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

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

2
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2
3 **25 Abstract**

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5 **26 Objective:** To count and describe the elements that overlap (i.e., present in 2 or more) and
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7
8 **27** diverge between models and frameworks of patient engagement in health services research. Our
9
10 **28** specific research question was, “What are the elements that underlie models and frameworks of
11
12 **29** patient engagement in health services research?”

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15 **30 Design:** Scoping review.

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18 **31 Eligibility criteria:** We included published and unpublished (i.e., grey) literature that presented
19
20 **32** (a) models or frameworks (b) of patient engagement (c) in health services research. We excluded
21
22 **33** articles unavailable as full-text or not written in English.

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24
25 **34 Data sources:** Using a search strategy co-developed with an academic librarian, published
26
27 **35** literature was identified by searching six electronic databases. Searches for unpublished literature
28
29 **36** included electronic databases and websites. These searches were supplemented by snowball
30
31 **37** sampling.

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35 **38 Data extraction and synthesis:** Two independent reviewers extracted data from included
36
37 **39** articles using an a-priori developed standardized form. Data were synthesized using both
38
39 **40** quantitative (i.e., counts) and qualitative (i.e., mapping) analyses.

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43 **41 Results:** We identified a total of 8069 articles and ultimately included 14 models and
44
45 **42** frameworks in the review. These models and frameworks were comprised of 18 overlapping and
46
47 **43** 57 diverging elements, that were organized into six conceptual categories (i.e., principles,
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49 **44** foundational components, contexts, actions, levels, and outcomes) and spanned intrapersonal,
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51 **45** interpersonal, process, and environment domains.
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3 46 **Conclusions:** There is little overlap between the elements that comprise existing models and
4
5 47 frameworks of patient engagement in health services research. Those seeking to apply these
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7 48 models and frameworks should consider the “fit” of each element, by conceptual category and
8
9 49 domain, within the context of their study.
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13 50 **Trial registration:** None.
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16 51
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19 52 **Keywords**
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22 53 Patient and public involvement, stakeholder engagement, patient involvement, patient-oriented
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24 54 research, patient engagement
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3 **55 Article Summary**
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5 **56 Strengths and limitations of this study**
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- 8 **57** • The study's methodological strengths included its published protocol and analysis
9
- 10 **58** approach, which supported mapping model and framework elements to generate a
11
- 12 **59** toolbox of options for researchers to use in their own patient-engaged research.
13
- 14
- 15 **60** • Another strength included the involvement of an interdisciplinary research team
16
- 17 **61** comprised of content experts, including patient co-researchers.
18
- 19 **62** • A weakness of this study concerns the grey boundaries between health services research
20
- 21 **63** and other forms of health research.
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- 23
- 24 **64** • Greater engagement of patient co-researchers would have likely resulted in other study
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- 26 **65** insights through more direct contact with the study.
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66 Introduction

67 Patient engagement in research involves the formation of meaningful and active collaborations
68 between academic researchers and patients (i.e., an overarching term that refers to individuals
69 with personal experience of a health issue and informal caregivers) in research governance,
70 priority setting, conduct, and knowledge translation¹. Globally, it is also commonly referred to
71 as patient and public involvement, patient involvement, and stakeholder engagement in research.
72 This research approach necessitates a shift from the patient's traditional role as a study
73 participant to that of a research collaborator or partner (i.e., patient co-researcher). This shift in
74 roles and power dynamics reflects the approach's roots in participatory research² and is founded
75 on the premises that those affected by a problem should be actively involved in the generation of
76 solutions to it³, and individuals' critical reflections on first-hand experiences are essential to
77 effecting individual and social change⁴. A growing body of evidence supports the benefits of
78 patient engagement in research, including improved enrollment and decreased attrition rates⁵,
79 increased relevance of research and accessibility of study materials to study participants⁶,
80 improved trial design⁷, and increased meaningfulness and understandability of disseminated
81 findings^{5,6}. However, despite its underlying rationale and documented benefits, academic
82 researchers report hesitance in adopting this research approach^{8,9}.

83
84 Some of the reported challenges of patient engagement in research include uncertainty about the
85 process (i.e., "how-to" engage patients in research) and the need for a culture shift that supports
86 these collaborations^{8,9}. As there is no standard process for engaging patients in research, this
87 first challenge is not surprising^{5,6,10}. That is, how patient engagement is operationalized may
88 vary according to the characteristics of a given project (e.g., design, scope, time, and financial

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3 89 resources) and patient and academic co-researchers (e.g., personal and professional backgrounds,
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5 90 interests, skills). Further, the underlying cultural shift needed to support patient engagement in
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7 91 research necessitates the redistribution of power and restructuring of traditional research
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10 92 paradigms to support shared planning and decision-making throughout a study ¹⁰. This requires a
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12 93 mutual understanding and vision for what these research collaborations entail. An exploration of
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14 94 the models and frameworks that underlie patient engagement in research may help clarify the
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16 95 processes and support the culture shift necessary for this approach by shedding light on the
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19 96 universal elements that underlie it.
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24 98 Three previous reviews have broadly synthesized the literature on models, frameworks and/or
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26 99 other systematic approaches to the engagement of patients (and in the case of Jull et al. - other
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28 100 knowledge users) in research ¹¹⁻¹³. Of these, only one described the elements that comprised
29
30 101 identified frameworks and best practice guidelines ¹². Perhaps more importantly, none of these
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33 102 reviews focused on health services research, which entails considerations unique to health
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35 103 research. Specifically, health services research focuses on the impact of social factors, financing
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37 104 systems, organizational structures and processes, health technologies, and personal behaviours
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39 105 on health care access, quality and cost, and population health and well-being ¹⁴. This differs
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42 106 substantially from general health research, where the goal of engagement is often to improve
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44 107 clinical outcomes, the effectiveness of a particular intervention, or uptake of research among
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46 108 patients with a specific condition. Engaging patients in health services research may require
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49 109 added considerations related to partnering with a more diverse group of patients with lived
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51 110 experience of different conditions, groups, interventions, and/or interactions with the health care
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54 111 system. Therefore, we set out to contribute to the existing literature on patient engagement in
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3 112 research by conducting a knowledge synthesis of models and frameworks of patient engagement
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5 113 in health services research. Specifically, we undertook a scoping review, which is “a type of
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7 114 knowledge synthesis that addresses an exploratory research question aimed at mapping key
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9 115 concepts, types of evidence, and gaps in research related to a defined area or field by
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11 116 systematically searching, selecting, and synthesizing existing knowledge ¹⁵.”
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16 17 118 **Objectives**

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19 119 The research question driving our review was, “What are the elements that underlie models and
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21 120 frameworks of patient engagement in health services research?” Our primary objective was to
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23 121 count and describe the elements that overlap (i.e., present in 2 or more models or frameworks)
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25 122 and diverge between identified models and frameworks. This objective intends to support a
26
27 123 clearer understanding of similarities in thinking about patient engagement rather than to judge
28
29 124 the relevance of elements or prescribe a “one size fits all” approach. Thus, rather than
30
31 125 synthesizing the identified elements, we chose to map them according to the categories
32
33 126 developed by the original authors and the themes that arose among them, with the vision of
34
35 127 presenting a “toolbox” of potential approaches that researchers may choose from for their given
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37 128 research endeavor. Finally, the exploratory nature of our research question and our desire to
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39 129 identify and map key concepts that underlie patient engagement in research led us to adopt a
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41 130 scoping review methodology ¹⁶.
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48 49 132 **Methods**

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51 133 Our scoping review’s design and conduct followed the methodological framework proposed by
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53 134 Arksey and O’Malley ¹⁷ and enhanced by Levac et al. ¹⁸. Its underlying protocol, including
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3 135 definitions of underlying key concepts, is published elsewhere¹⁹. Reporting was guided by the
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5 136 Preferred Reporting Items for Systematic Reviews and Meta-Analysis: Extension for Scoping
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8 137 Review (PRISMA-ScR)²⁰ and the revised Guidance for Reporting Involvement of Patients and
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10 138 the Public (GRIPP2) short form²¹ checklists. Research ethics approval was not required as this
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12 139 study did not involve participants.
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17 141 **Eligibility criteria:** We included published and unpublished (i.e., grey) literature that presented
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19 142 (i) models or frameworks (ii) of patient engagement (iii) in health services research. Both
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21 143 original and adapted models and frameworks were eligible as long as they were developed in or
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23 144 for health services research. We excluded articles unavailable as full-text due to the limited
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25 145 descriptive information they provide (e.g., context, description of development, underlying
26
27 146 elements) and not written in English due to feasibility-related considerations.
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33 148 **Information sources:** The lead author (A.M.C.) searched six electronic databases (CINAHL,
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35 149 Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice
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37 150 Database, MEDLine, PsycINFO, and Scopus) and Google Scholar for published literature.
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39 151 Electronic databases (ProQuest Dissertations & Theses and Conference Proceedings Citation
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41 152 Index), Google, and the websites of key agencies (Canadian Institutes of Health Research,
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43 153 INVOLVE, Patient-Centered Outcomes Research Institute) were searched for unpublished
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45 154 literature. These searches were supplemented by snowball sampling, which entailed backwards
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47 155 and forwards reference searches of included articles and contacting experts in patient
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49 156 engagement in research for recommendations about any potentially relevant models or
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53 157 frameworks.
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3 158 **Searches:** The search strategy was co-developed by an academic librarian (L.D.) and the lead
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5 159 author (A.M.C.) and finalized through collaboration with the rest of the research team. The
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7 160 search strategy used a combination of search terms related to our underlying concepts (i.e.,
8
9 161 “models,” “frameworks,” “patient engagement,” and “health services research”), was adapted to
10
11 162 the syntax used by each database and website, and used Boolean terms. The search strategy for
12
13 163 MEDLine is found in **Appendix 1**. There were no restrictions on publication dates. Searches of
14
15 164 the published and unpublished literature were conducted on July 6, 2021 and July 7, 2021,
16
17 165 respectively, while backward and forwards searches ended on January 13, 2022.
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24 167 **Selection:** Search results for the six electronic databases and Google Scholar were imported into
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26 168 a reference management software (Endnote), and duplicate references were removed. Only the
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28 169 first 10 pages of Google Scholar results (n = 100 citations) were imported as advised by an
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30 170 academic librarian (L.D.). One reviewer (A.M.C.) conducted the level 1 (title) screening. The
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32 171 remaining relevant references were then imported into an online systematic review production
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34 172 and management software (Covidence; Veritas Health Innovation, Melbourne Australia), where
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36 173 two reviewers (A.M.C. and T.H.) independently conducted the level 2 (abstract) and level 3
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38 174 (full-text) screening. Potentially relevant literature identified through websites and snowball
39
40 175 sampling was screened for inclusion by both reviewers (A.M.C. and T.H.).
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47 177 **Data charting process:** A standardized data charting (i.e., extraction) form was developed a
48
49 178 priori in Microsoft Word by the study team (**Appendix 2**). Items were chosen that described the
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51 179 eligible models and frameworks, as well as provided context, including how and by who the
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53 180 models were developed. Two reviewers (A.M.C. and T.H.) independently extracted information
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3 181 from the final set of included articles using the form. They met with the senior author (A.S) to
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5 182 compare their data after extracting from an initial set of five and ten articles and upon completion
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7 183 of data extraction. Any inconsistencies were resolved through discussion and ultimately referring
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10 184 back to the original article. Further, in two instances^{22 23}, the lead author (A.M.C.) contacted the
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12 185 corresponding authors of included frameworks to obtain clarifying information. Consistent with
13
14 186 methodological guidelines for scoping reviews, we did not appraise the methodological quality
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16
17 187 of included articles^{15 16}.

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19 188
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21 189 **Synthesis of results:** Data were compiled into a single Microsoft Excel (2019) spreadsheet for
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23 190 validation and synthesis, which included both quantitative (i.e., counts) and qualitative (i.e.,
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25 191 mapping) analyses. In conducting the analyses, the lead author (A.M.C) first immersed herself in
26
27 192 the included models and frameworks by thoroughly (and repeatedly) reading the associated
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29 193 literature and reviewing graphical representations (when available) along with element
30
31 194 definitions. With increased familiarity, the lead author was able to combine and reframe (where
32
33 195 appropriate) similar elements found in the original publications. The revised elements were used
34
35 196 to obtain counts of overlapping and diverging elements. During this process, it became evident
36
37 197 that the elements were organized into similar over-arching conceptual categories by the original
38
39 198 authors, and that they could also be located within multiple domains (**Table 1**). As such, in our
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41 199 analysis, we mapped each element according to conceptual category and domain. The validity
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43 200 (i.e., content and face) of the final set of elements was established through discussion with this
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45 201 study's authors. Visual representations of the data (i.e., concept maps) were created using Mind
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47 202 Manager 2020 software, version 20.1 (Corel Corp., Austin, T.X.). To support applicability, some
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49 203 elements within the concept maps contain clarifying examples in brackets.

204 **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

205

206 **Patient involvement**

207 We engaged two patient co-researchers (R.S. and S.H.), at the level of involve²⁴, in the design
 208 and conduct of this study. They were involved in development and publication of the scoping

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3 209 review protocol ¹⁹, provided feedback on the analyses (including element groupings), provided
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5 210 their perspectives on the interpretation of the study findings, and co-authored this manuscript. In
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8 211 helping shape these stages of the research cycle and related outputs, a notable impact of
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10 212 engagement also included identifying gaps in the current literature relevant to patient partners.
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12 213 The primary methods of engagement were 1-on-1 and small group meetings, as guided by a
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14 214 terms of reference co-developed at the outset of the research partnership. The patient co-
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16 215 researchers will continue to be involved in further knowledge translation activities, like
17
18 216 synthesizing the findings for our research group's website
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21 217 (www.patientengagementinresearch.ca), co-presenting about the engagement process and study
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23 218 findings, and identifying other appropriate methods of dissemination.
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28 220 **Results**

31 221 *Flow of models and frameworks into the study*

33 222 **Appendix 3** displays the flow of eligible articles into the review (PRISMA flow chart). Of the
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35 223 10,840 initially identified citations, 2771 duplicates were excluded. After applying inclusion
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37 224 criteria, 7150 articles were excluded at the title screen, 712 at the abstract screen, and 193 at the
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39 225 full-text screen, leaving 13 models and frameworks. One unpublished framework was then
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41 226 identified through snowball sampling, resulting in a total of 14 models and frameworks included
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43 227 in the review.
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49 229 *Characteristics of included articles and models/frameworks*

51 230 Select descriptive characteristics of the included models and frameworks and the articles they
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53 231 were published in (where applicable) are presented in **Appendix 4**. Included articles were
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3 232 published between 2012 - 2020 (n = 10 were published in or after 2015) and based in the United
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5 233 States (n = 5), United Kingdom (n = 4), Canada (n = 2), Australia (n = 1) and United Kingdom
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7 234 and Wales (n = 1). The included unpublished framework was developed in Canada as part of the
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9 235 CONNECT project (Caroline Jose, Patricia George-Zwicker, Louise Tardif, Aaron Bouma,
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11 236 Darlene Pugsley, Luke Pugsley, Mathieu Bélanger, Jeffrey Gaudet, Marc Robichaud,
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13 237 CONNECT framework). Models and frameworks were developed in a variety of contexts
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15 238 including organizations (i.e., Patient-Centered Outcomes Research Institute, British Columbia
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17 239 Support Unit, Sunnybrook Hospital, PRIME Centre Wales), medical specialties (i.e., pediatric
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19 240 subspecialty care, palliative care), research disciplines (i.e., healthcare operations research),
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21 241 diseases (i.e., chronic or long-term conditions; cancer; dementia; stroke), and other health
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23 242 conditions (e.g., autistic adults, persons with lived experience of long-term physical and/or
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25 243 mental health illness, parents of children with disabilities). They were developed for general use
26
27 244 as well in specific health services research contexts like healthcare operational research, practice-
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29 245 based research and innovation, pragmatic trials, patient-centered outcomes research, and
30
31 246 comparative effectiveness research. None targeted a specific component of the research process,
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33 247 and patients were involved in the development of slightly over half (n = 8) of the included
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35 248 models/frameworks. Eight of the models/frameworks took into account the public participation
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37 249 spectrum²⁴ either explicitly or by including considerations related to control of decision-making
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39 250 and/or directionality of information exchange.
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49 252 *Overview of the elements of included models and frameworks*

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51 253 A total of 112 elements of patient engagement were identified across the 14 included models and
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53 254 frameworks. Combining and reframing similar elements reduced the total number to 75. Among
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3 255 these, 18 elements overlapped across the included models and frameworks and 57 diverged (i.e.,
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5 256 were unique to individual models and frameworks). We present the elements by conceptual
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7 257 category and identify the domains they are situated in below. In considering these results, it
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9
10 258 should be noted that we placed select elements from the models by Deverka et al. ²⁵ (i.e., inputs,
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12 259 methods, outputs) and Evans et al. ²⁶ (i.e., opportunities for engagement, research environment
13
14 260 that actively supports engagement and its underlying principles, resources to support
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17 261 engagement, and system to enhance the breadth of patient and carer experience brought to
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19 262 research activities) in the “actions” conceptual category despite that not being what the original
20
21 263 authors labeled them. This is due to the fact that although these elements were uncategorized by
22
23 264 Deverka et al. ²⁵, they closely aligned with the actions category, and Evans et al.’s definition of
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25 265 the conceptual category that encompassed these elements closely aligned with other authors’
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27 266 conceptualization of “actions.”
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32 33 268 **Principles**

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35 269 Principles represent the values that orient and rule the conduct of a group. They form the ethical
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37 270 backdrop of engagement ²⁷ and underpin effective collaborative involvement in research ²⁶. For
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39 271 example, communication, which included mutual communication and feedback, was identified
40
41 272 as a core principle of patient engagement by Evans et al. ²⁶. Figure 1 displays the 13 elements in
42
43 273 this conceptual category, as reported by two articles ^{26 27}. These elements were situated in
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45 274 interpersonal (n = 7), process (n = 5), and environment (n = 1) domains.
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Figure 1 about here

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3 2784
5 279 Figure 1. Concept map – principles of patient engagement in health services research6
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8 280 Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements9
10 281 that overlap *across* conceptual categories; **Blue**: elements that reside in the interpersonal domain;11
12 282 **Orange**: elements that reside in the process domain; **Purple**: elements that reside in the13
14 283 environment domain.15
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17 28418
19 285 **Foundational components**20
21 286 Foundational components represent the core elements that comprise patient engagement in health22
23 287 services research (Figure 2). According to the eight models and frameworks²⁷⁻³³ represented24
25 288 here, the 26 underlying elements were primarily situated in the process domain (n = 1426
27 289 elements), with the remainder situated in intrapersonal (n = 7), interpersonal (n = 3), and28
29 290 environmental (n = 2) domains.30
31
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33 29134
35 292 ****Figure 2 about here****36
37 293 Figure 2. Concept map – foundational components of patient engagement in health services38
39 294 research40
41
42 295 Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements43
44 296 that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal45
46 297 domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in47
48 298 the process domain; **Purple**: elements that reside in the environment domain.49
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3 **301 Contexts**
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5 **302** This conceptual category identifies elements that relate to resources or decisions that are external
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8 **303** to but inform the engagement process³⁴. Figure 3 presents the four elements in this category, as
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10 **304** contributed by one article³⁴. Three of the underlying elements resided in the process, and one in
11
12 **305** the intrapersonal domains.
13

14 **306**

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17 **307** ****Figure 3 about here****
18

19 **308** Figure 3. Concept map – contexts of patient engagement in health services research

20
21 **309** Figure legend: **Oval**: elements that overlap *across* conceptual categories; **Green**: elements that
22
23
24 **310** reside in the intrapersonal domain; **Orange**: elements that reside in the process domain.
25

26 **311**

27
28 **312 Actions**
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30
31 **313** The elements within this category pertain to the activities (e.g., behaviors, phases, advisory
32
33 **314** bodies) involved in the actual engagement of patients in health services research. Figure 4
34
35 **315** presents the elements that comprised this category, as reported by six articles^{22 25-27 32 34}. These
36
37 **316** elements were primarily (n = 18) situated in the process domain, with one element in each of
38
39 **317** intrapersonal and environment domains, and two elements in the interpersonal domain. As
40
41 **318** displayed in the top half of elements in the process domain (Figure 4), some of the elements
42
43
44 **319** located here were conceptualized in terms of phases of research (i.e., preparatory, execution,
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46
47 **320** translational phases and inputs, methods, outputs).
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51 **322** ****Figure 4 about here****
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54 **323**
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3 324 Figure 4. Concept map – actions of patient engagement in health services research

4
5 325 Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements
6
7 326 that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal
8
9 327 domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in
10
11 328 the process domain; **Purple**: elements that reside in the environment domain.
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15 329

16 330 **Outcomes**

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19 331 Figure 5 presents the elements that comprise the outcomes of patient engagement in health
20
21 332 services research, based upon three articles^{25 27 34} and the unpublished CONNECT framework.
22
23 333 As reflected in the figure, all but the CONNECT framework further organized the elements by
24
25 334 time frame (i.e., immediate/near-term, intermediate, and long-term), with three elements (i.e.,
26
27 335 health decision-making, research culture, and research outcomes) belonging to two time frames.
28
29 336 Overall, this category's elements were primarily situated in the process (n = 11) and health
30
31 337 systems and outcomes (n = 8) domains. A further three elements were situated in the
32
33 338 intrapersonal, one in the interpersonal, and two in the environment domains.
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40 340 **Figure 5 about here**

41
42 341 Figure 5. Concept map – outcomes of patient engagement in health services research

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44 342 Figure legend: **Coloured**: elements that overlap *within* conceptual categories; **Oval**: elements
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46 343 that overlap *across* conceptual categories; **Green**: elements that reside in the intrapersonal
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48 344 domain; **Blue**: elements that reside in the interpersonal domain; **Orange**: elements that reside in
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50 345 the process domain; **Purple**: elements that reside in the environment domain; **Yellow**: elements
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52 346 that reside in the health system and outcomes domain.
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347 **Organizational levels**

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

352 ****Figure 6 about here****

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

357 **Discussion**

358 *Statement of principal findings*

359 Our scoping review identified 14 models and frameworks of patient engagement in health
360 services research, which were comprised of 18 overlapping and 57 diverging elements (n_{total}
361 elements = 75). This work represents a novel contribution as, to our knowledge, it is the first to
362 synthesize the literature on models and frameworks of patient engagement in health services
363 research. Our approach to data synthesis is also unique in that we attempt to maximize the
364 intuitiveness and applicability of our findings by presenting elements by overarching conceptual
365 categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and
366 corresponding domains (i.e., intrapersonal, interpersonal, process, and environment). We
367 anticipate this approach will facilitate the ready application of our findings to readers' own
368 research programs by serving as a "toolbox" of elements to consider according to the multi-level
369 facets of a research team and study. That is, by considering the applicability of elements within

1
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3 370 each conceptual category and domain (as well as other elements that may come to mind based on
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5 371 the groupings' definitions), our findings can serve as a map that allows researchers to pick and
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8 372 choose elements based on their study, resources, research partners, and context.
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11
12 374 *Strengths and weaknesses of the study*

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14 375 Strengths of this study include its scoping review design, which enabled us to gain a broad
15
16 376 perspective of the literature on models and frameworks of patient engagement in health services
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18 377 research. Other strengths include (i) a published protocol, (ii) the use of an established
19
20 378 methodological framework to guide its design and conduct, (iii) the involvement of an
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22 379 interdisciplinary research team that included patient co-researchers with lived experience of
23
24 380 engaging in health services research and other researchers with content expertise in health
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26 381 services research and participatory approaches (including patient engagement in research), and
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28 382 (iv) and the co-design of the search strategy with an academic librarian (LD) and the rest of the
29
30 383 research team. A weakness of this study concerns the grey boundaries between health services
31
32 384 research and other forms of health research, which may have resulted in the inclusion or
33
34 385 exclusion of models or frameworks that others could argue do/do not belong in this review. We
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36 386 attempted to minimize this possibility through a screening process that utilized two reviewers
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38 387 (with a third to resolve discrepancies), an a priori agreed upon definition of health services
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40 388 research, and reaching out to study authors for clarification when necessary. Further, the
41
42 389 engagement of patient co-researchers in our scoping review at the level of involve limited their
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44 390 opportunities to formally provide insights and expertise to pre-determined study milestones.
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46 391 Engagement at the level of collaborate or empower may have resulted in other insights through
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48 392 more direct contact with the study.
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3 393 *Comparison to other studies*
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5 394 Previous reviews of models, frameworks, and/or other systematic approaches to the engagement
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7 395 of patients and other knowledge users in research had a broader scope ¹¹⁻¹³ (albeit only one
8
9 396 involved a comprehensive search of multiple databases ¹¹) and different levels of analyses.

10 397 Specifically, rather than report underlying elements, Greenhalgh et al. developed a taxonomy for
11
12 398 the classification of identified systematic approaches (i.e., tools, frameworks, benchmarks,
13
14 399 guidelines, and critical appraisal checklists) based on their primary focus and intended purpose
15
16 400 (i.e., power-focused, priority-setting, study-focused, report-focused, partnership-focused) ¹¹.

17 401 Innovatively, they piloted co-design workshops that aimed to improve the aesthetic appeal and
18
19 402 usability of “best in-class” resources identified through the review. Similar to our questioning of
20
21 403 the appropriateness of a “one-size fits all” framework, preliminary findings from their co-design
22
23 404 workshops indicated that although stakeholders were presented with a common set of resources,
24
25 405 they generated widely differing frameworks suited to meet their different needs and purposes.

26 406 The review by Jull et al. reported 15 high-level concepts for knowledge user engagement in
27
28 407 research that they identified through a directed content analysis of underlying elements ¹³.

29 408 Although they organized these concepts across four general research phases (i.e., prepare, plan,
30
31 409 conduct, apply), they concluded that variation in the reported concepts between frameworks
32
33 410 indicated that research teams should consider the concepts as fluid rather than strictly required.

34 411 Lastly, the findings of Harrison et al.’s narrative review of frameworks and guidelines
35
36 412 culminated in the proposal of an overarching framework that (similar in principle to our scoping
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38 413 review) conceptualized three distinct but inter-related elements of the patient engagement in
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40 414 research process - engagement foundational principles (i.e., domains that original study authors
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42 415 “... considered foundational to patient engagement in research;” n = 15 elements), engagement
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3 416 best practices (i.e., best practice activities to support engagement; n = 25 elements), and research
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5 417 phases where engagement should occur (n = 3 elements) ¹².
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10 419 *The meaning of the study*

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12 420 Our findings indicate that the conceptualization of patient engagement in health services research
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14 421 varies between existing models and frameworks. Although models and frameworks posit to
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16 422 break down a concept into its base components (which implies the existence of convergence
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18 423 among underlying elements), this finding is not surprising as patient engagement is an *approach*
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20 424 to research rather than a *method*. Contributing to the heterogeneity among identified elements is
21
22 425 the diversity in the populations, contexts, and approaches used to develop the models and
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24 426 frameworks, which also emphasizes the importance of avoiding a “one size fits all” approach to
25
26 427 engagement. Perhaps congruence between models and frameworks actually exists at the level of
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28 428 conceptual categories. These could be taken to represent the essential components of patient
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30 429 engagement in health services research. The elements that underlie them, as identified across the
31
32 430 various models and frameworks, would then serve as considerations for researchers and patient
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34 431 co-researchers when planning and operationalizing patient engagement in health services
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36 432 research. For this to be better developed from the current literature, consensus is needed on the
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38 433 definitions of the underlying categories, followed by some re-shuffling of the elements across the
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40 434 conceptual categories in order to align with the agreed upon definitions.
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49 436 *Unanswered questions and future research.*

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51 437 In sum, our study found that there is little overlap between the elements that comprise existing
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53 438 models and frameworks of patient engagement in health services research. Readers seeking to
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3 439 apply our findings to their own engaged work should consider the “fit” of each element, by
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5 440 conceptual category and domain, within the context of their study. Future research that builds
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7 441 upon our work should consider addressing three major gaps. First, most existing models and
8
9 442 frameworks identified perceived deficits within patient co-researchers, with a lot less
10
11 443 consideration paid to deficits that exist within academic co-researchers. This may be in large part
12
13 444 due to the fact the majority of the models were written from an academic researcher lens (as
14
15 445 evidenced, for example, by the fact that only a third of the included models and frameworks
16
17 446 stated that they involved patients in their development). Second, there is a lack of focus on the
18
19 447 “soft skills” that underlie interpersonal interactions and relationships (e.g., body language,
20
21 448 wording, tone), as well as intrapersonal-level elements of patient engagement in health services
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23 449 research (e.g., attitudes, values, expectations). Third, patient co-researchers are not just
24
25 450 “patients” that can be lumped into a single homogenous category. They are people with different
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27 451 backgrounds, skills, and interests extending beyond their health conditions or needs. Thus, it is
28
29 452 important to incorporate a trauma-informed and intersectional approach that acknowledges and
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31 453 promotes an understanding of human beings as shaped by the interactions of different social
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33 454 locations and experiences³⁶. Relatedly, it is important for patient co-researchers and academic
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35 455 researchers to get to know each other as people, instead of making assumptions (including about
36
37 456 a patient co-researcher’s experience with healthcare services). Engagement is as much about
38
39 457 relational interactions as it is research processes. Careful attention needs to be paid to both for
40
41 458 academic-patient co-researcher relationships to thrive.
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50
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52
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20
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22
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24
25 472 for data collection and analysis. T.H., L.D., C.S., R.S., and S.H. were responsible for all major
26
27 473 areas of concept development and study planning, were consulted on the data analysis and its
28
29 474 interpretation, and provided manuscript edits. In addition, T.H. was responsible for data
30
31 475 collection. C.W. led concept development but passed away prior to the completion of the
32
33 476 manuscript.
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40 478 **Data sharing statement:** Data are available upon reasonable request to the corresponding author
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42 479 (<https://orcid.org/0000-0001-8476-2093>)
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47 481 **Ethics statements**

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51 483 **Ethics approval:** This paper synthesized publicly available articles and therefore did not require
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53 484 ethical approval.
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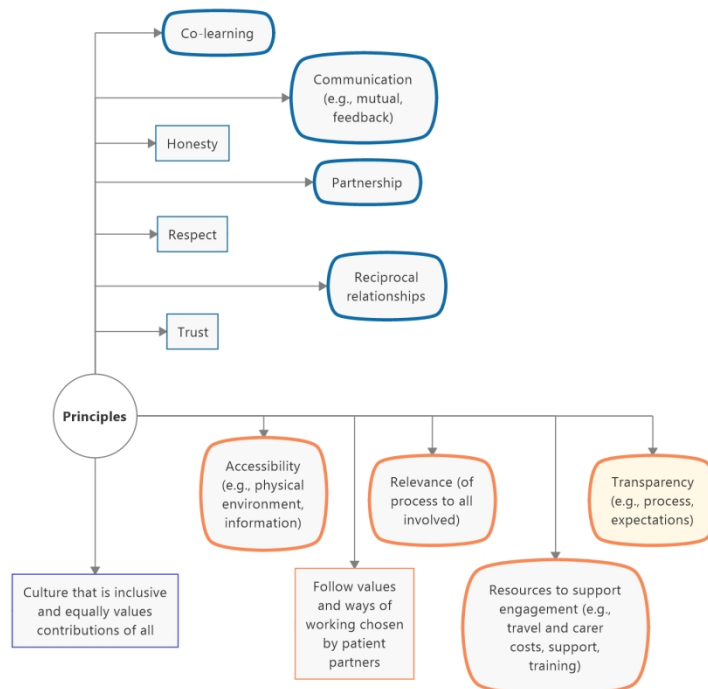


Figure 1. Concept map – principles of patient engagement in health services research
 Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Blue: elements that reside in the interpersonal domain; Orange: elements that reside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

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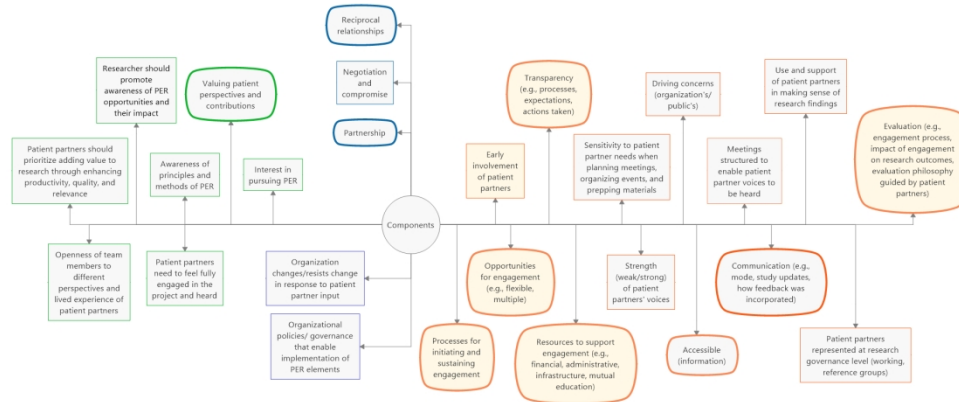


Figure 2. Concept map – foundational components of patient engagement in health services research
 Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Blue: elements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

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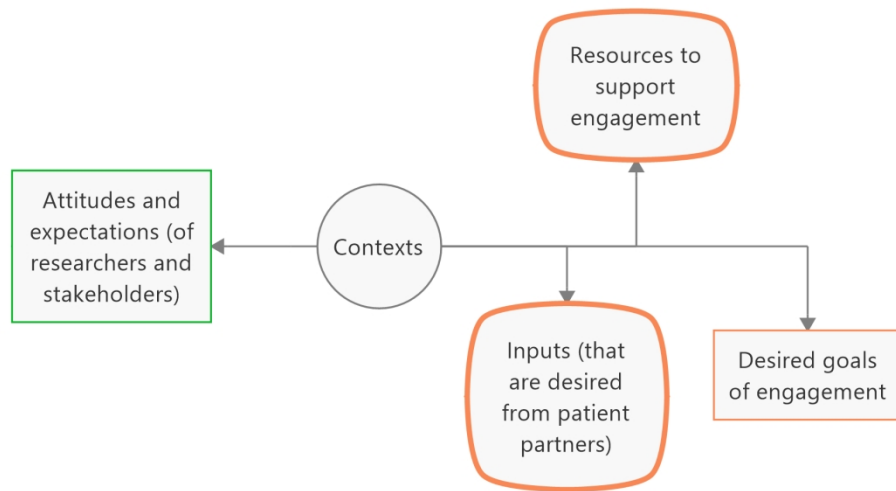


Figure 3. Concept map – contexts of patient engagement in health services research
Figure legend: Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Orange: elements that reside in the process domain.

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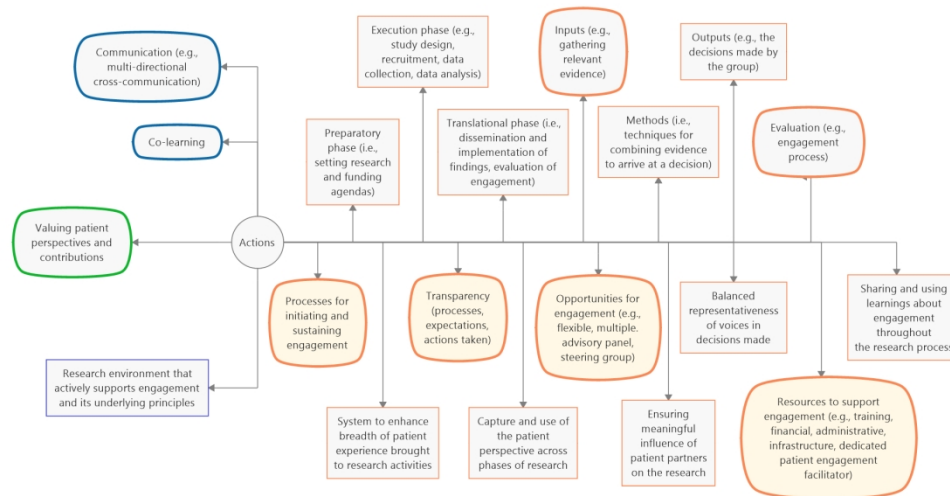
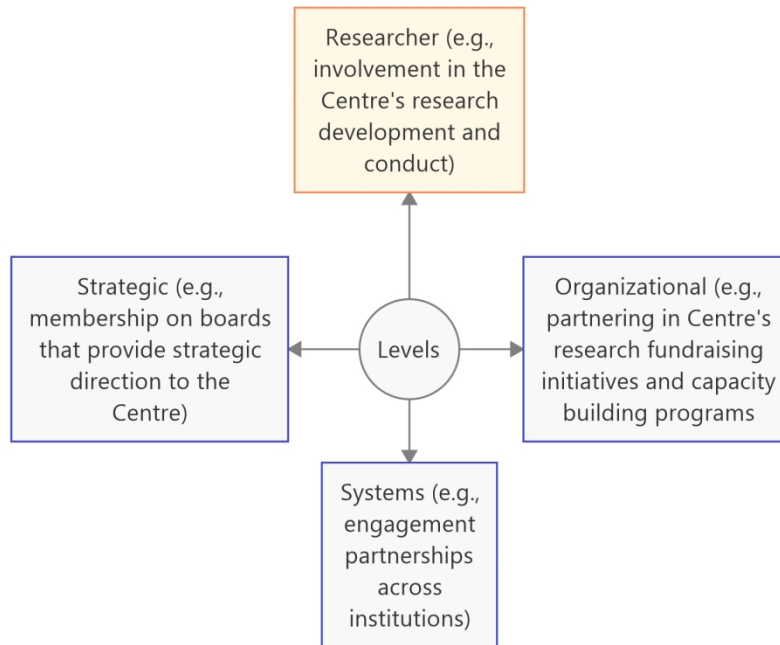


Figure 4. Concept map – actions of patient engagement in health services research
 Figure legend: Coloured: elements that overlap within conceptual categories; Oval: elements that overlap across conceptual categories; Green: elements that reside in the intrapersonal domain; Blue: elements that reside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



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

37 430x322mm (118 x 118 DPI)

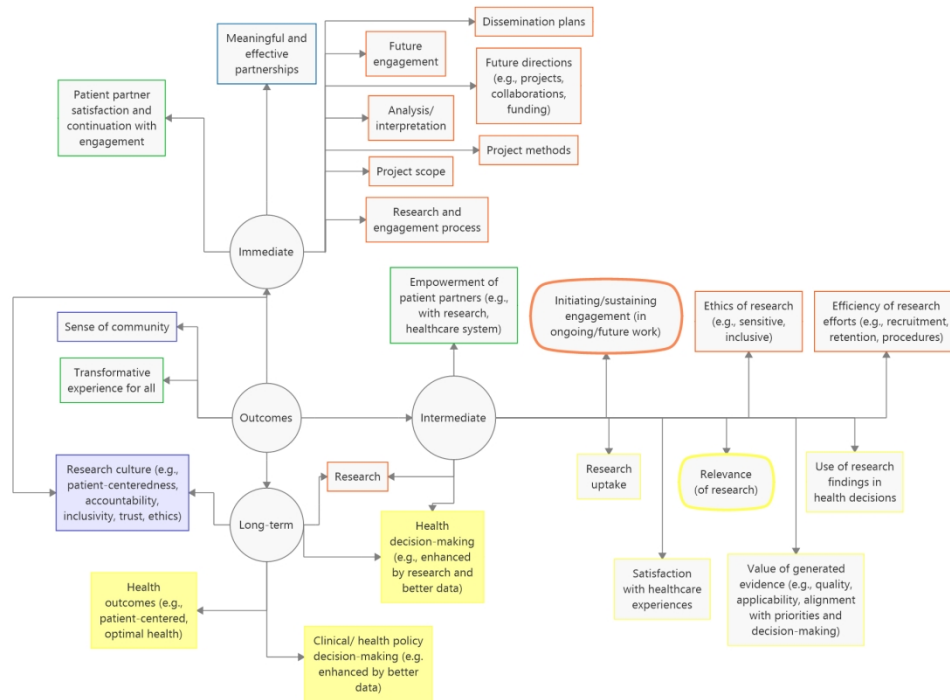


Figure 6. Concept map – organizational levels of patient engagement in health services research
 Figure legend: Coloured: elements that overlap within conceptual categories; Orange: elements that reside in the process domain; Purple: elements that reside in the environment domain.

430x322mm (118 x 118 DPI)

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4 Appendix 1 –Search strategy for MEDLINE (Ovid)
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- 7 1. Patient Participation/
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9 2. models, theoretical/ or patient-specific modeling/
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11 3. (patient* adj3 engag*).mp.
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13 4. (model or models or framework*).mp.
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15 5. research.mp.
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17 6. ("patient oriented research" or "patient and public involvement" or "patient involvement"
18 or "stakeholder engagement").mp.
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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 <i>(dd/mm/yyyy)</i>	
Name of data extractor	

Study authors (characteristics of)

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

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

Publication (characteristics of)

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

	Descriptions as stated in report/paper or note otherwise	Location in text or source (<i>pg & ¶/fig/table/other</i>)
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)

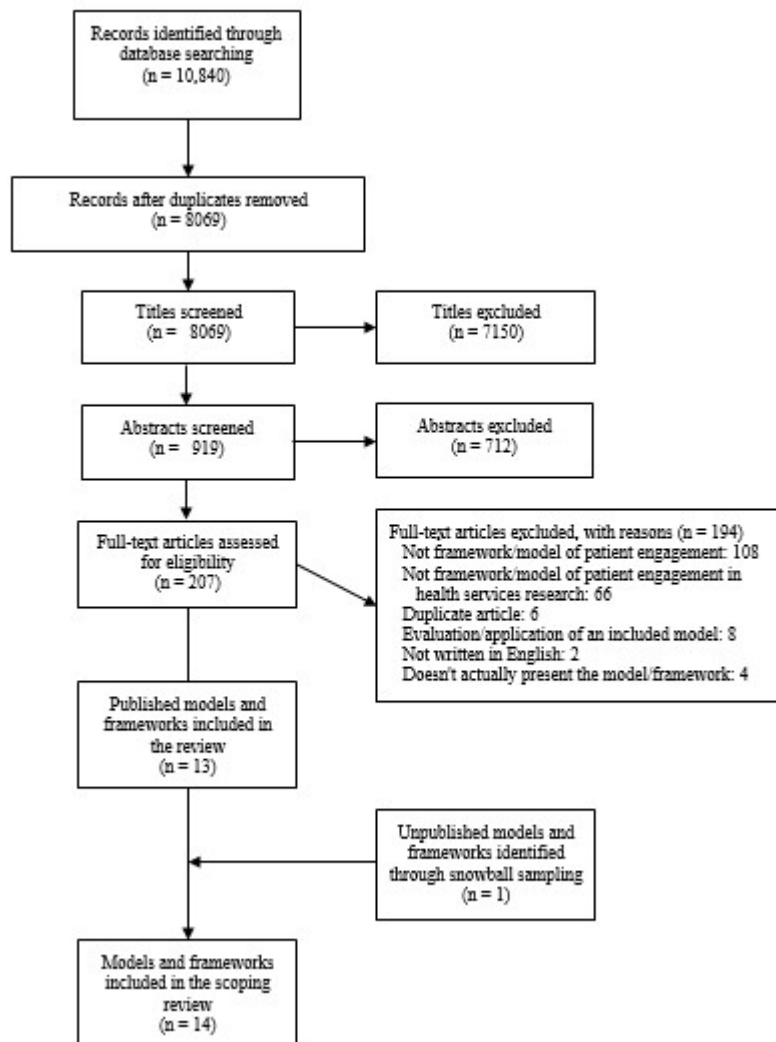
Definitions

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: <ul style="list-style-type: none"> • 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 ...

Model/framework

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)
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	with, a chronic or long- term condition 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 ...

Model/framework

Article				
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 ...

Model/framework

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)
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 (PRISMA-ScR): Checklist and Explanation. *Ann Intern Med.* 2018;169:467–473. doi: [10.7326/M18-0850](https://doi.org/10.7326/M18-0850).



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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

BMJ Open

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

Journal:	<i>BMJ Open</i>
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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3 **1 A scoping review of models and frameworks of patient engagement in health services**
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5 **2 research**
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12 **29**

13
14 **30 Abstract**

15 **31 Objective:** To count and describe the elements that overlap (i.e., present in 2 or more) and
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17 **32** diverge between models and frameworks of patient engagement in health services research. Our
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19 **33** specific research question was, “What are the elements that underlie models and frameworks of
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21 **34** patient engagement in health services research?”
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25 **35 Design:** Scoping review.
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29 **36 Data sources:** On July 6–7, 2021, we searched six electronic databases (CINAHL, Cochrane
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31 **37** Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database,
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33 **38** MEDLINE, PsycINFO, and Scopus) and Google Scholar for published literature, and ProQuest
34
35 **39** Dissertations & Theses, Conference Proceedings Citation Index, Google, and key agencies’
36
37 **40** websites for unpublished (i.e., grey) literature, with no date restrictions. These searches were
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39 **41** supplemented by snowball sampling.
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44 **42 Eligibility criteria:** We included published and unpublished literature that presented (a) models
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46 **43** or frameworks (b) of patient engagement (c) in health services research. We excluded articles
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48 **44** unavailable as full-text or not written in English.
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3 45 **Data extraction and synthesis:** Two independent reviewers extracted data from included
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5 46 articles using an a-priori developed standardized form. Data were synthesized using both
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7 47 quantitative (i.e., counts) and qualitative (i.e., mapping) analyses.
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10 48 **Results:** We identified a total of 8069 articles and ultimately included 14 models and
11
12 49 frameworks in the review. These models and frameworks were comprised of 18 overlapping and
13
14 50 57 diverging elements, that were organized into six conceptual categories (i.e., principles,
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16 51 foundational components, contexts, actions, levels, and outcomes) and spanned intrapersonal,
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18 52 interpersonal, process, environmental, and health systems and outcomes domains.
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21 53 **Conclusions:** There is little overlap between the elements that comprise existing models and
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23 54 frameworks of patient engagement in health services research. Those seeking to apply these
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25 55 models and frameworks should consider the “fit” of each element, by conceptual category and
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27 56 domain, within the context of their study.
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35 58 **Keywords**

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38 59 Patient and public involvement, stakeholder engagement, patient involvement, patient-oriented
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40 60 research, consumer and community involvement
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3 **61 Strengths and limitations of this study**
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- 5 **62** • The study's methodological strengths include use of a published protocol and the
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7 **63** analysis approach, which supported mapping model and framework elements to generate
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9 **64** a toolbox of options for researchers to use in their own patient-engaged research.
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12 **65** • Another strength is the involvement of an interdisciplinary research team comprised of
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14 **66** content experts, including patient co-researchers.
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17 **67** • A weakness of this study concerns the grey boundaries between health services research
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19 **68** and other forms of health research.
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22 **69** • Greater engagement of patient co-researchers would have likely resulted in other study
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24 **70** insights through more direct contact with the study.
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71 Introduction

72 Patient engagement in research involves the formation of meaningful and active collaborations
73 between academic researchers and patients (i.e., an overarching term that refers to individuals
74 with personal experience of a health issue and informal caregivers) in research governance,
75 priority setting, conduct, and knowledge translation¹. Globally, it is also commonly referred to
76 as patient and public involvement, patient involvement, consumer and community involvement,
77 and stakeholder engagement in research. This research approach necessitates a shift from the
78 patient's traditional role as a study participant to that of a research collaborator or partner (i.e.,
79 patient co-researcher). This shift in roles and power dynamics reflects the approach's roots in
80 participatory research² and is founded on the premises that those affected by a problem should
81 be actively involved in the generation of solutions to it³, and individuals' critical reflections on
82 first-hand experiences are essential to effecting individual and social change⁴. A growing body
83 of evidence supports the benefits of patient engagement in research, including improved
84 enrollment and decreased attrition rates⁵, increased relevance of research and accessibility of
85 study materials to study participants⁶, improved trial design⁷, and increased meaningfulness and
86 understandability of disseminated findings^{5 6}. However, despite its underlying rationale and
87 documented benefits, academic researchers report hesitance in adopting this research approach⁸
88 ⁹.

89
90 Some of the reported challenges of patient engagement in research include uncertainty about the
91 process (i.e., "how-to" engage patients in research) and the need for a culture shift that supports
92 these collaborations^{8 9}. As there is no standard process for engaging patients in research, this
93 first challenge is not surprising^{5 6 10}. That is, how patient engagement is operationalized may

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3 94 vary according to the characteristics of a given project (e.g., design, scope, time, and financial
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5 95 resources) and patient and academic co-researchers (e.g., personal and professional backgrounds,
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7 96 interests, skills). Further, the underlying cultural shift needed to support patient engagement in
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10 97 research necessitates the redistribution of power and restructuring of traditional research
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12 98 paradigms to support shared planning and decision-making throughout a study¹⁰. This requires a
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14 99 mutual understanding and vision for what these research collaborations entail. An exploration of
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17 100 the models and frameworks that underlie patient engagement in research may help clarify the
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19 101 processes and support the culture shift necessary for this approach by shedding light on the
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21 102 universal elements that underlie it.
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26 104 Three previous reviews have broadly synthesized the literature on models, frameworks and/or
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28 105 other systematic approaches to the engagement of patients (and in the case of Jull et al. - other
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30 106 knowledge users) in research¹¹⁻¹³. Of these, only one described the elements that comprised
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33 107 identified frameworks and guidelines describing best practices for engaging patients in
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35 108 research¹². Perhaps more importantly, none of these reviews focused on health services research,
36
37 109 which entails considerations unique to health research. Specifically, health services research
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39
40 110 focuses on the impact of social factors, financing systems, organizational structures and
41
42 111 processes, health technologies, and personal behaviours on health care access, quality and cost,
43
44 112 and population health and well-being¹⁴. This differs substantially from general health research,
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46
47 113 where the goal of engagement is often to improve clinical outcomes, the effectiveness of a
48
49 114 particular intervention, or uptake of research among patients with a specific condition. Engaging
50
51 115 patients in health services research may require added considerations related to partnering with a
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53
54 116 more diverse group of patients with lived experience of different conditions, groups,
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3 117 interventions, and/or interactions with the health care system. Therefore, we set out to contribute
4
5 118 to the existing literature on patient engagement in research by conducting a knowledge synthesis
6
7 119 of models and frameworks of patient engagement in health services research. Specifically, we
8
9 120 undertook a scoping review, which is “a type of knowledge synthesis that addresses an
10
11 121 exploratory research question aimed at mapping key concepts, types of evidence, and gaps in
12
13 122 research related to a defined area or field by systematically searching, selecting, and synthesizing
14
15 123 existing knowledge”¹⁵.
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19 124

125 ***Objectives***

126 The research question driving our review was, “What are the elements that underlie models and
127 frameworks of patient engagement in health services research?” Our primary objective was to
128 count and describe the elements that overlap (i.e., present in 2 or more models or frameworks)
129 and diverge between identified models and frameworks. This objective intended to support a
130 clearer understanding of similarities in thinking about patient engagement rather than to judge
131 the relevance of elements or prescribe a “one size fits all” approach. Thus, rather than
132 synthesizing the identified elements, we chose to map them according to the categories
133 developed by the original authors and the themes that arose among them, with the vision of
134 presenting a “toolbox” of potential approaches that researchers may choose from for their given
135 research endeavor. Finally, the exploratory nature of our research question and our desire to
136 identify and map key concepts that underlie patient engagement in research led us to adopt a
137 scoping review methodology¹⁶.

138

139 **Methods**

1
2
3 140 Our scoping review's design and conduct followed the methodological framework proposed by
4
5 141 Arksey and O'Malley¹⁷ and enhanced by Levac et al.¹⁸. The protocol for this scoping review,
6
7 142 including definitions of underlying key concepts, is published elsewhere¹⁹. Reporting was
8
9
10 143 guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analysis: Extension
11
12 144 for Scoping Review (PRISMA-ScR)²⁰ and the revised Guidance for Reporting Involvement of
13
14 145 Patients and the Public (GRIPP2) short form²¹ checklists. Research ethics approval was not
15
16
17 146 required as this study did not involve participants.
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147

21 148 *Eligibility criteria*

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23
24 149 We included published and unpublished (i.e., grey) literature that presented (i) models or
25
26 150 frameworks (ii) of patient engagement (iii) in health services research. Both original and adapted
27
28 151 models and frameworks were eligible as long as they were developed in or for health services
29
30
31 152 research. Since patient engagement also encompasses participation in research governance¹, we
32
33 153 included models and frameworks that conceptualized how patient engagement in research was
34
35 154 embedded across the different levels of health services research centers. We excluded articles
36
37
38 155 unavailable as full-text due to the limited descriptive information they provide (e.g., context,
39
40 156 description of development, underlying elements) and not written in English due to feasibility-
41
42 157 related considerations.
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44

158

47 159 *Information sources*

48
49 160 The lead author (A.M.C.) searched six electronic databases (CINAHL, Cochrane Database of
50
51 161 Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLINE,
52
53
54 162 PsycINFO, and Scopus) and Google Scholar for published literature. Electronic databases
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3 163 (ProQuest Dissertations & Theses and Conference Proceedings Citation Index), Google, and the
4
5 164 websites of key agencies (Canadian Institutes of Health Research, INVOLVE, Patient-Centered
6
7 165 Outcomes Research Institute) were searched for unpublished literature. These searches were
8
9
10 166 supplemented by snowball sampling, which entailed backwards and forwards reference searches
11
12 167 of included articles and contacting experts in patient engagement in research for
13
14 168 recommendations about any potentially relevant models or frameworks.
15
16
17 169

19 170 *Searches*

21 171 The search strategy was co-developed by an academic librarian (L.D.) and the lead author
22
23 172 (A.M.C.) and finalized through discussion with the rest of the research team. The search strategy
24
25 173 used a combination of search terms related to our underlying concepts (i.e., “models,”
26
27 174 “frameworks,” “patient engagement,” and “health services research”), was adapted to the syntax
28
29 175 used by each database, register, and website, and used Boolean terms. The search strategies for
30
31 176 all databases, registers, and websites are found in **Appendix 1**. There were no restrictions on
32
33 177 publication dates. Searches of the published and unpublished literature were conducted on July 6,
34
35 178 2021 and July 7, 2021, respectively, while backward and forwards searches ended on January 13,
36
37 179 2022.
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44 181 *Selection*

46 182 Search results for the six electronic databases and Google Scholar were imported into a reference
47
48 183 management software (Endnote), and duplicate references were removed. Only the first 10 pages
49
50 184 of Google Scholar results (n = 100 citations) were imported as advised by an academic librarian
51
52 185 (L.D.). One reviewer (A.M.C.) conducted the level 1 (title) screening. The remaining relevant
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2
3 186 references were then imported into an online systematic review production and management
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5 187 software (Covidence; Veritas Health Innovation, Melbourne Australia), where two reviewers
6
7 188 (A.M.C. and T.H.) independently conducted the level 2 (abstract) and level 3 (full-text)
8
9
10 189 screening. Potentially relevant literature identified through websites and snowball sampling was
11
12 190 screened for inclusion by both reviewers (A.M.C. and T.H.). A third reviewer (A.S.) helped
13
14 191 resolve discrepancies at the close of level 2 and 3 screening, which predominantly dealt with
15
16
17 192 whether identified literature focused on health services research.
18
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20 193

21 194 *Data charting process*

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23
24 195 A standardized data charting (i.e., extraction) form was developed a priori in Microsoft Word by
25
26 196 the study team (**Appendix 2**). Items were chosen that described the eligible models and
27
28 197 frameworks, as well as provided context, including how and by who the models were developed.
29
30
31 198 Two reviewers (A.M.C. and T.H.) independently extracted information from the final set of
32
33 199 included articles using the form. They met with the senior author (A.S) to compare their data
34
35 200 after extracting from an initial set of five and ten articles and upon completion of data extraction.
36
37
38 201 Any inconsistencies were resolved through discussion and ultimately referring back to the
39
40 202 original article. Further, in two instances ^{22 23}, the lead author (A.M.C.) contacted the
41
42 203 corresponding authors of included frameworks to obtain clarifying information. Consistent with
43
44 204 methodological guidelines for scoping reviews, we did not appraise the methodological quality
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47 205 of included articles ^{15 16}.
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50 206

51 207 *Synthesis of results*

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

227 **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.

1		
2		
3	Foundational components	Core elements that comprise patient engagement.
4		
5	Contexts	Resources or decisions that are external to but inform the
6		engagement process.
7		
8		
9		
10	Actions	Activities (e.g., behaviors, phases, advisory bodies) involved in
11		the actual conduct of the engagement and associated research.
12		
13		
14	Outcomes	Results of engagement and its associated research.
15		
16		
17	Organizational levels	Different organizational levels at which engagement may occur
18		in a research organization.
19		
20		
21	Domain	Elements pertain to...
22		
23	Intrapersonal	Individual-level knowledge, perceptions, attitudes, and beliefs.
24		
25	Interpersonal	Relationships with other people.
26		
27	Process	Carrying out the engagement or broader research.
28		
29	Environmental	Research or organizational policies, cultures, perceptions.
30		
31	Health systems and	Health systems and health outcomes.
32		
33	Outcomes	
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228

229 *Patient and public involvement*

230 We engaged two patient co-researchers (R.S. and S.H.), at the level of ‘involve’²⁴, in the design
 231 and conduct of this study. The patient co-researchers and lead author (A.M.C.) arrived at this
 232 mutual decision at the study’s outset, during the development of a terms of reference that guided
 233 the study’s engagement process. By ‘involve’ we mean that the lead author worked consistently
 234 with patient co-researchers to ensure that their ideas and perspectives were understood and
 235 considered at study milestones. Specifically, patient co-researchers contributed to developing and

1
2
3 236 publishing the scoping review protocol ¹⁹, provided feedback on the analyses (including element
4
5 237 groupings), provided their perspectives on the interpretation of the study findings, and co-
6
7 238 authored this manuscript. In helping shape these stages of the research cycle and related outputs,
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9
10 239 a notable impact of engagement also included identifying gaps in the current literature relevant to
11
12 240 patient partners. The primary methods of engagement were 1-on-1 and small group meetings, as
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14 241 guided by a terms of reference co-developed at the outset of the research partnership. The patient
15
16 242 co-researchers will continue to be involved in further knowledge translation activities, like
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18 243 synthesizing the findings for our research group's website
19
20 244 (www.patientengagementinresearch.ca), co-presenting about the engagement process and study
21
22 245 findings, and identifying other appropriate methods of dissemination.
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27 28 247 **Results**

29 30 248 *Flow of models and frameworks into the study*

31
32 249 **Appendix 3** displays the flow of eligible articles into the review (PRISMA flow chart). Of the
33
34 250 10,840 initially identified citations, 2771 duplicates were excluded. After applying inclusion
35
36 251 criteria, 7150 articles were excluded at the title screen, 712 at the abstract screen, and 194 at the
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38 252 full-text screen, leaving 13 models and frameworks. One unpublished framework was then
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40 253 identified through snowball sampling, resulting in a total of 14 models and frameworks included
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42 254 in the review.
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48 49 256 *Characteristics of included articles and models/frameworks*

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51 257 Select descriptive characteristics of the included models and frameworks and the articles they
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53 258 were published in (where applicable) are presented in **Appendix 4**. Included articles were
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3 259 published between 2012 - 2020 (n = 10 were published in or after 2015) and based in the United
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5 260 States (n = 5), United Kingdom (n = 4), Canada (n = 2), Australia (n = 1) and United Kingdom
6
7 261 and Wales (n = 1). The included unpublished framework was developed in Canada as part of the
8
9 262 CONNECT project (Caroline Jose, Patricia George-Zwicker, Louise Tardif, Aaron Bouma,
10
11 263 Darlene Pugsley, Luke Pugsley, Mathieu Bélanger, Jeffrey Gaudet, Marc Robichaud,
12
13 264 CONNECT framework). Models and frameworks were developed in a variety of contexts
14
15 265 including organizations (i.e., Patient-Centered Outcomes Research Institute, British Columbia
16
17 266 Support Unit, Sunnybrook Hospital, PRIME Centre Wales), medical specialties (i.e., pediatric
18
19 267 subspecialty care, palliative care), research disciplines (i.e., healthcare operations research),
20
21 268 diseases (i.e., chronic or long-term conditions; cancer; dementia; stroke), and other health
22
23 269 conditions (e.g., autistic adults, persons with lived experience of long-term physical and/or
24
25 270 mental health illness, parents of children with disabilities). They were developed for general use
26
27 271 as well in specific health services research contexts like healthcare operational research, practice-
28
29 272 based research and innovation, pragmatic trials, patient-centered outcomes research, and
30
31 273 comparative effectiveness research. None targeted a specific component of the research process,
32
33 274 and patients were involved in the development of slightly over half (n = 8) of the included
34
35 275 models/frameworks. Eight of the models/frameworks took into account the public participation
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37 276 spectrum²⁴ either explicitly or by including considerations related to control of decision-making
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39 277 and/or directionality of information exchange.
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49 279 ***Overview of the elements of included models and frameworks***

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51 280 A total of 112 elements of patient engagement were identified across the 14 included models and
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53 281 frameworks. Combining and reframing similar elements reduced the total number to 75. Among
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1
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3 282 these, 18 elements overlapped across the included models and frameworks and 57 diverged (i.e.,
4
5 283 were unique to individual models and frameworks). We present the elements by conceptual
6
7 284 category and identify the domains they are situated in below. In considering these results, it
8
9
10 285 should be noted that we placed select elements from the models by Deverka et al. ²⁵ (i.e., inputs,
11
12 286 methods, outputs) and Evans et al. ²⁶ (i.e., opportunities for engagement, research environment
13
14 287 that actively supports engagement and its underlying principles, resources to support
15
16 288 engagement, and system to enhance the breadth of patient and carer experience brought to
17
18
19 289 research activities) in the “actions” conceptual category despite that not being what the original
20
21 290 authors labeled them. This is due to the fact that although these elements were uncategorized by
22
23
24 291 Deverka et al. ²⁵, they closely aligned with the actions category, and Evans et al.’s definition of
25
26 292 the conceptual category that encompassed these elements closely aligned with other authors’
27
28 293 conceptualization of “actions.”
29
30
31 294

32 33 295 *Principles*

34
35 296 Principles represent the values that orient and rule the conduct of a group. They form the ethical
36
37 297 backdrop of engagement ²⁷ and underpin effective collaborative involvement in research ²⁶. For
38
39 298 example, communication, which included mutual communication and feedback, was identified
40
41
42 299 as a core principle of patient engagement by Evans et al. ²⁶. Figure 1 displays the 13 elements in
43
44 300 this conceptual category, as reported by two articles ^{26 27}. These elements were situated in
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46
47 301 interpersonal (n = 7), process (n = 5), and environmental (n = 1) domains.
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50 51 303 *Foundational components*

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3 304 Foundational components represent the core elements that comprise patient engagement in health
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5 305 services research (Figure 2). According to the eight models and frameworks²⁷⁻³³ represented
6
7 306 here, the 26 underlying elements were primarily situated in the process domain (n = 14
8
9
10 307 elements), with the remainder situated in intrapersonal (n = 7), interpersonal (n = 3), and
11
12 308 environmental (n = 2) domains.

13
14 309

15 310 *Contexts*

16
17 311 This conceptual category identifies elements that relate to resources or decisions that are external
18
19 312 to but inform the engagement process³⁴. Figure 3 presents the four elements in this category, as
20
21 313 contributed by one article³⁴. Three of the underlying elements resided in the process, and one in
22
23 314 the intrapersonal domains.

24
25 315

26 316 *Actions*

27
28 317 The elements within this category pertain to the activities (e.g., behaviors, phases, advisory
29
30 318 bodies) involved in the actual engagement of patients in health services research. Figure 4
31
32 319 presents the elements that comprised this category, as reported by six articles^{22 25-27 32 34}. These
33
34 320 elements were primarily (n = 18) situated in the process domain, with one element in each of
35
36 321 intrapersonal and environmental domains, and two elements in the interpersonal domain. As
37
38 322 displayed in the top half of elements in the process domain (Figure 4), some of the elements
39
40 323 located here were conceptualized in terms of phases of research (i.e., preparatory, execution,
41
42 324 translational phases and inputs, methods, outputs).

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44 325

45 326 *Outcomes*

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3 327 Figure 5 presents the elements that comprise the outcomes of patient engagement in health
4
5 328 services research, based upon three articles^{25 27 34} and the unpublished CONNECT framework.
6
7 329 As reflected in the figure, all but the CONNECT framework further organized the elements by
8
9
10 330 time frame (i.e., immediate/near-term, intermediate, and long-term), with three elements (i.e.,
11
12 331 health decision-making, research culture, and research outcomes) belonging to two time frames.
13
14 332 Overall, this category's elements were primarily situated in the process (n = 11) and health
15
16 333 systems and outcomes (n = 8) domains. A further three elements were situated in the
17
18 334 intrapersonal, one in the interpersonal, and two in the environmental domains.
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336 *Organizational levels*

26 337 Two articles presented frameworks whose elements captured the organizational levels at which
27
28 338 patient engagement in health services research occurred within research centres^{23 35}. As
29
30 339 displayed in Figure 6, these elements were located in environmental (n = 3) and process (n = 1)
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32 340 domains.
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342 **Discussion**

343 *Principal findings*

42 344 Our scoping review identified 14 models and frameworks of patient engagement in health
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44 345 services research, which were comprised of 18 overlapping and 57 diverging elements (n_{total}
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46 346 elements = 75). This work represents a novel contribution as, to our knowledge, it is the first to
47
48 347 synthesize the literature on models and frameworks of patient engagement in health services
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50 348 research. Our approach to data synthesis is also unique in that we attempt to maximize the
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52 349 intuitiveness and applicability of our findings by presenting elements by overarching conceptual
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3 350 categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and
4
5 351 corresponding domains (i.e., intrapersonal, interpersonal, process, environmental, and health
6
7 352 systems and outcomes). We anticipate this approach will facilitate the ready application of our
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9
10 353 findings to readers' own research programs by serving as a "toolbox" of elements to consider
11
12 354 according to the multi-level facets of a research team and study. To illustrate, research partners
13
14 355 could begin to meet at a study's outset to co-develop terms of reference that guide the relational
15
16 356 and activity related aspects of the study's engagement process. In doing so, they could reflect
17
18 357 upon whether/how elements within the domains of each conceptual category resonate with their
19
20 358 study's engagement process (as influenced by factors such as the study design, available
21
22 359 resources, research partner strengths and interests, etc.) These conversations could be guided by
23
24 360 prompts such as "do we want to embody or incorporate this element within our study (why/why
25
26 361 not)?", "what does the embodiment or incorporation of this element look and feel like to us?",
27
28 362 and "how will we know when we have or have not embodied or incorporated this element within
29
30 363 our study?" In doing so, the "toolbox" of elements found within our review is transformed into a
31
32 364 co-developed "roadmap" to help guide a study's engagement process.
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40 366 ***Strengths and weaknesses of the study***

41
42 367 Strengths of this study include its scoping review design, which enabled us to gain a broad
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44 368 perspective of the literature on models and frameworks of patient engagement in health services
45
46 369 research. Other strengths include (i) a published protocol, (ii) the use of an established
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48 370 methodological framework to guide its design and conduct, (iii) the involvement of an
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50 371 interdisciplinary research team that included patient co-researchers with lived experience of
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52 372 engaging in health services research and other researchers with content expertise in health
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3 373 services research and participatory approaches (including patient engagement in research), and
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5 374 (iv) and the co-design of the search strategy with an academic librarian (LD) and the rest of the
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7
8 375 research team. A weakness of this study concerns the grey boundaries between health services
9
10 376 research and other forms of health research, which may have resulted in the inclusion or
11
12 377 exclusion of models or frameworks that others could argue do/do not belong in this review. We
13
14 378 attempted to minimize this possibility through a screening process that utilized two reviewers
15
16 379 (with a third to resolve discrepancies), an a priori agreed upon definition of health services
17
18 380 research, and reaching out to study authors for clarification when necessary. Further, the
19
20 381 engagement of patient co-researchers in our scoping review at the level of involve limited their
21
22 382 opportunities to formally provide insights and expertise to pre-determined study milestones.
23
24 383 Engagement at the level of collaborate or empower may have resulted in other insights through
25
26 384 more direct contact with the study.
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33 386 *Comparison with other studies*

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35 387 Previous reviews of models, frameworks, and/or other systematic approaches to the engagement
36
37 388 of patients and other knowledge users in research had a broader scope¹¹⁻¹³ (albeit only one
38
39 389 involved a comprehensive search of multiple databases¹¹) and different levels of analyses.
40
41 390 Specifically, rather than report underlying elements, Greenhalgh et al. developed a taxonomy for
42
43 391 the classification of identified systematic approaches (i.e., tools, frameworks, benchmarks,
44
45 392 guidelines, and critical appraisal checklists) based on their primary focus and intended purpose
46
47 393 (i.e., power-focused, priority-setting, study-focused, report-focused, partnership-focused)¹¹.
48
49 394 Innovatively, they piloted co-design workshops that aimed to improve the aesthetic appeal and
50
51 395 usability of “best in-class” resources identified through the review. Similar to our questioning of
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3 396 the appropriateness of a “one-size fits all” framework, preliminary findings from their co-design
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5 397 workshops indicated that although stakeholders were presented with a common set of resources,
6
7 398 they generated widely differing frameworks suited to meet their different needs and purposes.
9
10 399 The review by Jull et al. reported 15 high-level concepts for knowledge user engagement in
11
12 400 research that they identified through a directed content analysis of underlying elements ¹³.
13
14 401 Although they organized these concepts across four general research phases (i.e., prepare, plan,
15
16 402 conduct, apply), they concluded that variation in the reported concepts between frameworks
17
18 403 indicated that research teams should consider the concepts as fluid rather than strictly required.
19
20 404 Lastly, the findings of Harrison et al.’s narrative review of frameworks and guidelines
21
22 405 culminated in the proposal of an overarching framework that (similar in principle to our scoping
23
24 406 review) conceptualized three distinct but inter-related elements of the patient engagement in
25
26 407 research process - engagement foundational principles (i.e., domains that original study authors
27
28 408 “... considered foundational to patient engagement in research;” n = 15 elements), engagement
29
30 409 best practices (i.e., best practice activities to support engagement; n = 25 elements), and research
31
32 410 phases where engagement should occur (n = 3 elements) ¹². Taken together, existing reviews
33
34 411 demonstrate diversity in how patient engagement in research has been conceptualized through
35
36 412 models, frameworks, and other systematic approaches and the approaches used to synthesize
37
38 413 their findings.
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45 415 *The meaning of the study*

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47 416 Our findings indicate that the conceptualization of patient engagement in health services research
48
49 417 varies between existing models and frameworks. Although models and frameworks posit to
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51 418 break down a concept into its base components (which implies the existence of convergence
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3 419 among underlying elements), this finding is not surprising as patient engagement is an *approach*
4
5 420 to research rather than a *method*. Contributing to the heterogeneity among identified elements is
6
7 421 the diversity in the populations, contexts, and approaches used to develop the models and
8
9 422 frameworks, which also emphasizes the importance of avoiding a “one size fits all” approach to
10
11 423 engagement. Perhaps congruence between models and frameworks actually exists at the level of
12
13 424 conceptual categories. These could be taken to represent the essential components of patient
14
15 425 engagement in health services research. The elements that underlie them, as identified across the
16
17 426 various models and frameworks, would then serve as considerations for researchers and patient
18
19 427 co-researchers when planning and operationalizing patient engagement in health services
20
21 428 research. For this to be better developed from the current literature, consensus is needed on the
22
23 429 definitions of the underlying categories, followed by some re-shuffling of the elements across the
24
25 430 conceptual categories in order to align with the agreed upon definitions.
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33 432 ***Unanswered questions and future research***

34
35 433 In sum, our study found that there is little overlap between the elements that comprise existing
36
37 434 models and frameworks of patient engagement in health services research. Readers seeking to
38
39 435 apply our findings to their own engaged work should consider the “fit” of each element, by
40
41 436 conceptual category and domain, within the context of their study. Future research that builds
42
43 437 upon our work should consider addressing three major gaps. First, most existing models and
44
45 438 frameworks identified factors that needed improvement for patient co-researchers to be better
46
47 439 research partners, with a lot less consideration paid to factors that needed improvement for
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49 440 academic co-researchers to be better research partners. This may be in large part due to the fact
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51 441 the majority of the models were written from an academic researcher lens (as evidenced, for
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3 442 example, by the fact that only a third of the included models and frameworks stated that they
4
5 443 involved patients in their development). Second, there is a lack of focus on the intrapersonal
6
7 444 domain of engagement, including the “soft skills” that underlie interpersonal interactions and
8
9 445 relationships (e.g., body language, wording, tone), and the environmental domain of engagement
10
11 446 which shapes the context in which engagement is set. Third, patient co-researchers are not just
12
13 447 “patients” that can be lumped into a single homogenous category. They are people with different
14
15 448 backgrounds, skills, and interests extending beyond their health conditions or needs. Thus, it is
16
17 449 important to incorporate a trauma-informed and intersectional approach that acknowledges and
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19 450 promotes an understanding of human beings as shaped by the interactions of different social
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21 451 locations and experiences³⁶. Relatedly, it is important for patient co-researchers and academic
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23 452 researchers to get to know each other as people, instead of making assumptions (including about
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25 453 a patient co-researcher’s experience with healthcare services). Engagement is as much about
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27 454 relational interactions as it is research processes. Careful attention needs to be paid to both for
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29 455 academic-patient co-researcher relationships to thrive.
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17
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22 473 interpretation, and provided manuscript edits. In addition, T.H. was responsible for data
23
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44 482 author.
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49 484 **Ethics statements**

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51 485 **Patient consent for publication:** Not applicable.
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3 486 **Ethics approval:** This paper synthesized publicly available articles and therefore did not require
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40 613 **FIGURE TITLES**

41
42 614 **Figure 1. Concept map – principles of patient engagement in health services research**

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44 615 **Figure 2. Concept map – foundational components of patient engagement in health services**
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46 616 **research**

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49 617 **Figure 3. Concept map – contexts of patient engagement in health services research**

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51 618 **Figure 4. Concept map – actions of patient engagement in health services research**

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53 619 **Figure 5. Concept map – outcomes of patient engagement in health services research**

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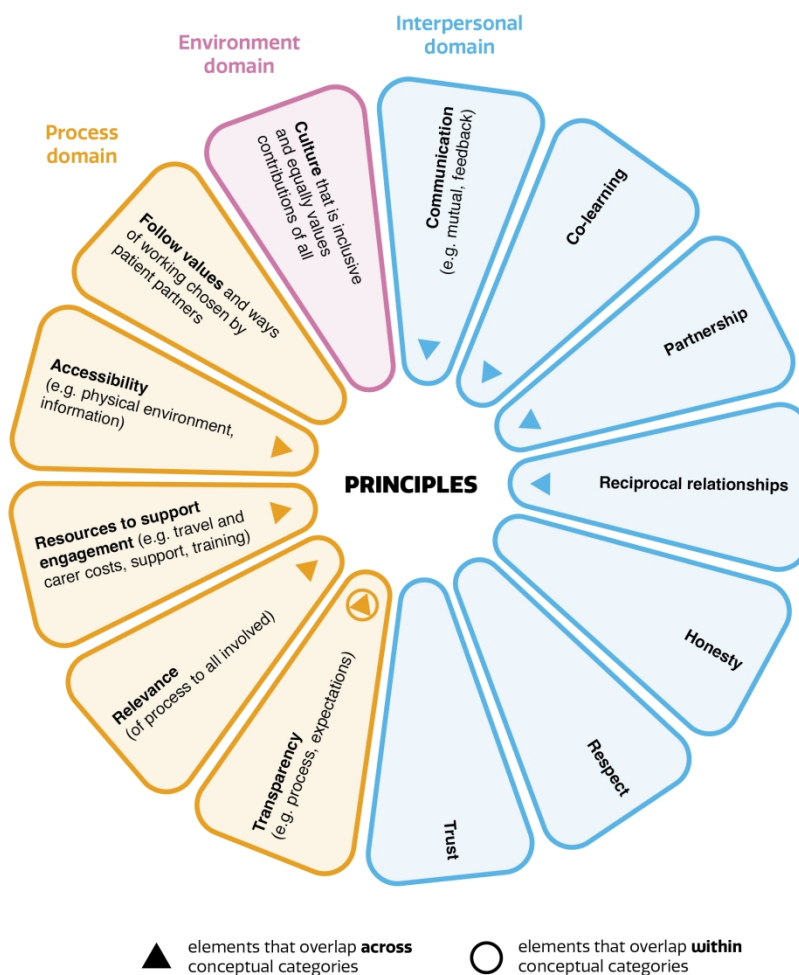


Figure 1. Concept map – principles of patient engagement in health services research

548x580mm (118 x 118 DPI)

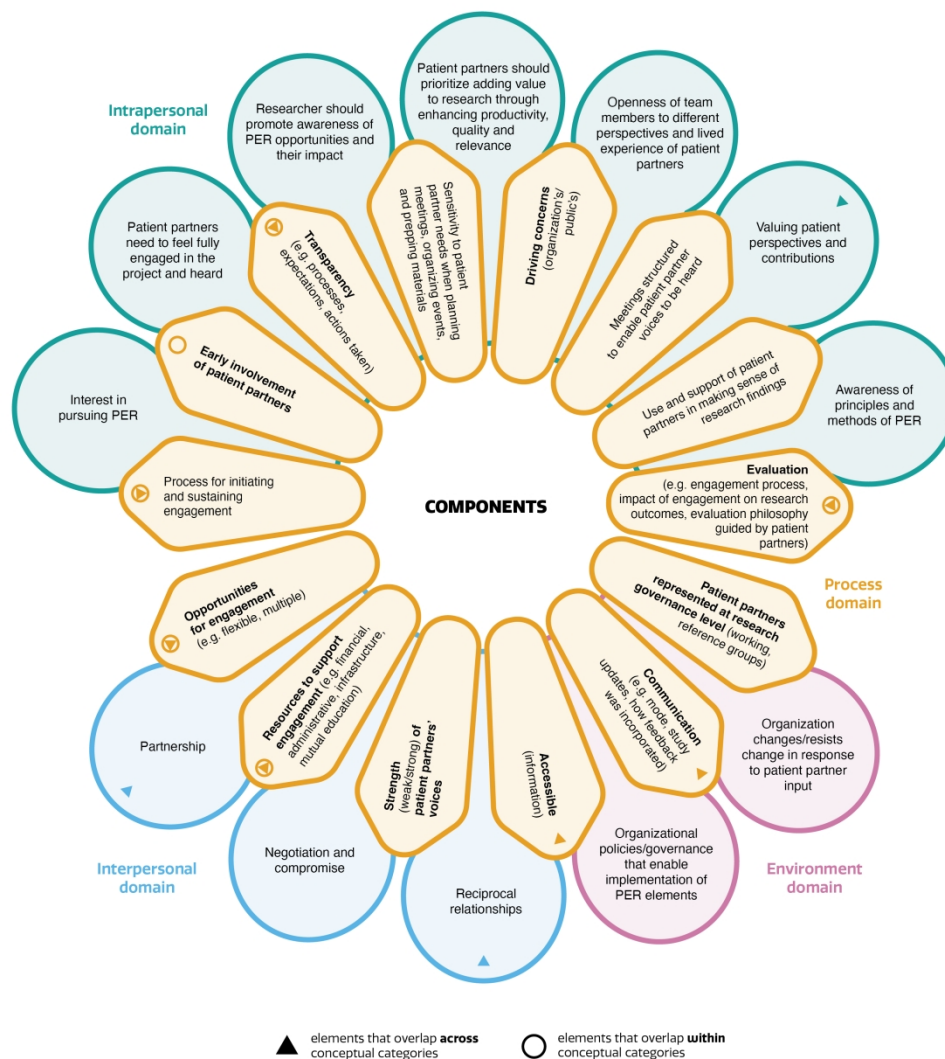


Figure 2. Concept map – foundational components of patient engagement in health services research

696x773mm (118 x 118 DPI)

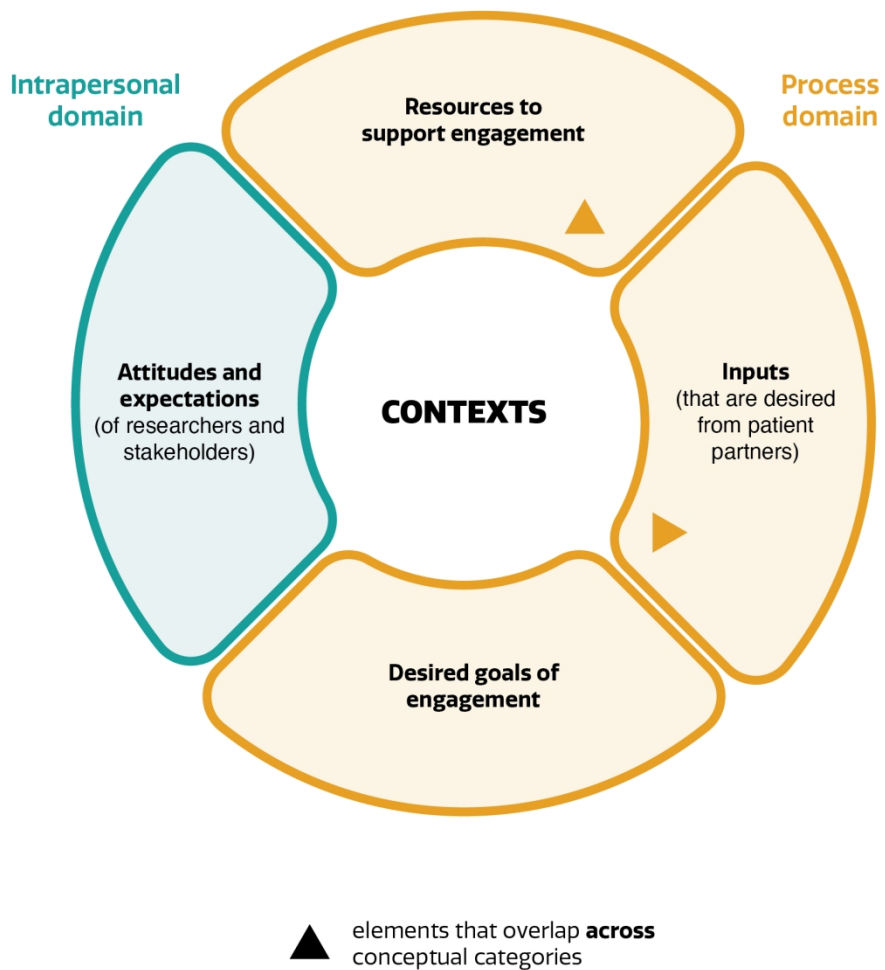


Figure 3. Concept map – contexts of patient engagement in health services research

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

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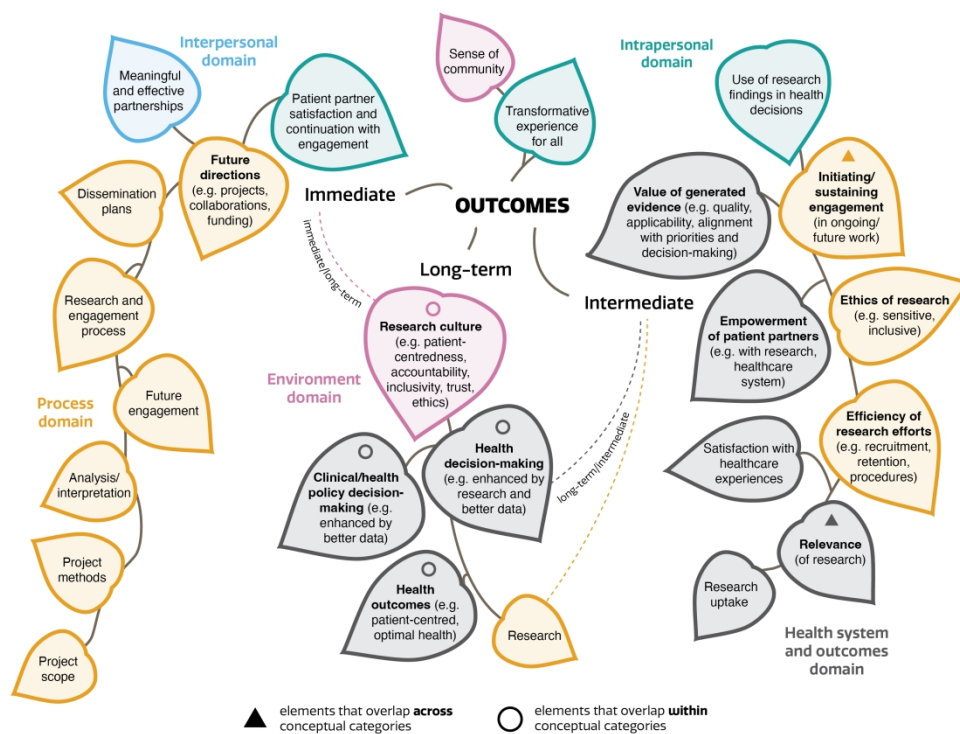


Figure 5. Concept map – outcomes of patient engagement in health services research

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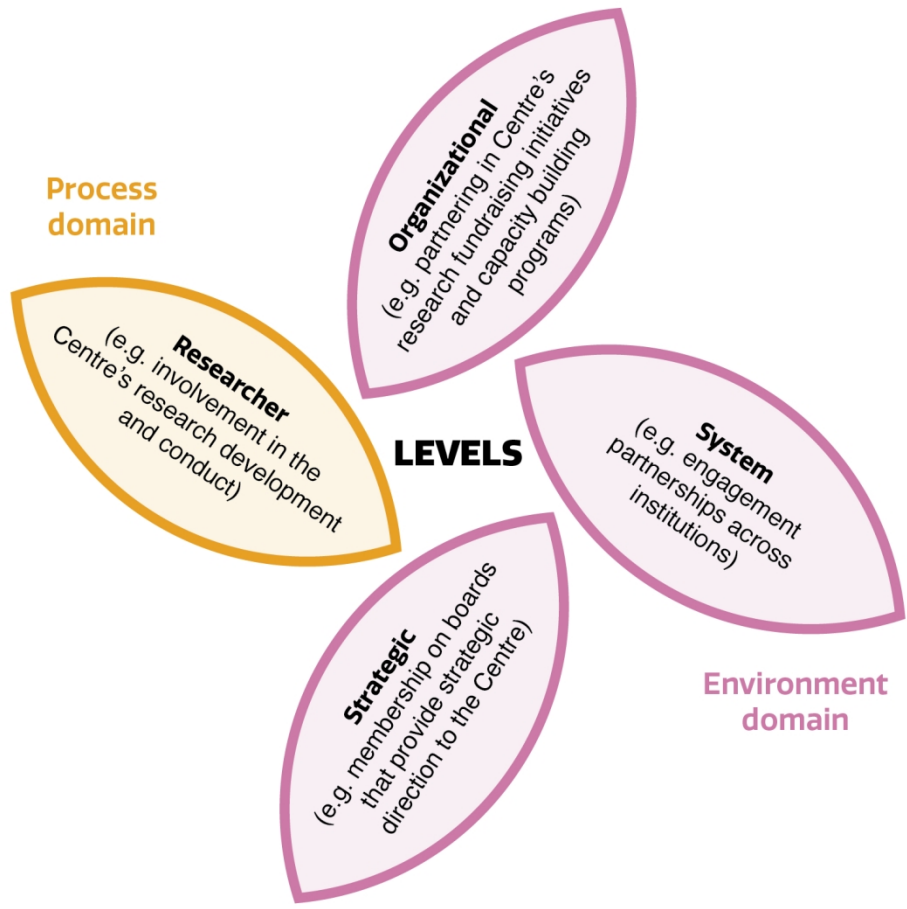


Figure 6. Concept map – organizational levels of patient engagement in health services research

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 (<i>dd/mm/yyyy</i>)	
Name of data extractor	

Study authors (characteristics of)

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

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

Publication (characteristics of)

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

	Descriptions as stated in report/paper or note otherwise	Location in text or source (<i>pg & ¶/fig/table/other</i>)
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)

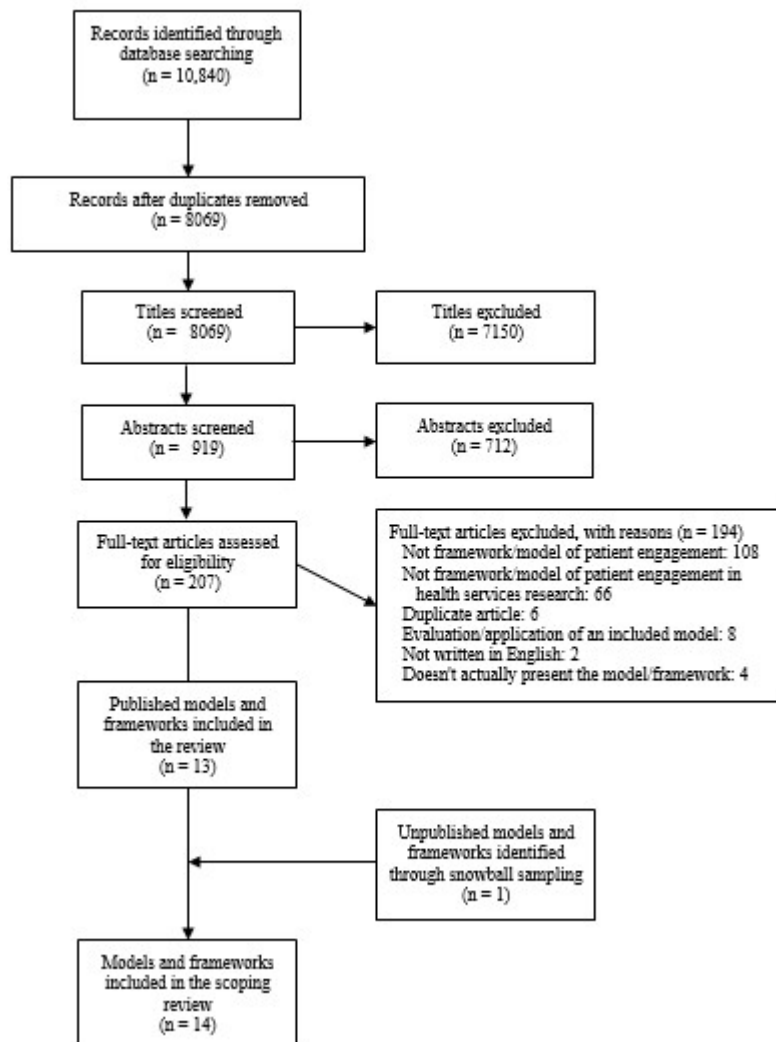
Definitions

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: <ul style="list-style-type: none"> • 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 ...

Model/framework

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)
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	with, a chronic or long- term condition 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 ...

Model/framework

Article				
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 ...

Model/framework

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)
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 (PRISMA-ScR): Checklist and Explanation. *Ann Intern Med.* 2018;169:467–473. doi: 10.7326/M18-0850.



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