **Yellow**: elements that reside in the health system and outcomes domain.

Organizational levels

Two articles presented frameworks whose elements captured the organizational levels at which patient engagement in health services research occurred within research centres [23 35]. As displayed in Figure 6, these elements were located in environment (n = 3) and process (n = 1) domains.

Figure 6 about here

Figure 6. Concept map – organizational levels of patient engagement in health services research

Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the environment domain.

Discussion

Statement of principal findings

Our scoping review identified 14 models and frameworks of patient engagement in health services research, which were comprised of 18 overlapping and 57 diverging elements (n_{total} elements = 75). This work represents a novel contribution as, to our knowledge, it is the first to synthesize the literature on models and frameworks of patient engagement in health services research. Our approach to data synthesis is also unique in that we attempt to maximize the intuitiveness and applicability of our findings by presenting elements by overarching conceptual categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and corresponding domains (i.e., intrapersonal, interpersonal, process, and environment). We anticipate this approach will facilitate the ready application of our findings to readers' own research programs by serving as a "toolbox" of elements to consider according to the multi-level facets of a research team and study. That is, by considering the applicability of elements within each conceptual category and domain (as well as other elements that may come to mind based on the groupings' definitions), our findings can serve as a map that allows researchers to pick and choose elements based on their study, resources, research partners, and context.

Strengths and weaknesses of the study

Strengths of this study include its scoping review design, which enabled us to gain a broad perspective of the literature on models and frameworks of patient engagement in health services research. Other strengths include (i) a published protocol, (ii) the use of an established methodological framework to guide its design and conduct, (iii) the involvement of an interdisciplinary research team that included patient co-researchers with lived experience of engaging in health services research and other researchers with content expertise in health services research and participatory approaches (including patient engagement in research), and (iv) and the co-design of the search strategy with an academic librarian (LD) and the rest of the research team. A weakness of this study concerns the grey boundaries between health services research and other forms of health research, which may have resulted in the inclusion or exclusion of models or frameworks that others could argue do/do not belong in this review. We attempted to minimize this possibility through a screening process that utilized two reviewers (with a third to resolve discrepancies), an a priori agreed upon definition of health services research, and reaching out to study authors for clarification when necessary. Further, the engagement of patient co-researchers in our scoping review at the level of involve limited their opportunities to formally provide insights and expertise to pre-determined study milestones. Engagement at the level of collaborate or empower may have resulted in other insights through more direct contact with the study.

Comparison to other studies

Previous reviews of models, frameworks, and/or other systematic approaches to the engagement of patients and other knowledge users in research had a broader scope [11-13] (albeit only one involved a comprehensive search of multiple databases [11]) and different levels of analyses. Specifically, rather than report underlying elements, Greenhalgh et al. developed a taxonomy for the classification of identified systematic approaches (i.e., tools, frameworks, benchmarks, guidelines, and critical appraisal checklists) based on their primary focus and intended purpose (i.e., power-focused, priority-setting, study-focused, report-focused, partnership-focused) [11]. Innovatively, they piloted co-design workshops that aimed to improve the aesthetic appeal and usability of "best in-class" resources identified through the review. Similar to our questioning of the appropriateness of a "one-size fits all" framework, preliminary findings from their co-design workshops indicated that although stakeholders were presented with a common set of resources, they generated widely differing frameworks suited to meet their different needs and purposes. The review by Jull et al. reported 15 high-level concepts for knowledge user engagement in research that they identified through a directed content analysis of underlying elements [13]. Although they organized these concepts across four general research phases (i.e., prepare, plan, conduct, apply), they concluded that variation in the reported concepts between frameworks indicated that research teams should consider the concepts as fluid rather than strictly required. Lastly, the findings of Harrison et al.'s narrative review of frameworks and guidelines culminated in the proposal of an overarching framework that (similar in principle to our scoping review) conceptualized three distinct but inter-related elements of the patient engagement in research process - engagement foundational principles (i.e., domains that original study authors "… considered foundational to patient engagement in research;" n = 15 elements), engagement best practices (i.e., best practice activities to support engagement; n = 25 elements), and research phases where engagement should occur (n = 3 elements) [12].

The meaning of the study

Our findings indicate that the conceptualization of patient engagement in health services research varies between existing models and frameworks. Although models and frameworks posit to break down a concept into its base components (which implies the existence of convergence among underlying elements), this finding is not surprising as patient engagement is an *approach* to research rather than a *method*. Contributing to the heterogeneity among identified elements is the diversity in the populations, contexts, and approaches used to develop the models and frameworks, which also emphasizes the importance of avoiding a "one size fits all" approach to engagement. Perhaps congruence between models and frameworks actually exists at the level of conceptual categories. These could be taken to represent the essential components of patient engagement in health services research. The elements that underlie them, as identified across the various models and frameworks, would then serve as considerations for researchers and patient co-researchers when planning and operationalizing patient engagement in health services research. For this to be better developed from the current literature, consensus is needed on the definitions of the underlying categories, followed by some re-shuffling of the elements across the conceptual categories in order to align with the agreed upon definitions.

Unanswered questions and future research.

In sum, our study found that there is little overlap between the elements that comprise existing models and frameworks of patient engagement in health services research. Readers seeking to apply our findings to their own engaged work should consider the “fit” of each element, by conceptual category and domain, within the context of their study. Future research that builds upon our work should consider addressing three major gaps. First, most existing models and frameworks identified perceived deficits within patient co-researchers, with a lot less consideration paid to deficits that exist within academic co-researchers. This may be in large part due to the fact the majority of the models were written from an academic researcher lens (as evidenced, for example, by the fact that only a third of the included models and frameworks stated that they involved patients in their development). Second, there is a lack of focus on the “soft skills” that underlie interpersonal interactions and relationships (e.g., body language, wording, tone), as well as intrapersonal-level elements of patient engagement in health services research (e.g., attitudes, values, expectations). Third, patient co-researchers are not just “patients” that can be lumped into a single homogenous category. They are people with different backgrounds, skills, and interests extending beyond their health conditions or needs. Thus, it is important to incorporate a trauma-informed and intersectional approach that acknowledges and promotes an understanding of human beings as shaped by the interactions of different social locations and experiences [36]. Relatedly, it is important for patient co-researchers and academic researchers to get to know each other as people, instead of making assumptions (including about a patient co-researcher’s experience with healthcare services). Engagement is as much about relational interactions as it is research processes. Careful attention needs to be paid to both for academic-patient co-researcher relationships to thrive.

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Competing interests statement: None declared.

Author Contributions: A.M.C and A.S. were responsible for all major areas of concept development, study planning, and manuscript writing. In addition, A.M.C. was also responsible for data collection and analysis. T.H., L.D., C.S., R.S., and S.H. were responsible for all major areas of concept development and study planning, were consulted on the data analysis and its interpretation, and provided manuscript edits. In addition, T.H. was responsible for data collection. C.W. led concept development but passed away prior to the completion of the manuscript.

Data sharing statement: Data are available upon reasonable request to the corresponding author (https://orcid.org/0000-0001-8476-2093)

Ethics statements

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Ethics approval: This paper synthesized publicly available articles and therefore did not require ethical approval.

Word count: 4000

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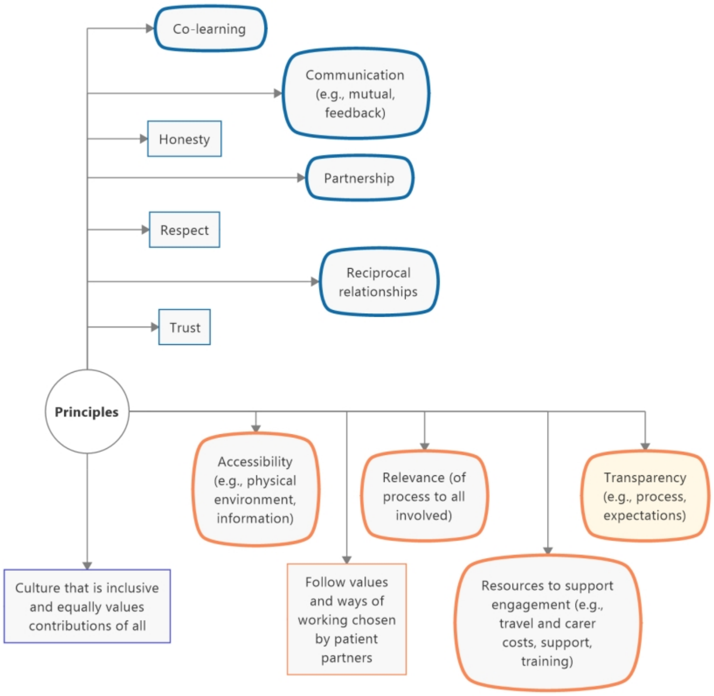


Figure 1. Concept map – principles of patient engagement in health services research
Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Blue: elements that reside in the interpersonal domain; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain.

430x322mm (118 x 118 DPI)

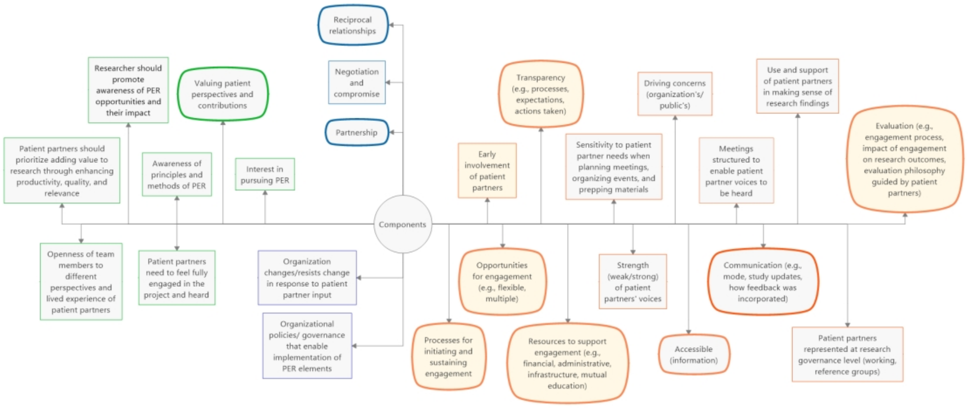


Figure 2. Concept map – foundational components of patient engagement in health services research
Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Blue: elements that reside in the interpersonal domain; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain.

430x322mm (118 x 118 DPI)

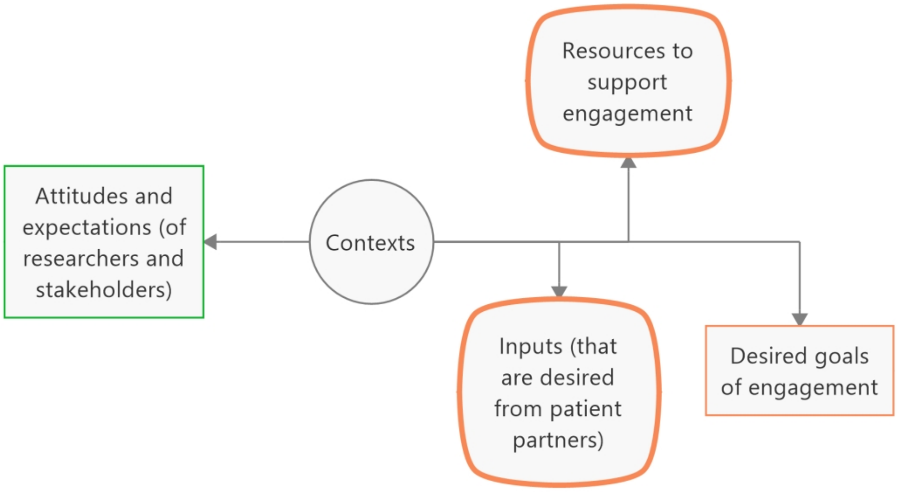


Figure 3. Concept map – contexts of patient engagement in health services research
Figure legend: Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Orange: elements that reside in the process domain.

430x322mm (118 x 118 DPI)

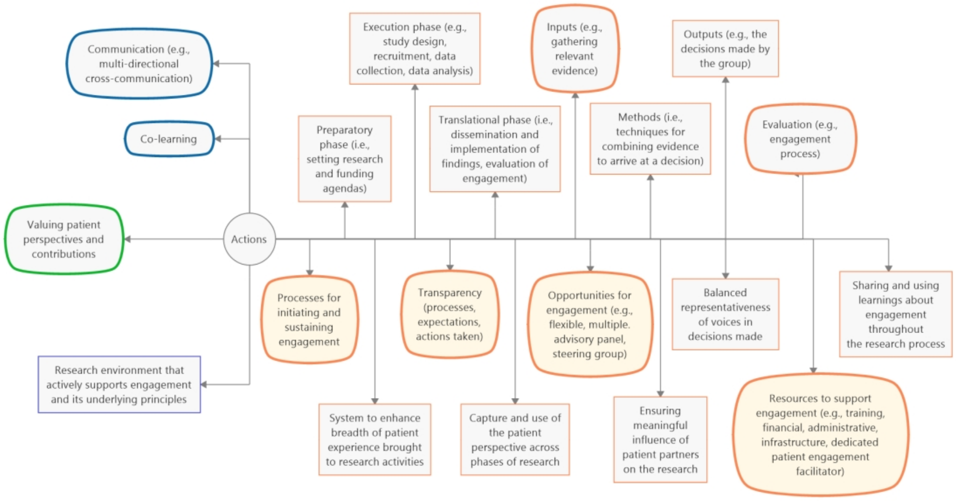


Figure 4. Concept map – actions of patient engagement in health services research
Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Blue: elements that reside in the interpersonal domain; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain.

430x322mm (118 x 118 DPI)

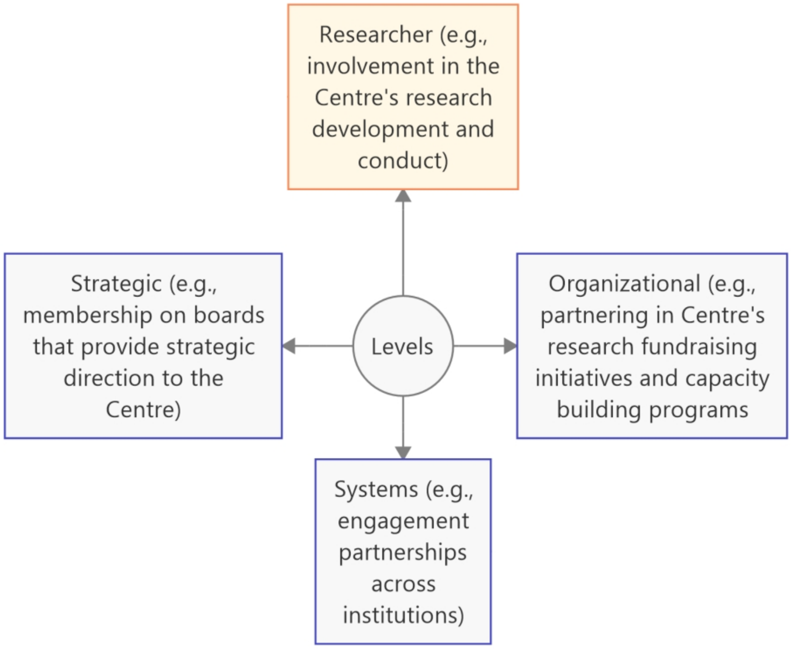


Figure 5. Concept map – outcomes of patient engagement in health services research
Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Blue: elements that reside in the interpersonal domain; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain; Yellow: elements that reside in the health system and outcomes domain.

430x322mm (118 x 118 DPI)

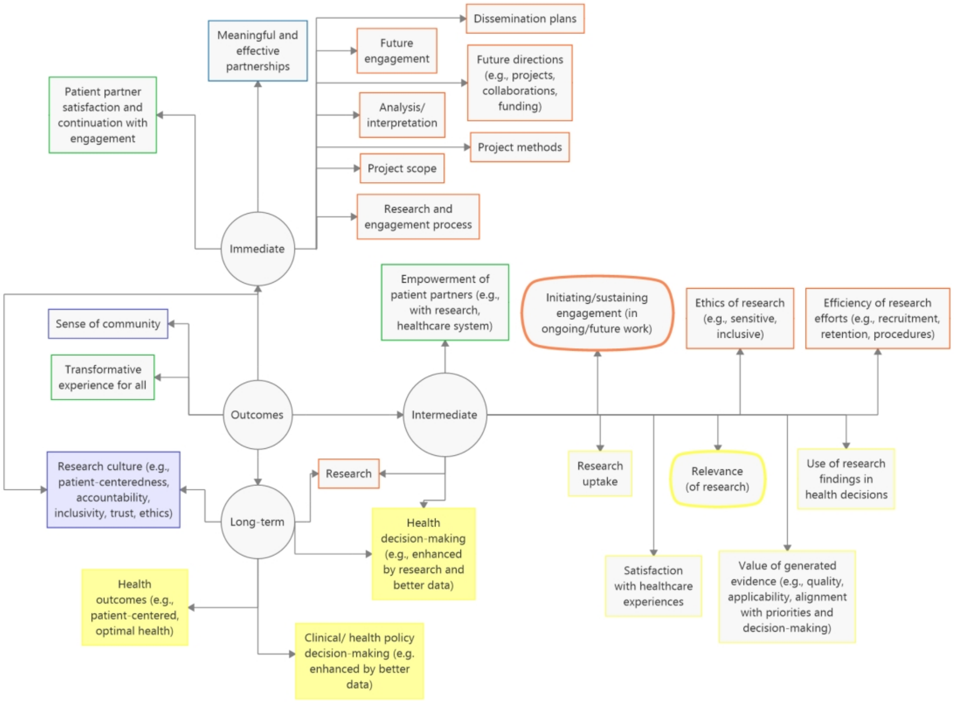


Figure 6. Concept map – organizational levels of patient engagement in health services research
Figure legend: Coloured: elements that overlap within conceptual categories; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain.

430x322mm (118 x 118 DPI)

Appendix 1 –Search strategy for MEDLINE (Ovid)

1. Patient Participation/

2. models, theoretical/ or patient-specific modeling/

3. (patient* adj3 engag*).mp.

4. (model or models or framework*).mp.

5. research.mp.

6. ("patient oriented research" or "patient and public involvement" or "patient involvement" or "stakeholder engagement").mp.

7. 1 or 3 or 6

8. 2 or 4

9. 5 and 7 and 8

Appendix 2 – Standardized data extraction form

Notes on using a data extraction form:

- Be consistent in the order and style you use to describe the information for each report.
- Record any missing information as unclear or not described, to make it clear that the information was not found in the study report(s), not that you forgot to extract it.
- Include any instructions and decision rules on the data collection form, or in an accompanying document. It is important to practice using the form and give training to any other authors using the form.

Decision rules

- Only include models and framework that are explicitly labelled by study authors as such

General Information

Date form completed *(dd/mm/yyyy)*	
Name of data extractor	

Study authors (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Author last names		
Country *(of authors' institutional affiliations)*		

Lens of authors *(by individuals, cluster/ groups or body parts)*		
Notes:		

Publication (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Study name		
Publication year		
Journal		
Is study published?	**Yes** **No**	
Notes:		

Model/framework (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Name of model/framework		
Purpose/aim of model/framework		

	Descriptions as stated in report/paper or note otherwise	Location in text or source *(pg & ¶/fig/table/other)*
Population developed in/for		
Methods used to develop m/f?		
Patients involved in m/f development (and if so how)?		
Targeted stage of research process		
Elements (description)		
Proposed relations between elements		
Location on spectrum of engagement		
Strs/weaknesses of model/framework (stated in study)		
Notes:		

Model/framework (figure presented in text)

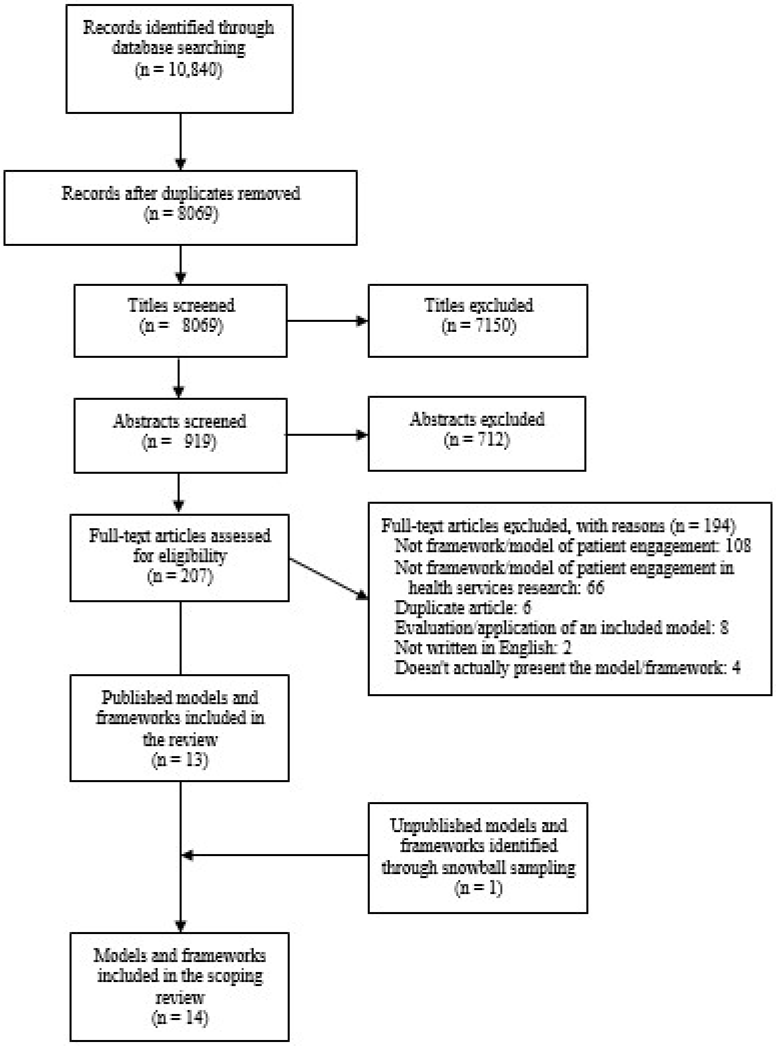
Definitions

Model	A descriptive and deliberate simplification of a phenomenon or an aspect of a phenomenon
Framework	A shared orientation for studying, explaining, and understanding phenomena of interest through the description and identification of the universal elements underlying a phenomenon of interest
Patient engagement	Meaningful and active collaboration of patients in research governance, priority setting, conduct, and knowledge translation
Health services research	The study of how social factors, financing systems, organizational structures and processes, health technologies, and personal behaviours affect access to health care, the quality and cost of health care, and, ultimately, the population's health and well-being. It includes research with the goal of improving the efficiency and effectiveness of health professionals and the health care system, through changes to practice and policy

Inclusion/exclusion criteria

Inclusion	**Exclusion**
Published or unpublished model or framework of	Models/frameworks not focused on health services research, such as those related to: • clinical decision-making/practice, • active role of patients in health management, • therapeutic engagement, • understanding disease experience • technology engagement • the pharmaceutical industry
Patient engagement in	Not written in English
Health services research	Unavailable as full-text

Appendix 3 – PRISMA diagram



Appendix 4. Select characteristics of included articles and/or models and frameworks

Characteristics of the …				
Article	**Model/framework**			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
Daveson/ [28] / UK/ 2015	Not specified	An optimal involvement model that fulfills the needs of both users and researchers while acknowledging any potential diversity between these groups	Consultation workshop attended by patients and caregivers	Yes – patients actively involved in Cicely Saunders Institute palliative care research
Deverka/ [25] / USA/ 2012	Conceptual model for stakeholder engagement in CER	A conceptual model for involving stakeholders in the CER process	Literature search, followed by practical experience with an existing stakeholder engagement process/project, and review and revision by an expert panel (including patients)	Yes - patients engaged with the Center for CANCERGEN project
Evans/ [26] / UK/ 2019	SUCCESS model	Not stated	Initially developed through a workshop, then reviewed and refined through meetings and email discussions.	Yes - people diagnosed with, or caring for someone with, a chronic or long-term condition
Evans/ [23] / UK and Wales/ 2020	PRIME Centre Wales	To enable and promote successful patient engagement across research activities within their research centre.	Informed by the research literature and the development of an engagement plan with the help of patients.	Yes – people with experience of using health services and caring for family members as well as people diagnosed with, or caring for someone

Characteristics of the …

Article	**Model/framework**			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
				with, a chronic or long-term condition
Frank/ [27] / USA/ 2015	Conceptual model for PCOR	To provide the basis for subsequent evaluative frameworks, to guide evaluation of PCOR, and to serve as a foundation for measurement models, to allow testing of hypothesized relationships between elements in the model.	Literature search and worked with a patient advisory panel	Yes - members of the PCORI Patient Engagement Advisory Panel
Gessell/ [29] / USA/ 2017	Not specified	To maximize stakeholder engagement within the context of a pragmatic trial	"Expanded" engagement standards that were previously proposed to PCORI standards	Not specified
Gibson/ [30] / UK/ 2017	Not specified	To map and evaluate patient engagement interactions in health services research	Adapted the researchers' previously developed model through workshops held with various patient groups	Yes - the language used within the adapted model passed through iterative changes involving both the researchers and workshop participants
Goeman/ [33] / Australia/ 2019	Model for successful inclusion of consumers and community	The successful inclusion of consumers in research	Co-design process involving expert working and reference groups	Yes - People living with dementia and their care-partners (“consumers”)

Article	Characteristics of the … Model/framework			
First author/ reference/ country/ year	Name	Purpose	Methods of development	Were patient co-researchers involved in its development (yes/no/not specified – population)
	representatives in research			
Holmes/ [22]/ Canada/ 2018	BC SUPPORT Unit Framework	To conceptualize and organize patient engagement activities	Not specified	Not specified
Pearson/ [31] / UK/ 2013	A provisional model for patient engagement in healthcare OR	A model for patient engagement in healthcare OR	Not specified	Not specified
Ray/ [34] / USA/ 2017	Conceptual model for understanding impact of stakeholder engagement and differentiating stakeholder-engaged research from research on stakeholder engagement	A framework for planning, evaluating, and reporting both stakeholder-engaged research and research on stakeholder engagement	Initial framework developed based on synthesis of existing literature; refined through use in a patient engagement research project which examined families' experiences of access to pediatric subspecialty care	No
Ruco/ [35] / Canada/ 2016	Sunnybrook Health Sciences Centre patient engagement in PBRI framework	To guide and capture the range and scope of patient engagement in PBRI at Sunnybrook Health Sciences Centre, Toronto	Developed based on CIHR's Supporting POR Visual Value Model for Patient Engagement and in alignment with Sunnybrook's PBRI Strategic plan	Not specified
Shippee/ [32] /	Framework for PSUE	To develop a standardized, evidence-based	Systematic review/environmental scan/manual	Yes - patient advisory group consisting of

Characteristics of the …

Article	**Model/framework**			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
USA/ 2013		framework for understanding, reporting, and assessing PSUE in biomedical and health services research	literature search that was synthesized into a two-part framework and supplemented with input from a patient advisory group	community member PSUEs
Jose/ Unpublished/ 2020	CONNECT framework	To engage members of the autistic community in research	Developed as part of work on the CONNECT project, including 2 feedback questionnaires designed for the study	Yes – members of the autistic community

CANCERGEN = comparative effectiveness research in cancer genomics; CER = comparative effectiveness research; CIHR = Canadian Institutes of Research; OR = operations research; PBRI = practice-based research and innovation; PCORI = patient-centered outcomes research institute; POR = patient-oriented research; PSUE = patient and service user engagement; SUCCESS = service users with chronic conditions encouraging sensible solutions.

Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	1
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	4-7
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	7
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	7-8
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	8
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	9
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	Appendix 1
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	9
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	9-10
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	Appendix 2
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	N/A

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	10
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	12, Appendix 3
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	12-13, Appendix 4
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	N/A
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	13-18
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	13-14
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	18-21
Limitations	20	Discuss the limitations of the scoping review process.	19
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	21-22
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	22-23

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.

* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.

† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).

‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.

§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy document).

From: Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMAScR): Checklist and Explanation. Ann Intern Med. 2018;169:467–473. doi: 10.7326/M18-0850.

GRIPP2-short form checklist

Section and topic	Item	Line numbers
1: Aim Report the aim of the study	Our primary objective was to count and describe the elements that overlap (i.e., present in 2 or more models or frameworks) and diverge between identified models and frameworks.	120-122
2: Methods Provide a clear description of the methods used for patient and public involvement (PPI) in the study	We engaged two patient co-researchers (R.S. and S.H.), at the level of involve (24), in the design and conduct of this study… The primary methods of engagement were 1-on-1 and small group meetings, as guided by a terms of reference co-developed at the outset of the research partnership.	207-208, 213-214
3: Results Outcomes—Report the results of PPI in the study, including both positive and negative outcomes	They were involved in development and publication of the scoping review protocol (19), provided feedback on the analyses (including element groupings), provided their perspectives on the interpretation of the study findings, and co-authored this manuscript.	208-210
4: Discussion Outcomes—Comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	In helping shape these stages of the research cycle and related outputs, a notable impact of engagement also included identifying gaps in the current literature relevant to patient partners.	210-212
5: Reflections Critical perspective—Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	Further, the engagement of patient co-researchers in our scoping review at the level of involve limited their opportunities to formally provide insights and expertise to pre-determined study milestones. Engagement at the level of collaborate or empower may have resulted in other insights through more direct contact with the study.	388-392

Adapted from: *BMJ 2017;358:j3453*

A scoping review of models and frameworks of patient engagement in health services research

Journal:	*BMJ Open*
Manuscript ID	bmjopen-2022-063507.R1
Article Type:	Original research
Date Submitted by the Author:	21-Jul-2022
Complete List of Authors:	Chudyk, Anna; University of Manitoba Faculty of Health Sciences Horrill, Tara; The University of British Columbia School of Nursing Waldman, Celeste; University of Manitoba Demczuk, Lisa; University of Manitoba Elizabeth Dafoe Library Shimmin, Carolyn; George and Fay Yee Centre for Healthcare Innovation Stoddard, Roger; Horizon Health Network Hickes, Serena; University of Manitoba Children's Hospital Research Institute of Manitoba Schultz, Annette; University of Manitoba College of Nursing
Primary Subject Heading:	Health services research
Secondary Subject Heading:	Research methods
Keywords:	STATISTICS & RESEARCH METHODS, HEALTH SERVICES ADMINISTRATION & MANAGEMENT, MEDICAL EDUCATION & TRAINING

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For peer review only

A scoping review of models and frameworks of patient engagement in health services research

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Abstract

Objective: To count and describe the elements that overlap (i.e., present in 2 or more) and diverge between models and frameworks of patient engagement in health services research. Our specific research question was, “What are the elements that underlie models and frameworks of patient engagement in health services research?”

Design: Scoping review.

Data sources: On July 6–7, 2021, we searched six electronic databases (CINAHL, Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLINE, PsycINFO, and Scopus) and Google Scholar for published literature, and ProQuest Dissertations & Theses, Conference Proceedings Citation Index, Google, and key agencies’ websites for unpublished (i.e., grey) literature, with no date restrictions. These searches were supplemented by snowball sampling.

Eligibility criteria: We included published and unpublished literature that presented (a) models or frameworks (b) of patient engagement (c) in health services research. We excluded articles unavailable as full-text or not written in English.

Data extraction and synthesis: Two independent reviewers extracted data from included articles using an a-priori developed standardized form. Data were synthesized using both quantitative (i.e., counts) and qualitative (i.e., mapping) analyses.

Results: We identified a total of 8069 articles and ultimately included 14 models and frameworks in the review. These models and frameworks were comprised of 18 overlapping and 57 diverging elements, that were organized into six conceptual categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and spanned intrapersonal, interpersonal, process, environmental, and health systems and outcomes domains.

Conclusions: There is little overlap between the elements that comprise existing models and frameworks of patient engagement in health services research. Those seeking to apply these models and frameworks should consider the "fit" of each element, by conceptual category and domain, within the context of their study.

Keywords

Patient and public involvement, stakeholder engagement, patient involvement, patient-oriented research, consumer and community involvement

Strengths and limitations of this study

- The study’s methodological strengths include use of a published protocol and the analysis approach, which supported mapping model and framework elements to generate a toolbox of options for researchers to use in their own patient-engaged research.
- Another strength is the involvement of an interdisciplinary research team comprised of content experts, including patient co-researchers.
- A weakness of this study concerns the grey boundaries between health services research and other forms of health research.
- Greater engagement of patient co-researchers would have likely resulted in other study insights through more direct contact with the study.

Introduction

Patient engagement in research involves the formation of meaningful and active collaborations between academic researchers and patients (i.e., an overarching term that refers to individuals with personal experience of a health issue and informal caregivers) in research governance, priority setting, conduct, and knowledge translation [1]. Globally, it is also commonly referred to as patient and public involvement, patient involvement, consumer and community involvement, and stakeholder engagement in research. This research approach necessitates a shift from the patient's traditional role as a study participant to that of a research collaborator or partner (i.e., patient co-researcher). This shift in roles and power dynamics reflects the approach's roots in participatory research [2] and is founded on the premises that those affected by a problem should be actively involved in the generation of solutions to it [3], and individuals' critical reflections on first-hand experiences are essential to effecting individual and social change [4]. A growing body of evidence supports the benefits of patient engagement in research, including improved enrollment and decreased attrition rates [5], increased relevance of research and accessibility of study materials to study participants [6], improved trial design [7], and increased meaningfulness and understandability of disseminated findings [5 6]. However, despite its underlying rationale and documented benefits, academic researchers report hesitance in adopting this research approach [8 9].

Some of the reported challenges of patient engagement in research include uncertainty about the process (i.e., "how-to" engage patients in research) and the need for a culture shift that supports these collaborations [8 9]. As there is no standard process for engaging patients in research, this first challenge is not surprising [5 6 10]. That is, how patient engagement is operationalized may vary according to the characteristics of a given project (e.g., design, scope, time, and financial resources) and patient and academic co-researchers (e.g., personal and professional backgrounds, interests, skills). Further, the underlying cultural shift needed to support patient engagement in research necessitates the redistribution of power and restructuring of traditional research paradigms to support shared planning and decision-making throughout a study [10]. This requires a mutual understanding and vision for what these research collaborations entail. An exploration of the models and frameworks that underlie patient engagement in research may help clarify the processes and support the culture shift necessary for this approach by shedding light on the universal elements that underlie it.

Three previous reviews have broadly synthesized the literature on models, frameworks and/or other systematic approaches to the engagement of patients (and in the case of Jull et al. - other knowledge users) in research [11-13]. Of these, only one described the elements that comprised identified frameworks and guidelines describing best practices for engaging patients in research[12]. Perhaps more importantly, none of these reviews focused on health services research, which entails considerations unique to health research. Specifically, health services research focuses on the impact of social factors, financing systems, organizational structures and processes, health technologies, and personal behaviours on health care access, quality and cost, and population health and well-being [14]. This differs substantially from general health research, where the goal of engagement is often to improve clinical outcomes, the effectiveness of a particular intervention, or uptake of research among patients with a specific condition. Engaging patients in health services research may require added considerations related to partnering with a more diverse group of patients with lived experience of different conditions, groups, interventions, and/or interactions with the health care system. Therefore, we set out to contribute to the existing literature on patient engagement in research by conducting a knowledge synthesis of models and frameworks of patient engagement in health services research. Specifically, we undertook a scoping review, which is "a type of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesizing existing knowledge" [15].

Objectives

The research question driving our review was, "What are the elements that underlie models and frameworks of patient engagement in health services research?" Our primary objective was to count and describe the elements that overlap (i.e., present in 2 or more models or frameworks) and diverge between identified models and frameworks. This objective intended to support a clearer understanding of similarities in thinking about patient engagement rather than to judge the relevance of elements or prescribe a "one size fits all" approach. Thus, rather than synthesizing the identified elements, we chose to map them according to the categories developed by the original authors and the themes that arose among them, with the vision of presenting a "toolbox" of potential approaches that researchers may choose from for their given research endeavor. Finally, the exploratory nature of our research question and our desire to identify and map key concepts that underlie patient engagement in research led us to adopt a scoping review methodology [16].

Methods

Our scoping review's design and conduct followed the methodological framework proposed by Arksey and O'Malley [17] and enhanced by Levac et al. [18]. The protocol for this scoping review, including definitions of underlying key concepts, is published elsewhere [19]. Reporting was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analysis: Extension for Scoping Review (PRISMA-ScR) [20] and the revised Guidance for Reporting Involvement of Patients and the Public (GRIPP2) short form [21] checklists. Research ethics approval was not required as this study did not involve participants.

Eligibility criteria

We included published and unpublished (i.e., grey) literature that presented (i) models or frameworks (ii) of patient engagement (iii) in health services research. Both original and adapted models and frameworks were eligible as long as they were developed in or for health services research. Since patient engagement also encompasses participation in research governance[1], we included models and frameworks that conceptualized how patient engagement in research was embedded across the different levels of health services research centers. We excluded articles unavailable as full-text due to the limited descriptive information they provide (e.g., context, description of development, underlying elements) and not written in English due to feasibility-related considerations.

Information sources

The lead author (A.M.C.) searched six electronic databases (CINAHL, Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLINE, PsycINFO, and Scopus) and Google Scholar for published literature. Electronic databases

(ProQuest Dissertations & Theses and Conference Proceedings Citation Index), Google, and the websites of key agencies (Canadian Institutes of Health Research, INVOLVE, Patient-Centered Outcomes Research Institute) were searched for unpublished literature. These searches were supplemented by snowball sampling, which entailed backwards and forwards reference searches of included articles and contacting experts in patient engagement in research for recommendations about any potentially relevant models or frameworks.

Searches

The search strategy was co-developed by an academic librarian (L.D.) and the lead author (A.M.C.) and finalized through discussion with the rest of the research team. The search strategy used a combination of search terms related to our underlying concepts (i.e., “models,” “frameworks,” “patient engagement,” and “health services research”), was adapted to the syntax used by each database, register, and website, and used Boolean terms. The search strategies for all databases, registers, and websites are found in **Appendix 1**. There were no restrictions on publication dates. Searches of the published and unpublished literature were conducted on July 6, 2021 and July 7, 2021, respectively, while backward and forwards searches ended on January 13, 2022.

Selection

Search results for the six electronic databases and Google Scholar were imported into a reference management software (Endnote), and duplicate references were removed. Only the first 10 pages of Google Scholar results (n = 100 citations) were imported as advised by an academic librarian (L.D.). One reviewer (A.M.C.) conducted the level 1 (title) screening. The remaining relevant references were then imported into an online systematic review production and management software (Covidence; Veritas Health Innovation, Melbourne Australia), where two reviewers (A.M.C. and T.H.) independently conducted the level 2 (abstract) and level 3 (full-text) screening. Potentially relevant literature identified through websites and snowball sampling was screened for inclusion by both reviewers (A.M.C. and T.H.). A third reviewer (A.S.) helped resolve discrepancies at the close of level 2 and 3 screening, which predominantly dealt with whether identified literature focused on health services research.

Data charting process

A standardized data charting (i.e., extraction) form was developed a priori in Microsoft Word by the study team (**Appendix 2**). Items were chosen that described the eligible models and frameworks, as well as provided context, including how and by who the models were developed. Two reviewers (A.M.C. and T.H.) independently extracted information from the final set of included articles using the form. They met with the senior author (A.S) to compare their data after extracting from an initial set of five and ten articles and upon completion of data extraction. Any inconsistencies were resolved through discussion and ultimately referring back to the original article. Further, in two instances [22 23], the lead author (A.M.C.) contacted the corresponding authors of included frameworks to obtain clarifying information. Consistent with methodological guidelines for scoping reviews, we did not appraise the methodological quality of included articles [15 16].

Synthesis of results

Data were compiled into a single Microsoft Excel (2019) spreadsheet for synthesis, which included both quantitative (i.e., counts) and qualitative (i.e., mapping) analyses. In conducting the analyses, the lead author (A.M.C) first immersed herself in the included models and frameworks by thoroughly (and repeatedly) reading the associated literature and reviewing graphical representations (when available) along with element definitions. With increased familiarity, the lead author was able to combine and reframe (where appropriate) similar elements found in the original publications. For example, ‘improved quality health decisions’ and ‘improved patient decision making’ were combined into the element ‘health decision-making’ based on similarities in the element names and descriptions provided by the original study authors. The revised elements were used to obtain counts of overlapping and diverging elements. During this process, it became evident that the elements were organized into similar over-arching conceptual categories by the original authors, and that they could also be located within multiple domains (**Table 1**). These categories and domains were identified inductively during the synthesis exercise. As such, in our analysis, we mapped each element according to conceptual category and domain. The validity (i.e., content and face) of the final set of elements was established through discussion with this study’s authors. Visual representations of the data (i.e., concept maps) were created using Mind Manager 2020 software, version 20.1 (Corel Corp., Austin, T.X.). To support applicability, some elements within the concept maps contain clarifying examples in brackets.

Table 1. Explanation of over-arching conceptual categories and domains

Over-arching conceptual category	Explanation
Principles	Values that orient and rule the conduct of a group.

Foundational components	Core elements that comprise patient engagement.
Contexts	Resources or decisions that are external to but inform the engagement process.
Actions	Activities (e.g., behaviors, phases, advisory bodies) involved in the actual conduct of the engagement and associated research.
Outcomes	Results of engagement and its associated research.
Organizational levels	Different organizational levels at which engagement may occur in a research organization.
Domain	**Elements pertain to…**
Intrapersonal	Individual-level knowledge, perceptions, attitudes, and beliefs.
Interpersonal	Relationships with other people.
Process	Carrying out the engagement or broader research.
Environmental	Research or organizational policies, cultures, perceptions.
Health systems and Outcomes	Health systems and health outcomes.

Patient and public involvement

We engaged two patient co-researchers (R.S. and S.H.), at the level of 'involve' [24], in the design and conduct of this study. The patient co-researchers and lead author (A.M.C.) arrived at this mutual decision at the study's outset, during the development of a terms of reference that guided the study's engagement process. By 'involve' we mean that the lead author worked consistently with patient co-researchers to ensure that their ideas and perspectives were understood and considered at study milestones. Specifically, patient co-researchers contributed to developing and publishing the scoping review protocol [19], provided feedback on the analyses (including element groupings), provided their perspectives on the interpretation of the study findings, and co-authored this manuscript. In helping shape these stages of the research cycle and related outputs, a notable impact of engagement also included identifying gaps in the current literature relevant to patient partners. The primary methods of engagement were 1-on-1 and small group meetings, as guided by a terms of reference co-developed at the outset of the research partnership. The patient co-researchers will continue to be involved in further knowledge translation activities, like synthesizing the findings for our research group's website (www.patientengagementinresearch.ca), co-presenting about the engagement process and study findings, and identifying other appropriate methods of dissemination.

Results

Flow of models and frameworks into the study

Appendix 3 displays the flow of eligible articles into the review (PRISMA flow chart). Of the 10,840 initially identified citations, 2771 duplicates were excluded. After applying inclusion criteria, 7150 articles were excluded at the title screen, 712 at the abstract screen, and 194 at the full-text screen, leaving 13 models and frameworks. One unpublished framework was then identified through snowball sampling, resulting in a total of 14 models and frameworks included in the review.

Characteristics of included articles and models/frameworks

Select descriptive characteristics of the included models and frameworks and the articles they were published in (where applicable) are presented in **Appendix 4**. Included articles were published between 2012 - 2020 (n = 10 were published in or after 2015) and based in the United States (n = 5), United Kingdom (n = 4), Canada (n = 2), Australia (n = 1) and United Kingdom and Wales (n = 1). The included unpublished framework was developed in Canada as part of the CONNECT project (Caroline Jose, Patricia George-Zwicker, Louise Tardif, Aaron Bouma, Darlene Pugsley, Luke Pugsley, Mathieu Bélanger, Jeffrey Gaudet, Marc Robichaud, CONNECT framework). Models and frameworks were developed in a variety of contexts including organizations (i.e., Patient-Centered Outcomes Research Institute, British Columbia Support Unit, Sunnybrook Hospital, PRIME Centre Wales), medical specialties (i.e., pediatric subspecialty care, palliative care), research disciplines (i.e., healthcare operations research), diseases (i.e., chronic or long-term conditions; cancer; dementia; stroke), and other health conditions (e.g., autistic adults, persons with lived experience of long-term physical and/or mental health illness, parents of children with disabilities). They were developed for general use as well in specific health services research contexts like healthcare operational research, practice-based research and innovation, pragmatic trials, patient-centered outcomes research, and comparative effectiveness research. None targeted a specific component of the research process, and patients were involved in the development of slightly over half (n = 8) of the included models/frameworks. Eight of the models/frameworks took into account the public participation spectrum [24] either explicitly or by including considerations related to control of decision-making and/or directionality of information exchange.

Overview of the elements of included models and frameworks

A total of 112 elements of patient engagement were identified across the 14 included models and frameworks. Combining and reframing similar elements reduced the total number to 75. Among these, 18 elements overlapped across the included models and frameworks and 57 diverged (i.e., were unique to individual models and frameworks). We present the elements by conceptual category and identify the domains they are situated in below. In considering these results, it should be noted that we placed select elements from the models by Deverka et al. [25] (i.e., inputs, methods, outputs) and Evans et al. [26] (i.e., opportunities for engagement, research environment that actively supports engagement and its underlying principles, resources to support engagement, and system to enhance the breadth of patient and carer experience brought to research activities) in the "actions" conceptual category despite that not being what the original authors labeled them. This is due to the fact that although these elements were uncategorized by Deverka et al. [25], they closely aligned with the actions category, and Evans et al.'s definition of the conceptual category that encompassed these elements closely aligned with other authors' conceptualization of "actions."

Principles

Principles represent the values that orient and rule the conduct of a group. They form the ethical backdrop of engagement [27] and underpin effective collaborative involvement in research [26]. For example, communication, which included mutual communication and feedback, was identified as a core principle of patient engagement by Evans et al. [26]. Figure 1 displays the 13 elements in this conceptual category, as reported by two articles [26 27]. These elements were situated in interpersonal (n = 7), process (n = 5), and environmental (n = 1) domains.

Foundational components

Foundational components represent the core elements that comprise patient engagement in health services research (Figure 2). According to the eight models and frameworks [27-33] represented here, the 26 underlying elements were primarily situated in the process domain (n = 14 elements), with the remainder situated in intrapersonal (n = 7), interpersonal (n = 3), and environmental (n = 2) domains.

Contexts

This conceptual category identifies elements that relate to resources or decisions that are external to but inform the engagement process [34]. Figure 3 presents the four elements in this category, as contributed by one article [34]. Three of the underlying elements resided in the process, and one in the intrapersonal domains.

Actions

The elements within this category pertain to the activities (e.g., behaviors, phases, advisory bodies) involved in the actual engagement of patients in health services research. Figure 4 presents the elements that comprised this category, as reported by six articles [22 25-27 32 34]. These elements were primarily (n = 18) situated in the process domain, with one element in each of intrapersonal and environmental domains, and two elements in the interpersonal domain. As displayed in the top half of elements in the process domain (Figure 4), some of the elements located here were conceptualized in terms of phases of research (i.e., preparatory, execution, translational phases and inputs, methods, outputs).

Outcomes

Figure 5 presents the elements that comprise the outcomes of patient engagement in health services research, based upon three articles [25 27 34] and the unpublished CONNECT framework. As reflected in the figure, all but the CONNECT framework further organized the elements by time frame (i.e., immediate/near-term, intermediate, and long-term), with three elements (i.e., health decision-making, research culture, and research outcomes) belonging to two time frames. Overall, this category's elements were primarily situated in the process (n = 11) and health systems and outcomes (n = 8) domains. A further three elements were situated in the intrapersonal, one in the interpersonal, and two in the environmental domains.

Organizational levels

Two articles presented frameworks whose elements captured the organizational levels at which patient engagement in health services research occurred within research centres [23 35]. As displayed in Figure 6, these elements were located in environmental (n = 3) and process (n = 1) domains.

Discussion

Principal findings

Our scoping review identified 14 models and frameworks of patient engagement in health services research, which were comprised of 18 overlapping and 57 diverging elements (n_{total} elements = 75). This work represents a novel contribution as, to our knowledge, it is the first to synthesize the literature on models and frameworks of patient engagement in health services research. Our approach to data synthesis is also unique in that we attempt to maximize the intuitiveness and applicability of our findings by presenting elements by overarching conceptual categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and corresponding domains (i.e., intrapersonal, interpersonal, process, environmental, and health systems and outcomes). We anticipate this approach will facilitate the ready application of our findings to readers' own research programs by serving as a "toolbox" of elements to consider according to the multi-level facets of a research team and study. To illustrate, research partners could begin to meet at a study's outset to co-develop terms of reference that guide the relational and activity related aspects of the study's engagement process. In doing so, they could reflect upon whether/how elements within the domains of each conceptual category resonate with their study's engagement process (as influenced by factors such as the study design, available resources, research partner strengths and interests, etc.) These conversations could be guided by prompts such as "do we want to embody or incorporate this element within our study (why/why not)?", "what does the embodiment or incorporation of this element look and feel like to us?", and "how will we know when we have or have not embodied or incorporated this element within our study?" In doing so, the "toolbox" of elements found within our review is transformed into a co-developed "roadmap" to help guide a study's engagement process.

Strengths and weaknesses of the study

Strengths of this study include its scoping review design, which enabled us to gain a broad perspective of the literature on models and frameworks of patient engagement in health services research. Other strengths include (i) a published protocol, (ii) the use of an established methodological framework to guide its design and conduct, (iii) the involvement of an interdisciplinary research team that included patient co-researchers with lived experience of engaging in health services research and other researchers with content expertise in health services research and participatory approaches (including patient engagement in research), and (iv) and the co-design of the search strategy with an academic librarian (LD) and the rest of the research team. A weakness of this study concerns the grey boundaries between health services research and other forms of health research, which may have resulted in the inclusion or exclusion of models or frameworks that others could argue do/do not belong in this review. We attempted to minimize this possibility through a screening process that utilized two reviewers (with a third to resolve discrepancies), an a priori agreed upon definition of health services research, and reaching out to study authors for clarification when necessary. Further, the engagement of patient co-researchers in our scoping review at the level of involve limited their opportunities to formally provide insights and expertise to pre-determined study milestones. Engagement at the level of collaborate or empower may have resulted in other insights through more direct contact with the study.

Comparison with other studies

Previous reviews of models, frameworks, and/or other systematic approaches to the engagement of patients and other knowledge users in research had a broader scope [11-13] (albeit only one involved a comprehensive search of multiple databases [11]) and different levels of analyses. Specifically, rather than report underlying elements, Greenhalgh et al. developed a taxonomy for the classification of identified systematic approaches (i.e., tools, frameworks, benchmarks, guidelines, and critical appraisal checklists) based on their primary focus and intended purpose (i.e., power-focused, priority-setting, study-focused, report-focused, partnership-focused) [11]. Innovatively, they piloted co-design workshops that aimed to improve the aesthetic appeal and usability of “best in-class” resources identified through the review. Similar to our questioning of the appropriateness of a "one-size fits all" framework, preliminary findings from their co-design workshops indicated that although stakeholders were presented with a common set of resources, they generated widely differing frameworks suited to meet their different needs and purposes. The review by Jull et al. reported 15 high-level concepts for knowledge user engagement in research that they identified through a directed content analysis of underlying elements [13]. Although they organized these concepts across four general research phases (i.e., prepare, plan, conduct, apply), they concluded that variation in the reported concepts between frameworks indicated that research teams should consider the concepts as fluid rather than strictly required. Lastly, the findings of Harrison et al.'s narrative review of frameworks and guidelines culminated in the proposal of an overarching framework that (similar in principle to our scoping review) conceptualized three distinct but inter-related elements of the patient engagement in research process - engagement foundational principles (i.e., domains that original study authors "… considered foundational to patient engagement in research;" n = 15 elements), engagement best practices (i.e., best practice activities to support engagement; n = 25 elements), and research phases where engagement should occur (n = 3 elements) [12]. Taken together, existing reviews demonstrate diversity in how patient engagement in research has been conceptualized through models, frameworks, and other systematic approaches and the approaches used to synthesize their findings.

The meaning of the study

Our findings indicate that the conceptualization of patient engagement in health services research varies between existing models and frameworks. Although models and frameworks posit to break down a concept into its base components (which implies the existence of convergence among underlying elements), this finding is not surprising as patient engagement is an *approach* to research rather than a *method*. Contributing to the heterogeneity among identified elements is the diversity in the populations, contexts, and approaches used to develop the models and frameworks, which also emphasizes the importance of avoiding a “one size fits all” approach to engagement. Perhaps congruence between models and frameworks actually exists at the level of conceptual categories. These could be taken to represent the essential components of patient engagement in health services research. The elements that underlie them, as identified across the various models and frameworks, would then serve as considerations for researchers and patient co-researchers when planning and operationalizing patient engagement in health services research. For this to be better developed from the current literature, consensus is needed on the definitions of the underlying categories, followed by some re-shuffling of the elements across the conceptual categories in order to align with the agreed upon definitions.

Unanswered questions and future research

In sum, our study found that there is little overlap between the elements that comprise existing models and frameworks of patient engagement in health services research. Readers seeking to apply our findings to their own engaged work should consider the “fit” of each element, by conceptual category and domain, within the context of their study. Future research that builds upon our work should consider addressing three major gaps. First, most existing models and frameworks identified factors that needed improvement for patient co-researchers to be better research partners, with a lot less consideration paid to factors that needed improvement for academic co-researchers to be better research partners. This may be in large part due to the fact the majority of the models were written from an academic researcher lens (as evidenced, for example, by the fact that only a third of the included models and frameworks stated that they involved patients in their development). Second, there is a lack of focus on the intrapersonal domain of engagement, including the "soft skills" that underlie interpersonal interactions and relationships (e.g., body language, wording, tone), and the environmental domain of engagement which shapes the context in which engagement is set. Third, patient co-researchers are not just "patients" that can be lumped into a single homogenous category. They are people with different backgrounds, skills, and interests extending beyond their health conditions or needs. Thus, it is important to incorporate a trauma-informed and intersectional approach that acknowledges and promotes an understanding of human beings as shaped by the interactions of different social locations and experiences [36]. Relatedly, it is important for patient co-researchers and academic researchers to get to know each other as people, instead of making assumptions (including about a patient co-researcher's experience with healthcare services). Engagement is as much about relational interactions as it is research processes. Careful attention needs to be paid to both for academic-patient co-researcher relationships to thrive.

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Contributors: A.M.C and A.S. were responsible for all major areas of concept development, study planning, and manuscript writing. In addition, A.M.C. was also responsible for data collection and analysis. T.H., L.D., C.S., R.S., and S.H. were responsible for all major areas of concept development and study planning, were consulted on the data analysis and its interpretation, and provided manuscript edits. In addition, T.H. was responsible for data collection. C.W. led concept development but passed away prior to the completion of the manuscript.

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Ethics approval: This paper synthesized publicly available articles and therefore did not require ethical approval.

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FIGURE TITLES

Figure 1. Concept map – principles of patient engagement in health services research

Figure 2. Concept map – foundational components of patient engagement in health services research

Figure 3. Concept map – contexts of patient engagement in health services research

Figure 4. Concept map – actions of patient engagement in health services research

Figure 5. Concept map – outcomes of patient engagement in health services research

Figure 6. Concept map – organizational levels of patient engagement in health services research

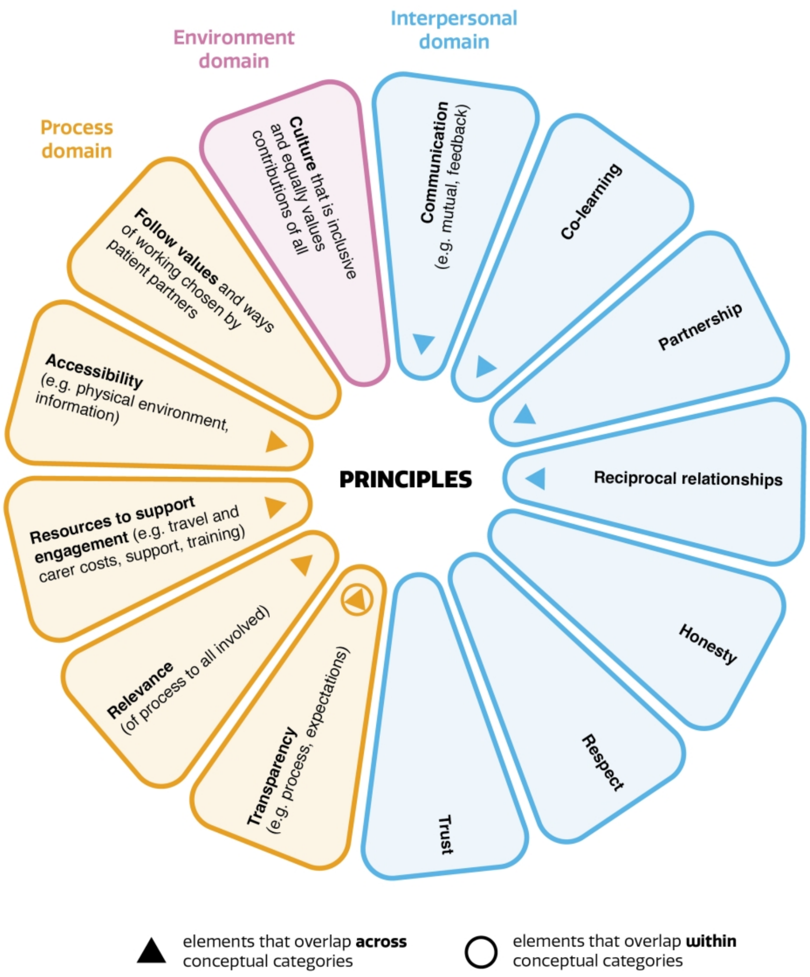


Figure 1. Concept map – principles of patient engagement in health services research

548x580mm (118 x 118 DPI)

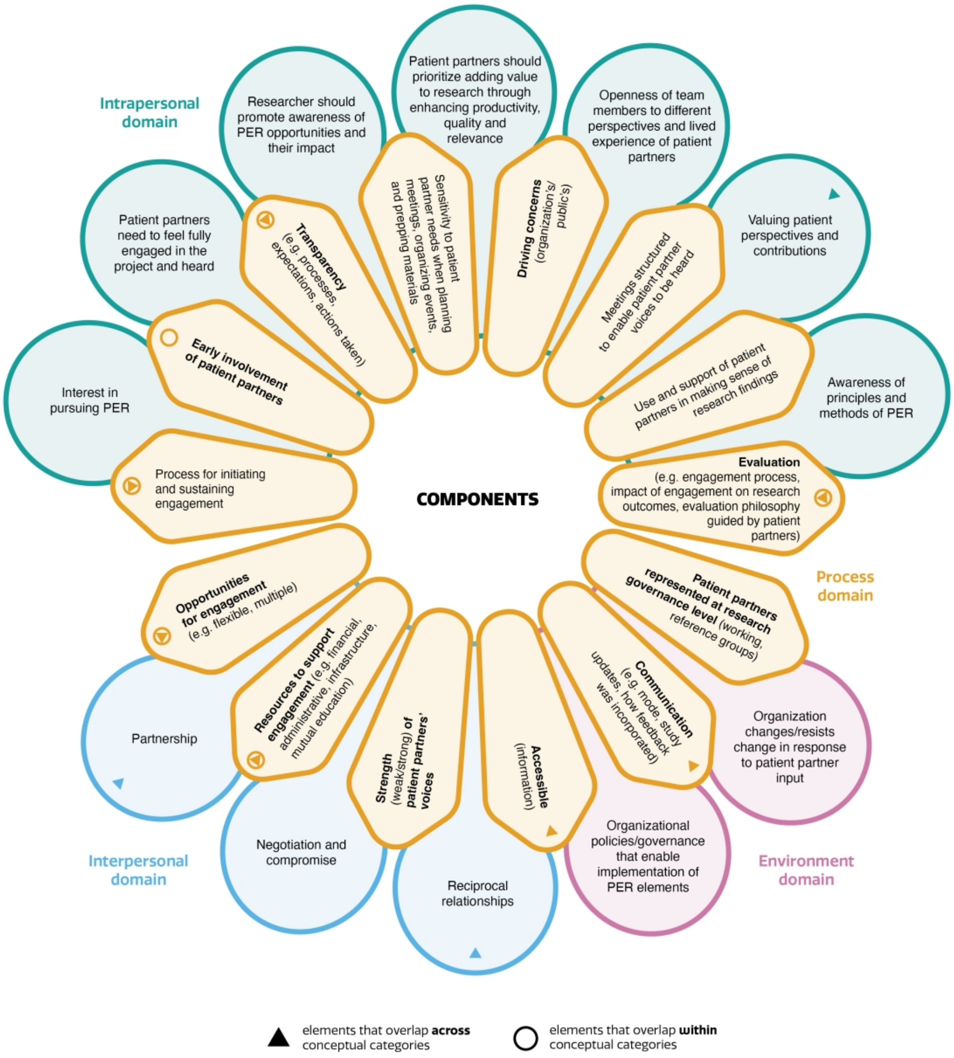


Figure 2. Concept map – foundational components of patient engagement in health services research

696x773mm (118 x 118 DPI)

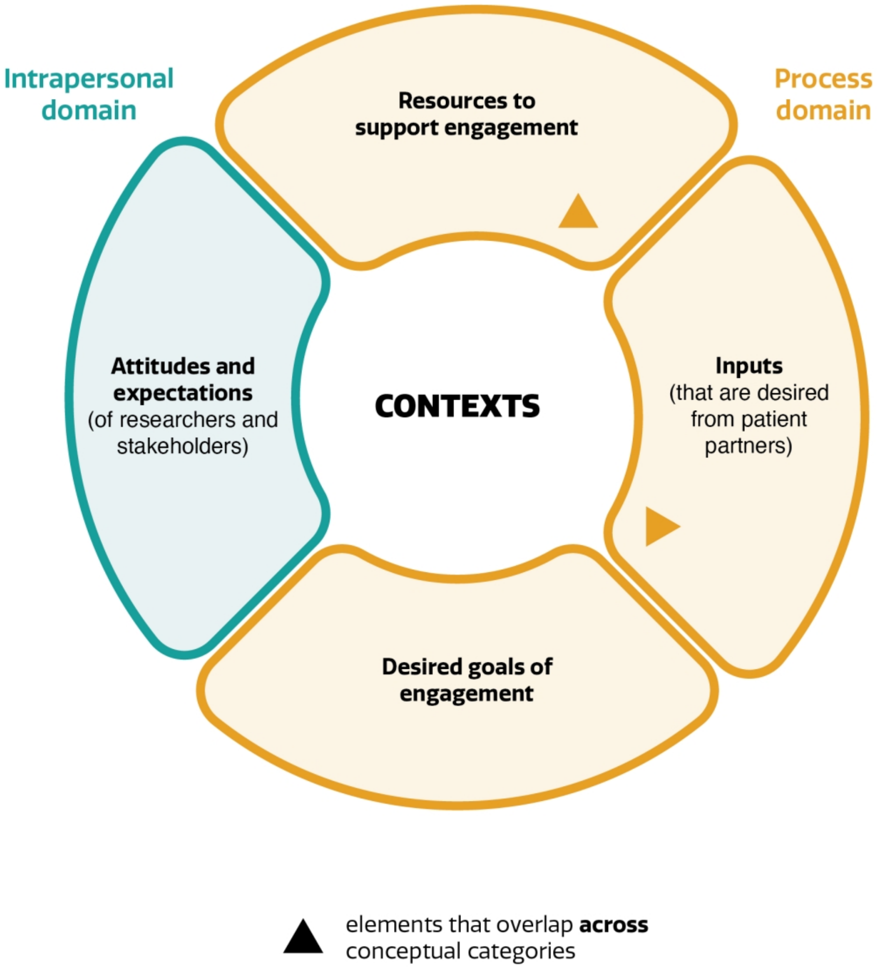


Figure 3. Concept map – contexts of patient engagement in health services research

406x436mm (118 x 118 DPI)

Figure 4. Concept map – actions of patient engagement in health services research

548x656mm (118 x 118 DPI)

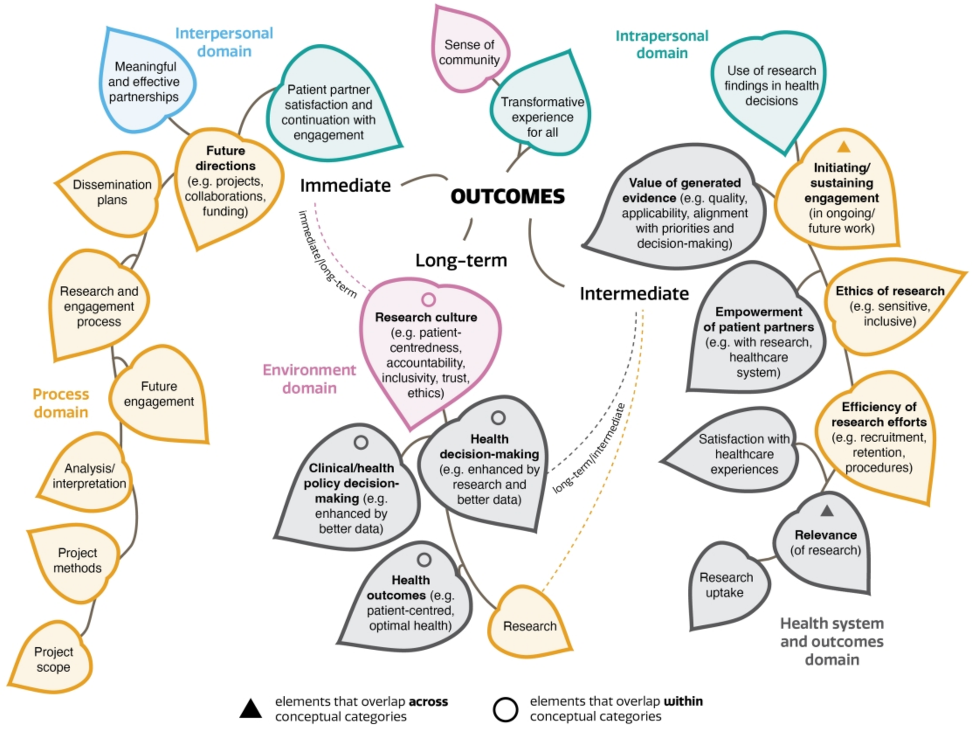


Figure 5. Concept map – outcomes of patient engagement in health services research

710x549mm (118 x 118 DPI)

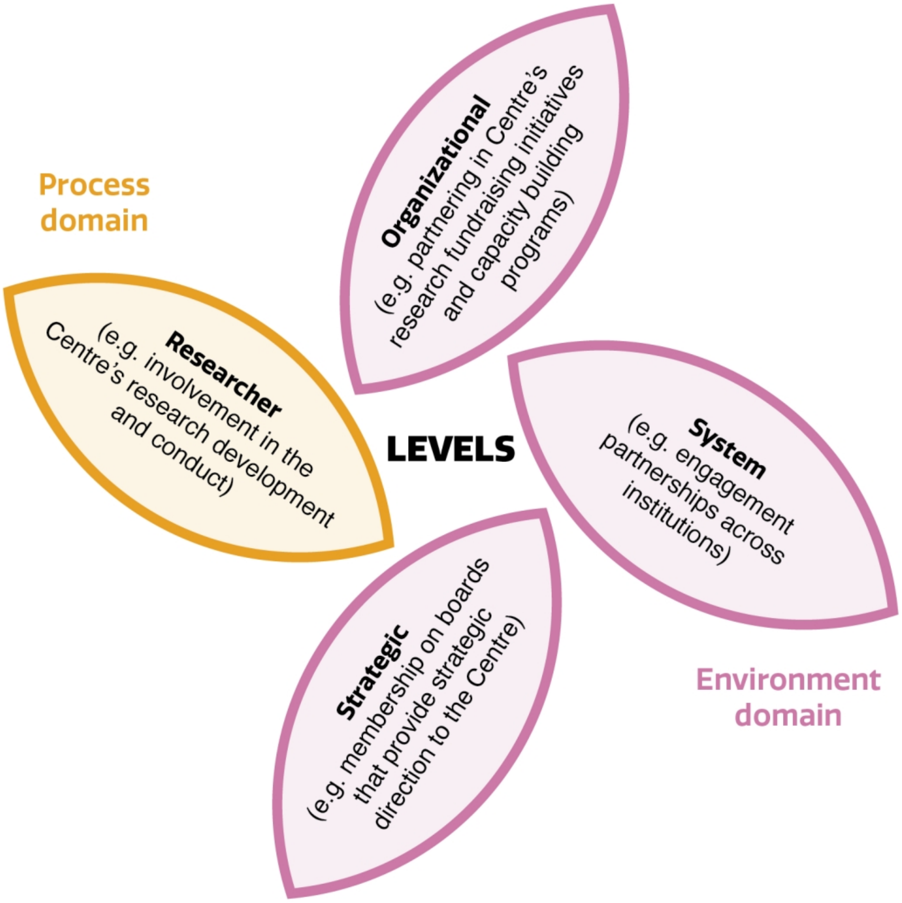


Figure 6. Concept map – organizational levels of patient engagement in health services research

360x373mm (118 x 118 DPI)

Appendix 1 –Search strategies for all databases, registers, and websites

CINAHL

1. (patient* n3 engag*) OR “patient oriented research”
2. model OR models OR framework* OR (MH “Conceptual Framework”) OR (MH “Models, Theoretical”)
3. research
4. “patient and public involvement” OR “patient involvement” or “stakeholder engagement”
5. S1 OR S4
6. S2 AND S3 AND S5

Cochrane Database of Systematic Reviews

“patient engagement” OR “patient oriented research” OR “patient and public involvement” OR “patient involvement” OR “stakeholder engagement” in Title Abstract Keyword

Joanna Briggs Institute Evidence Based Practice Database

(“patient engagement” OR “patient oriented research” OR “patient and public involvement” OR “patient involvement” OR “stakeholder engagement”).mp.

MEDLINE (Ovid)

1. Patient Participation/
2. models, theoretical/ or patient-specific modeling/
3. (patient* adj3 engag*).mp.
4. (model or models or framework*).mp.
5. research.mp.
6. ("patient oriented research" or "patient and public involvement" or "patient involvement" or "stakeholder engagement").mp.
7. 1 or 3 or 6
8. 2 or 4
9. 5 and 7 and 8

Scopus

((TITLE-ABS-KEY (patient* W/3 engag*)) OR (TITLE-ABS-KEY (“patient oriented research”)) OR (TITLE-ABS-KEY (“patient and public involvement”)) OR (TITLE-ABS-KEY (“patient involvement”)) OR (TITLE-ABS-KEY (“stakeholder engagement”))) AND (TITLE-ABS-KEY (model or models or framework*)) AND (TITLE-ABS-KEY (research))

PsycINFO

1. Client Participation/
2. Models/
3. (patient* adj3 engag*).mp.
4. (model OR models OR framework*).mp.
5. research.mp.
6. (“patient oriented research” OR “patient and public involvement” OR “patient involvement” or “stakeholder engagement”).mp.
7. 1 OR 3 OR 6
8. 2 OR 4
9. 5 AND 7 AND 8

Web of Science - Conference Proceedings Citation Index

(TS=(patient* NEAR/3 engag*) OR (TS=("patient oriented research" OR "patient and public involvement" OR "patient involvement" OR "stakeholder engagement"))) AND (TS=(model OR models OR framework*)) AND (TS=(research))

Google Scholar

("patient engagement" OR "patient oriented research" OR “patient and public involvement” OR “patient involvement” OR “stakeholder engagement”) AND (model or framework) AND research

Conference Proceedings Citation Index

(TS=(patient* NEAR/3 engag*) OR (TS=("patient oriented research" OR "patient and public involvement" OR "patient involvement" OR "stakeholder engagement"))) AND (TS=(model OR models OR framework*)) AND (TS=(research))

ProQuest Dissertations & Theses

noft((patient* near/3 engag*) OR "patient oriented research" OR "patient and public involvement" OR "patient involvement" OR "stakeholder engagement") AND noft(model OR models OR framework*) AND noft(research)

Google and websites

(model OR framework) AND “health services research”

Appendix 2 – Standardized data extraction form

Notes on using a data extraction form:

- Be consistent in the order and style you use to describe the information for each report.
- Record any missing information as unclear or not described, to make it clear that the information was not found in the study report(s), not that you forgot to extract it.
- Include any instructions and decision rules on the data collection form, or in an accompanying document. It is important to practice using the form and give training to any other authors using the form.

Decision rules

- Only include models and framework that are explicitly labelled by study authors as such

General Information

Date form completed *(dd/mm/yyyy)*	
Name of data extractor	

Study authors (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Author last names		
Country *(of authors' institutional affiliations)*		

Lens of authors *(by individuals, cluster/ groups or body parts)*		
Notes:		

Publication (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Study name		
Publication year		
Journal		
Is study published?	**Yes** **No**	
Notes:		

Model/framework (characteristics of)

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Name of model/framework		
Purpose/aim of model/framework		

	Descriptions as stated in report/paper or note otherwise	**Location in text or source** *(pg & ¶/fig/table/other)*
Population developed in/for		
Methods used to develop m/f?		
Patients involved in m/f development (and if so how)?		
Targeted stage of research process		
Elements (description)		
Proposed relations between elements		
Location on spectrum of engagement		
Strs/weaknesses of model/framework (stated in study)		
Notes:		

Model/framework (figure presented in text)

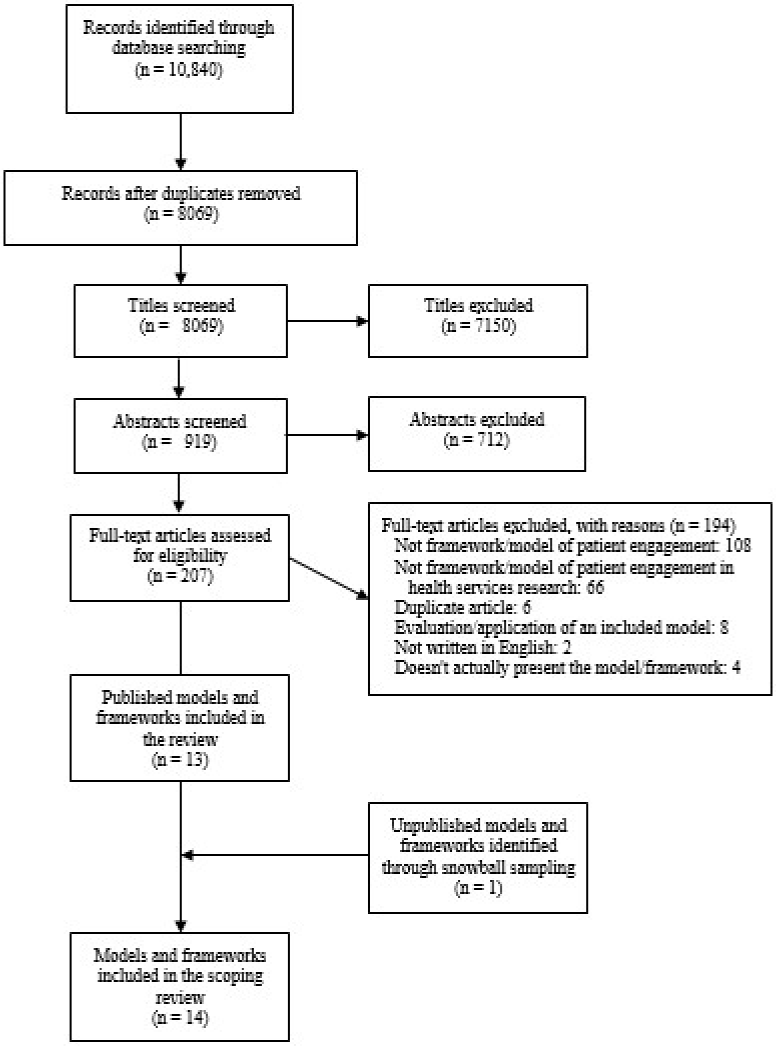
Definitions

Model	A descriptive and deliberate simplification of a phenomenon or an aspect of a phenomenon
Framework	A shared orientation for studying, explaining, and understanding phenomena of interest through the description and identification of the universal elements underlying a phenomenon of interest
Patient engagement	Meaningful and active collaboration of patients in research governance, priority setting, conduct, and knowledge translation
Health services research	The study of how social factors, financing systems, organizational structures and processes, health technologies, and personal behaviours affect access to health care, the quality and cost of health care, and, ultimately, the population's health and well-being. It includes research with the goal of improving the efficiency and effectiveness of health professionals and the health care system, through changes to practice and policy

Inclusion/exclusion criteria

Inclusion	**Exclusion**
Published or unpublished model or framework of	Models/frameworks not focused on health services research, such as those related to: • clinical decision-making/practice, • active role of patients in health management, • therapeutic engagement, • understanding disease experience • technology engagement • the pharmaceutical industry
Patient engagement in	Not written in English
Health services research	Unavailable as full-text

Appendix 3 – PRISMA diagram



Appendix 4. Select characteristics of included articles and/or models and frameworks

Article	Characteristics of the … Model/framework			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
Daveson/ [28] / UK/ 2015	Not specified	An optimal involvement model that fulfills the needs of both users and researchers while acknowledging any potential diversity between these groups	Consultation workshop attended by patients and caregivers	Yes – patients actively involved in Cicely Saunders Institute palliative care research
Deverka/ [25] / USA/ 2012	Conceptual model for stakeholder engagement in CER	A conceptual model for involving stakeholders in the CER process	Literature search, followed by practical experience with an existing stakeholder engagement process/project, and review and revision by an expert panel (including patients)	Yes - patients engaged with the Center for CANCERGEN project
Evans/ [26] / UK/ 2019	SUCCESS model	Not stated	Initially developed through a workshop, then reviewed and refined through meetings and email discussions.	Yes - people diagnosed with, or caring for someone with, a chronic or long-term condition
Evans/ [23] / UK and Wales/ 2020	PRIME Centre Wales	To enable and promote successful patient engagement across research activities within their research centre.	Informed by the research literature and the development of an engagement plan with the help of patients.	Yes – people with experience of using health services and caring for family members as well as people diagnosed with, or caring for someone

Characteristics of the …

Article	Model/framework			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
				with, a chronic or long-term condition
Frank/ [27] / USA/ 2015	Conceptual model for PCOR	To provide the basis for subsequent evaluative frameworks, to guide evaluation of PCOR, and to serve as a foundation for measurement models, to allow testing of hypothesized relationships between elements in the model.	Literature search and worked with a patient advisory panel	Yes - members of the PCORI Patient Engagement Advisory Panel
Gessell/ [29] / USA/ 2017	Not specified	To maximize stakeholder engagement within the context of a pragmatic trial	"Expanded" engagement standards that were previously proposed to PCORI standards	Not specified
Gibson/ [30] / UK/ 2017	Not specified	To map and evaluate patient engagement interactions in health services research	Adapted the researchers' previously developed model through workshops held with various patient groups	Yes - the language used within the adapted model passed through iterative changes involving both the researchers and workshop participants
Goeman/ [33] / Australia/ 2019	Model for successful inclusion of consumers and community	The successful inclusion of consumers in research	Co-design process involving expert working and reference groups	Yes - People living with dementia and their care-partners (“consumers”)

Characteristics of the …

Article	Model/framework			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
	representatives in research			
Holmes/ [22]/ Canada/ 2018	BC SUPPORT Unit Framework	To conceptualize and organize patient engagement activities	Not specified	Not specified
Pearson/ [31] / UK/ 2013	A provisional model for patient engagement in healthcare OR	A model for patient engagement in healthcare OR	Not specified	Not specified
Ray/ [34] / USA/ 2017	Conceptual model for understanding impact of stakeholder engagement and differentiating stakeholder-engaged research from research on stakeholder engagement	A framework for planning, evaluating, and reporting both stakeholder-engaged research and research on stakeholder engagement	Initial framework developed based on synthesis of existing literature; refined through use in a patient engagement research project which examined families' experiences of access to pediatric subspecialty care	No
Ruco/ [35] / Canada/ 2016	Sunnybrook Health Sciences Centre patient engagement in PBRI framework	To guide and capture the range and scope of patient engagement in PBRI at Sunnybrook Health Sciences Centre, Toronto	Developed based on CIHR's Supporting POR Visual Value Model for Patient Engagement and in alignment with Sunnybrook's PBRI Strategic plan	Not specified
Shippee/ [32] /	Framework for PSUE	To develop a standardized, evidence-based	Systematic review/environmental scan/manual	Yes - patient advisory group consisting of

Characteristics of the …

Article	Model/framework			
First author/ reference/ country/ year	**Name**	**Purpose**	**Methods of development**	**Were patient co-researchers involved in its development (yes/no/not specified – population)**
USA/ 2013		framework for understanding, reporting, and assessing PSUE in biomedical and health services research	literature search that was synthesized into a two-part framework and supplemented with input from a patient advisory group	community member PSUEs
Jose/ Unpublished/ 2020	CONNECT framework	To engage members of the autistic community in research	Developed as part of work on the CONNECT project, including 2 feedback questionnaires designed for the study	Yes – members of the autistic community

CANCERGEN = comparative effectiveness research in cancer genomics; CER = comparative effectiveness research; CIHR = Canadian Institutes of Research; OR = operations research; PBRI = practice-based research and innovation; PCORI = patient-centered outcomes research institute; POR = patient-oriented research; PSUE = patient and service user engagement; SUCCESS = service users with chronic conditions encouraging sensible solutions.

GRIPP2-short form checklist

Section and topic	Item	Line numbers
1: Aim Report the aim of the study	Our primary objective was to count and describe the elements that overlap (i.e., present in 2 or more models or frameworks) and diverge between identified models and frameworks.	120-122
2: Methods Provide a clear description of the methods used for patient and public involvement (PPI) in the study	We engaged two patient co-researchers (R.S. and S.H.), at the level of involve (24), in the design and conduct of this study… The primary methods of engagement were 1-on-1 and small group meetings, as guided by a terms of reference co-developed at the outset of the research partnership.	207-208, 213-214
3: Results Outcomes—Report the results of PPI in the study, including both positive and negative outcomes	They were involved in development and publication of the scoping review protocol (19), provided feedback on the analyses (including element groupings), provided their perspectives on the interpretation of the study findings, and co-authored this manuscript.	208-210
4: Discussion Outcomes—Comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	In helping shape these stages of the research cycle and related outputs, a notable impact of engagement also included identifying gaps in the current literature relevant to patient partners.	210-212
5: Reflections Critical perspective—Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	Further, the engagement of patient co-researchers in our scoping review at the level of involve limited their opportunities to formally provide insights and expertise to pre-determined study milestones. Engagement at the level of collaborate or empower may have resulted in other insights through more direct contact with the study.	388-392

Adapted from: *BMJ 2017;358:j3453*

Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	1
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	4-7
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	7
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	7-8
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	8
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	9
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	Appendix 1
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	9
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	9-10
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	Appendix 2
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	N/A

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	10
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	12, Appendix 3
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	12-13, Appendix 4
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	N/A
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	13-18
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	13-14
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	18-21
Limitations	20	Discuss the limitations of the scoping review process.	19
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	21-22
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	22-23

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.

* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.

† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).

‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.

§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy document).

From: Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMAScR): Checklist and Explanation. Ann Intern Med. 2018;169:467–473. doi: 10.7326/M18-0850.