

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	A scoping review of models and frameworks of patient engagement in health services research
AUTHORS	Chudyk, Anna; Horrill, Tara; Waldman, Celeste; Demczuk, Lisa; Shimmin, Carolyn; Stoddard, Roger; Hickes, Serena; Schultz, Annette

VERSION 1 – REVIEW

REVIEWER	Majid, Umair University of Toronto
REVIEW RETURNED	18-May-2022

GENERAL COMMENTS	<p>Abstract</p> <ul style="list-style-type: none">• Great! <p>Article Summary</p> <ul style="list-style-type: none">• One limitation is also the exclusion of models and frameworks used in governments and organizations at the regional, provincial, and national levels. These frameworks would have been found in grey literature searching. <p>Introduction</p> <ul style="list-style-type: none">• Great! <p>Methods</p> <ul style="list-style-type: none">• I am a bit confused about whether this focuses on models and framework of health services research, or on health services management and organization. It seems like you have conflated both or at least not acknowledged this overlap in your work. Patient engagement can happen in research and health services management. <p>Results</p> <ul style="list-style-type: none">• Great! <p>Discussion</p> <ul style="list-style-type: none">• Thank you for a great discussion. You mention that this paper will allow people to consider different elements of patient engagement in health services research. However, I do not see a meaningful discussion on how those considerations can be made by someone who is looking to apply the findings of this paper in their work. I think one thing that would strengthen this paper is a conversation about how the various elements you have identified can be used in
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	programs and initiatives. Examples can also be used from the elements you generated to develop this conversation.
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REVIEWER	Tobiano, Georgia Griffith University, Centre for Health Practice Innovation, Menzies Institute for Health (Queensland)
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REVIEW RETURNED	28-Jun-2022
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GENERAL COMMENTS	<p>Thank you for the opportunity to read this well written manuscript. It is a scoping review of models and frameworks for patient engagement in health services research. Through a clear and in-depth synthesis process the authors are able to demonstrate the lack of overlap between models.</p> <ul style="list-style-type: none">- Very clear and great introduction. On page 6 line 71 consider adding consumer and community involvement to your list of commonly referred to terms - this is the term often used in my country- On page 7 line 101 the sentence: "only one describes the elements that comprised identified frameworks and best practice guidelines". The term "best practice guidelines" seemed out of context, could you please provide more detail about this. Great to see that you have clearly stated what previous reviews have been conducted in the field.- Page 8 line 134 the sentence states "its underlying protocol" consider changing to "the protocol for this scoping review". The language was a little unclear as a reader- For "selection" on page 10 was a third reviewer available to resolve any discrepancies and did this need to occur?- The "synthesis of results" section on page 11 is largely clear and has great detail. Please consider the use of the word validation in the first sentence - I'm not sure if this is the best use of term in the context of the synthesis. This section could be strengthened by providing more detail in the middle section of this paragraph. First the sentence that states "with increased familiarity the lead author was able to confine and reframe where appropriate similar elements found in the original publication". I got a little lost on this sentence, what is meant by the term elements, and could you give an example of what you mean by reframe where appropriate- an example might make this clear in the reader's mind. Second, the sentence that states "during this process it became evident that the elements were organised into similar overarching...". Could you provide a little bit more detail about how this table emerged? Were these categories and domains identified aprior or did these come inductively during the synthesis exercise?- Compare appendix 3 to the description in the text as they differ in terms of one stating that 193 excluded at full text whereas the other said 194 excluded at full text- Great to see the two patient co-researchers were involved in the research process and that strategies like terms of references were developed at the start of the study- When I get to the results, I got the sense that Table 1 might actually be informed by Deverka et al and Evans et al research (refer to my earlier comment about whether Table 1 content was inductive or deductive)? I think this needs to be clearer in the methods.
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	<ul style="list-style-type: none"> - In the discussion for the section “comparison to other studies” the research team provides a really great overview of three previous reviews. Could they conclude this paragraph with a sentence that gives their own critical voice about the main message they're trying to send about the comparison between their scoping review and these three reviews? - For the “future directions to research” I was a little unclear about what this meant by “identified perceived deficits” could there be a little bit more description here? - I was intrigued by the researchers mapping their elements to the “domains” such as interpersonal and intrapersonal process etc. Did the researchers find any interesting findings relating to the domains or have any interesting reflections on this that they can add to the discussion? - Great use of the GRIPP2 short form checklist in the supplementary material and great reflections on this process
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1		
1	N/A (no change requested).	N/A
2	Clarified that unpublished encompasses grey literature.	31-36
3	N/A (no change requested).	N/A
4	Clarified that we also included models and frameworks that conceptualized how patient engagement in research was embedded across the different levels of health services research organizations.	146-148
5	N/A (no change requested).	N/A
6	Better describe the direct applications of our findings.	363-379
Reviewer 2		
1	N/A (no change requested).	N/A
2	Added consumer and community involvement to our list of commonly referred to terms.	71-73
3	Clarified what is meant by ‘best practice guidelines.’	101-103
4	Changed wording to that suggested by reviewer.	136-137
5	Added information on the role of a third reviewer during article screening.	182-184

6	More clearly describe our synthesis, including removing the word ‘validation’ from the first sentence of the applicable section, providing a clarifying example of how similar elements identified in the original studies were combined into an over-arching element, and specifying that conceptual categories and domains were identified inductively.	202-212
7	Consistently state that we screened 194 full-text articles.	240-242
8	N/A (no change requested).	N/A
9	Clarify that our conceptual categories and domains were identified inductively during the synthesis exercise.	210-211
10	Added a sentence describing the main message we are trying to send about the comparison between ours and others’ reviews.	425-428
11	Rewrote the identified sentence to more clearly state what we meant.	452-455
12	Added a statement about the lack of focus on the intrapersonal and environmental domains of engagement to our unanswered questions section.	458-461
13	N/A (no change requested).	N/A

Editor(s)' Comments to the Authors:

Comment 1:

Please reorder the abstract format so that 'Data sources' comes before 'Eligibility criteria'. Please also ensure that the abstract contains an adequate description of the search strategy and selection criteria. The "Data sources" section should list all the databases searched and the date range covered by the search. The "Eligibility criteria" section should include language restrictions as well as key inclusion and exclusion criteria. Please also delete the 'Trial registration' section, as this is not relevant so is not required.

RESPONSE

We have revised the abstract to reflect the suggested changes.

CHANGES IN MANUSCRIPT

Lines 31-36:

“Data sources: On July 6 and 7, 2021, we searched six electronic databases (CINAHL, Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLINE, PsycINFO, and Scopus) and Google Scholar for published literature, and ProQuest Dissertations & Theses and Conference Proceedings Citation Index, Google, and key agencies’ websites for unpublished (i.e., grey) literature. These searches were supplemented by snowball sampling.

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

Comment 2:

If there were any deviations from the published protocol (reference 19), please ensure that these deviations (and explanations for them) are highlighted in the main text Methods section.

RESPONSE

We have doubled checked that we do not have any protocol deviations to report. Thanks for this reminder!

CHANGES IN MANUSCRIPT

None required.

Comment 3:

Please change the heading “Patient involvement” to “Patient and public involvement”.
Please also clarify what is meant by “at the level of involve”.

RESPONSE

We have changed the heading and clarify what we meant by involve and how we arrived at this decision.

CHANGES IN MANUSCRIPT

Lines 219-225:

“Patient **and public** involvement

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

Comment 4:

Please include, as a supplemental appendix, the precise, full search strategies for all databases, registers and websites, including any filters and limits used. This should replace the current Appendix 1 (MEDLINE-only search strategy) and the citation of this appendix in the main text should be updated accordingly.

RESPONSE

We now include the full search strategies for all databases, registers, and websites in Appendix 1 and have updated the description of the appendix in the main text.

CHANGES IN MANUSCRIPT

Lines 168-169:

“The search **strategies for all databases, registers, and websites** are found in Appendix 1.”

Appendix 1

Comment 5:

Throughout the manuscript, please change ‘MEDLine’ to ‘MEDLINE’.

RESPONSE

We have made this change throughout the manuscript.

CHANGES IN MANUSCRIPT

Lines 33 and 155:

Changed 'MEDLine' to 'MEDLINE'

Comment 6:

Please change the headings 'Funding statement', 'Competing interests statement', 'Author Contributions' and 'Data sharing statement' to 'Funding', 'Competing interests', 'Contributors' and 'Data availability statement', respectively.

RESPONSE

We have changed the headings as suggested. We also added an 'acknowledgements' heading after 'contributors.'

CHANGES IN MANUSCRIPT

Relevant headings changed to ‘Funding, ‘Competing interests’, ‘Contributors’ and ‘Data availability statement.’

Lines 420-44:

“Acknowledgements: We offer our sincerest thanks to Mr. Patrick Faucher for designing our figures, and to the Manitoba SPOR Support Unit (George & Fay Yee Centre for Healthcare Innovation) for Mr. Faucher’s services.”

Reviewer 1:

Comment 1:

Abstract - Great!

RESPONSE

Thank you!

CHANGES IN MANUSCRIPT

None.

Comment 2:

Article Summary - One limitation is also the exclusion of models and frameworks used in governments and organizations at the regional, provincial, and national levels. These frameworks would have been found in grey literature searching.

RESPONSE

Great suggestion about grey literature searching – one of the reasons we searched the unpublished/grey literature was to identify these models and frameworks. We now clarify that by “unpublished literature” we also mean grey literature the first time we use the term unpublished in the abstract and have doubled checked that we also do this the first time the word “unpublished” appears in the body of the manuscript.

CHANGES IN MANUSCRIPT

Lines 31- 36:

“Data sources: On July 6 and 7, 2021, we searched six electronic databases (CINAHL, Cochrane Database of Systematic Reviews, Joanna Briggs Institute Evidence Based Practice Database, MEDLINE, PsycINFO, and Scopus) and Google Scholar for published literature, and ProQuest Dissertations & Theses and Conference Proceedings Citation Index, Google, and key agencies’ websites for unpublished (**i.e., grey**) literature.”

Comment 3:

Introduction - Great!

RESPONSE

Thanks so much for incorporating positivity into your review!

CHANGES IN MANUSCRIPT

None required.

Comment 4:

Methods - I am a bit confused about whether this focuses on models and framework of health services research, or on health services management and organization. It seems like you have conflated both or at least not acknowledged this overlap in your work. Patient engagement can happen in research and health services management.

RESPONSE

You're right. Thanks for the opportunity to clarify this point in our methods.

CHANGES IN MANUSCRIPT

Lines 146-148:

“Since patient engagement also encompasses participation in research governance¹, we included models and frameworks that conceptualized how patient engagement in research was embedded across the different levels of health services research organizations.”

Comment 5:

Results - Great!

RESPONSE:

Thank you! Please note that we have worked with a knowledge translation expert to update the visual presentation of our figures.

CHANGES IN MANUSCRIPT

None required.

Comment 6:

Discussion - Thank you for a great discussion. You mention that this paper will allow people to consider different elements of patient engagement in health services research. However, I do not see a meaningful discussion on how those considerations can be made by someone who is looking to apply the findings of this paper in their work. I think one thing that would strengthen this paper is a conversation about how the various elements you have identified can be used in programs and initiatives. Examples can also be used from the elements you generated to develop this conversation.

RESPONSE

Thank you for this suggestion. We originally attempted to describe how our findings could be applied to programs and initiatives at the end of the first paragraph of the discussion. We have now revised this section to be clearer.

CHANGES IN MANUSCRIPT

Lines 363-379:

“Our approach to data synthesis is also unique in that we attempt to maximize the intuitiveness and applicability of our findings by presenting elements by overarching conceptual categories (i.e., principles, foundational components, contexts, actions, levels, and outcomes) and corresponding domains (i.e., intrapersonal, interpersonal, process, and environment). We anticipate this approach will facilitate the ready application of our findings to readers’ own research programs by serving as a “toolbox” of elements to consider according to the multi-level facets of a research team and study. **To illustrate, research partners could begin to meet at a study’s outset to co-develop terms of reference that guide the relational and activity related aspects of the study’s engagement process. In doing so, they could reflect upon whether/how elements within the domains of each conceptual category resonate with their study’s engagement process (as influenced by factors such as the study design, available resources, research partner strengths and interests, etc.) These conversations could be guided by prompts such as “do we want to embody or incorporate this element within our study (why/why not)?”, “what does the embodiment or incorporation of this element look and feel like to us?”, “how will we know when we have or have not embodied or incorporated this element within our study?” In doing so, the “toolbox” of elements found within our review is transformed into a co-developed “roadmap” to help guide a study’s engagement process.**

Reviewer 2:

Comment 1:

Thank you for the opportunity to read this well written manuscript. It is a scoping review of models and frameworks for patient engagement in health services research. Through a clear and in-depth synthesis process the authors are able to demonstrate the lack of overlap between models.

RESPONSE

Thank you for the kind words!

CHANGES IN MANUSCRIPT

None required.

Comment 2:

Very clear and great introduction. On page 6 line 71 consider adding consumer and community involvement to your list of commonly referred to terms - this is the term often used in my country

RESPONSE

Thanks for helping us to make this list even more inclusive of common terms for patient engagement in research.

CHANGES IN MANUSCRIPT

Lines 71-73:

“Globally, it is also commonly referred to as patient and public involvement, patient involvement, **consumer and community involvement**, and stakeholder engagement in research.”

Comment 3:

On page 7 line 101 the sentence: “only one describes the elements that comprised identified frameworks and best practice guidelines”. The term “best practice guidelines” seemed out of context, could you please provide more detail about this. Great to see that you have clearly stated what previous reviews have been conducted in the field.

RESPONSE

We now clarify what is meant by ‘best practice guidelines’ based on the information provided in the relevant review.

CHANGES IN MANUSCRIPT

Lines 101-103:

“Of these, only one described the elements that comprised identified frameworks and **guidelines describing best practices for engaging patients in research.**”

Comment 4:

Page 8 line 134 the sentence states “its underlying protocol” consider changing to “the protocol for this scoping review”. The language was a little unclear as a reader

RESPONSE

We have changed the wording as suggested. We appreciate all of your ideas on how to make our writing clearer.

CHANGES IN MANUSCRIPT

Lines 136-137:

“The **protocol for this scoping review**, including definitions of underlying key concepts, is published elsewhere.”

Comment 5:

- For “selection” on page 10 was a third reviewer available to resolve any discrepancies and did this need to occur?

RESPONSE

We now provide this information under ‘selection,’

CHANGES IN MANUSCRIPT

Lines 182-184:

“A third reviewer (A.S.) helped resolve discrepancies at the close of level 2 and 3 screening, which predominantly dealt with whether identified literature focused on health services research.”

Comment 6:

- The “synthesis of results” section on page 11 is largely clear and has great detail. Please consider the use of the word validation in the first sentence - I'm not sure if this is the best use of term in the context of the synthesis. This section could be strengthened by providing more detail in the middle section of this paragraph. First the sentence that states “with increased familiarity the lead author was able to confine and reframe where appropriate similar elements found in the original publication”. I got a little lost on this sentence, what is meant by the term elements, and could you give an example of what you mean by reframe where appropriate- an example might make this clear in the reader's mind. Second, the sentence that states “during this process it became evident that the elements were organised into similar overarching...”. Could you provide a little bit more detail about how this table emerged? Were these categories and domains identified aprior or did these come inductively during the synthesis exercise?

RESPONSE

Thanks so much for helping us more clearly describe our analysis. We have removed the word validation from the first sentence of this paragraph and also provide a clarifying example of how similar elements identified in the original studies were combined into an over-arching element. We now also specify that these categories and domains were identified inductively during the synthesis exercise.

CHANGES IN MANUSCRIPT

Lines 202-212:

“With increased familiarity, the lead author was able to combine and reframe (where appropriate) similar elements found in the original publications. **For example, the elements ‘improved quality health decisions’ and ‘improved patient decision making’ were combined into the element ‘health decision-making’ based on similarities in the element**

names and descriptions provided by the original study authors. The revised elements were used to obtain counts of overlapping and diverging elements. During this process, it became evident that elements were organized into similar over-arching conceptual categories by the original authors and that they could also be located within multiple domains (Table 1). **These categories and domains were identified inductively during the synthesis exercise.** As such, in our analysis, we mapped each element according to conceptual category and domain.”

Comment 7:

- Compare appendix 3 to the description in the text as they differ in terms of one stating that 193 excluded at full text whereas the other said 194 excluded at full text

RESPONSE

Thank you so much for catching that. We have changed the text to consistently state the correct number (194).

CHANGES IN MANUSCRIPT

Lines 240-242:

“After applying inclusion criteria, 7150 articles were excluded at the title screen, 712 at the abstract screen, and **194** at the full-text screen, leaving 13 models and frameworks.”

Comment 8:

- Great to see the two patient co-researchers were involved in the research process and that strategies like terms of references were developed at the start of the study

RESPONSE

Thank you! It is always nice to cross paths with individuals that also appreciate the value of patient co-researchers.

CHANGES IN MANUSCRIPT

Comment 9:

- When I get to the results, I got the sense that Table 1 might actually be informed by Deverka et al and Evans et al research (refer to my earlier comment about whether Table 1 content was inductive or deductive)? I think this needs to be clearer in the methods.

RESPONSE

Yes. You are right. As a result of this comment and your 6th comment, we now clarify in the methods that our conceptual categories and domains were identified inductively during the synthesis exercise. In terms of the results, please also note that we have worked with a knowledge translation expert to update the visual presentation of our figures (but their content has remained the same).

CHANGES IN MANUSCRIPT

Lines 210-211:

“These categories and domains were identified inductively during the synthesis exercise.”

Comment 10:

- In the discussion for the section “comparison to other studies” the research team provides a really great overview of three previous reviews. Could they conclude this paragraph with a sentence that gives their own critical voice about the main message they're trying to send about the comparison between their scoping review and these three reviews?

RESPONSE

We now conclude this section with a statement that summarizes the main messages we're trying to send about the comparison between ours and others' reviews.

CHANGES IN MANUSCRIPT

Lines 425-428:

“Taken together, existing reviews demonstrate diversity in how patient engagement in research has been conceptualized through models, frameworks, and other systematic approaches and the approaches used to synthesize their findings.”

Comment 11:

- For the “future directions to research” I was a little unclear about what this meant by “identified perceived deficits” could there be a little bit more description here?

RESPONSE

Thank you for bringing this confusing sentence to our attention. We have re-written this sentence to more clearly state what we mean.

CHANGES IN MANUSCRIPT

Lines 452-455:

“First, most existing models and frameworks identified factors that needed improvement for patient co-researchers to be better research partners, with a lot less consideration paid to factors that needed improvement in order for academic co-researchers to be better research partners.

Comment 12:

- I was intrigued by the researchers mapping their elements to the “domains” such as interpersonal and intrapersonal process etc. Did the researchers find any interesting findings relating to the domains or have any interesting reflections on this that they can add to the discussion?

RESPONSE

In our unanswered questions section, we now state that there is a lack of focus on the intrapersonal and environmental domains of engagement.

CHANGES IN MANUSCRIPT

Lines 458-461:

“Second, there is a lack of focus on the intrapersonal domain of engagement, including the “soft skills” that underlie interpersonal interactions and relationships (e.g., body language, wording, tone), and the environment domain of engagement which shapes the context in which engagement is set.”

Comment 13:

- Great use of the GRIPP2 short form checklist in the supplementary material and great reflections on this process

RESPONSE

Thanks again for all of your kind words and thoughtful suggestions. We really appreciate it.

CHANGES IN MANUSCRIPT

None required.

VERSION 2 – REVIEW

REVIEWER	Tobiano, Georgia Griffith University, Centre for Health Practice Innovation, Menzies Institute for Health (Queensland)
REVIEW RETURNED	29-Jul-2022
GENERAL COMMENTS	Thank you for taking the time to thoughtfully address revisions suggested. Research team have done fantastic job addressing reviewer comments.