

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making in healthcare systems: a scoping review
AUTHORS	Hamilton, Clayton; Dehnadi, Maryam; Snow, M. Elizabeth; Clark, Nancy; Lui, Michelle; McLean, Janet; Mamdani, Hussein; Kooijman, Allison; Bubber, Vikram; Hoefer, Tammy; Patients as Partners, Team; Li, Linda

VERSION 1 – REVIEW

REVIEWER	Kirk, Andrew University of Saskatchewan, Medicine
REVIEW RETURNED	04-May-2021

GENERAL COMMENTS	<p>Hamilton et al. present a scoping review of literature reviews evaluating initiatives to include patients and family caregivers in making decisions concerning healthcare systems. This seems a useful endeavor which may yield progress in helping to define appropriate means of involving patients and caregivers in healthcare system improvement. Previous individual reviews may have been idiosyncratic and not informed by themes used in other reviews. The information presented may thus prove helpful to future researchers and planners. I have only a few comments.</p> <ol style="list-style-type: none"> 1. There is ambiguity in the title. A reader doesn't immediately know whether "patient and family caregiver" means two groups consisting firstly of patient caregivers (which to me may suggest professional caregivers such as physicians, nurses, etc) and secondly of family caregivers or whether it refers to the two groups of patients and of family caregivers. It turns out from reading the article that it means the latter. Until reading the article one doesn't know whether the word "patient" is used as an ordinary noun or as a noun adjunct describing the word "caregiver." One could fix this for greater clarity by changing the grammar of the title so that the words "patient" and "caregiver" could be pluralized. 2. The sentence on page 6, line 146 seems to be missing a verb. 3. On page 8 we're told that only studies from upper middle and high income countries were selected. It might be helpful to explicitly state in the text how much this reduced the number of studies included. My guess is that it may not have resulted in much of a reduction but this would be helpful to know. 4. In Table 2, under outcome, number 6, "improve" should presumably read "improved" in both appearances. 5. On page 20, the word "parameters" is used. I don't think it's used again and I'm therefore not sure if it's meant in the scientific
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	<p>sense or in the colloquial sense which has come to mean “limits.” If the latter is intended, it might be better in a scientific paper to use a clearer word such as “limits.”</p> <p>6. On page 23, line 350, “allow” should read “allows.”</p> <p>Overall, this paper may prove helpful to future researchers as well as to groups wishing to promote involvement of patients and caregivers in healthcare planning and decision-making.</p>
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REVIEWER	Al-Tannir, Mohamad King Fahad Medical City Faculty of Medicine, Clinical and Translational Research
REVIEW RETURNED	20-May-2021

GENERAL COMMENTS	none
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REVIEWER	van Veenendaal , Haske Erasmus University Rotterdam, ESHPM
REVIEW RETURNED	27-Jul-2021

GENERAL COMMENTS	<p>This report is a comprehensive review aiming to contribute to evaluation of the extent to which policy-driven patient and family caregiver engagement initiatives are effective. Key themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making across the organization and system domains were gathered. Patient and family caregiver partners were engaged as research team members in this study. The subject of this review is current and the results are valuable for research and practice. However, especially the methods section needs to report additional information.</p> <ul style="list-style-type: none"> - The research question aims at evaluation of a provincial engagement initiative? In many countries health care systems are predominantly organized at the national and/or local level. What does this mean for the generalizability of the results? - Although information is available in the protocol that has been published previously, I would like to understand how the predetermined categories were determined? The same accounts for only using reviews in the review. One could argue that valuable information is available in original research. - Please explain why and how 37 original articles are added to the selection of 42 reviews? - The coding process is not clear to me. In the abstract, the number of 5 coders is mentioned. In the methods section, I can only find 4 (CBH, HM, ML, and MD). Next, it is not clear what % is independently coded and what part was checked by a second coder and/or CBH - Results: Implementation/impact is not one of the outcome themes? Or is Cost-effectiveness almost similar to implementation? - The research team of multiple stakeholders met and discussed the preliminary themes identified and their corresponding excerpts. At what time, how many times, who was in the team? - Discussion/conclusion: Of course, it is very relevant to determine initiatives that are effective and investigate their effectiveness. But even so, effectiveness might depend on the context as well. Is it
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	<p>possible that we need to tailor approaches to get them more effective?</p> <ul style="list-style-type: none"> - Is there an explanation for the fact that so few process themes were identified? One would expect that the most information would be available on the process of engaging? - Limitations: I can only find 1 limitation (relevant (grey) literature and input from experts) that might be missed, but I think there are more. E.g. only studies conducted in upper middle- and high-income countries were selected. Is the context of engagement initiatives that sensitive or might interesting research be missed this way? The authors did not assess the methodological quality of the included studies, what could be the effect of this approach.
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REVIEWER	Heier, Lina University Hospital Bonn, Institute for Patient Safety
REVIEW RETURNED	30-Jul-2021

GENERAL COMMENTS	<p>Dear authors</p> <p>Thank you very much for the opportunity to read and comment on your manuscript. This is a relevant and interesting topic addressing key themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making across the organization and system domains of healthcare systems. You have created a very advanced manuscript and I have only very few comments for you which I have addressed below:</p> <ul style="list-style-type: none"> - Abstract: Please add a sentence that the categories structure, process and outcome are adapted from the Donabedian conceptual framework. - Introduction, lines 110 & 111: Although the terms are described in more detail in the study protocol, naming them here would be helpful to the reader. - Methods, lines 142 & 143: Please explain why you decided on \$35/hour (is it a common hourly wage for external experts/patient partners?) - Methods, lines 149 & 150: Please clarify “very active in patient engagement” or give an example. - Methods, line 180: Please explain why the search strategy is so broad (in terms of time of publication) (one publication is published over 19 years ago and seems a bit old). - Methods, lines 230 – 232: Please clarify the step “renaming the codes as themes”. What is the exact difference between codes and themes? Maybe you can give an example. - Results (in general): I would suggest to add a brief description of what the following categories and themes are about. E.g., After “Selection themes: Structure comprises the settings or context within which engagement occur, such as organisational structure, materials and human resources”. - Results (in general): Please check citation again, e.g. citation in line 277 differs from citation in line 279.
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	<ul style="list-style-type: none"> - Discussion: Please discuss the involvement and influence of the patient partners on your results. - Discussion & conclusion: Although you selected studies only from upper middle and high income countries, however, healthcare systems differ greatly. Please discuss if and what impact the differences can have on the identified themes and your conclusion.
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VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

Dr. Andrew Kirk, University of Saskatchewan Comments to the Author:

Reviewer comment

Hamilton et al. present a scoping review of literature reviews evaluating initiatives to include patients and family caregivers in making decisions concerning healthcare systems. This seems a useful endeavor which may yield progress in helping to define appropriate means of involving patients and caregivers in healthcare system improvement. Previous individual reviews may have been idiosyncratic and not informed by themes used in other reviews. The information presented may thus prove helpful to future researchers and planners. I have only a few comments.

Authors' response

Thanks for your positive overall assessment of our scoping review. We truly appreciate the valuable suggestions that you have made to strengthen our paper. Please see our responses to each of your comments below.

Reviewer comment

1. There is ambiguity in the title. A reader doesn't immediately know whether "patient and family caregiver" means two groups consisting firstly of patient caregivers (which to me may suggest professional caregivers such as physicians, nurses, etc) and secondly of family caregivers or whether it refers to the two groups of patients and of family caregivers. It turns out from reading the article that it means the latter. Until reading the article one doesn't know whether the word "patient" is used as an ordinary noun or as a noun adjunct describing the word "caregiver." One could fix this for greater clarity by changing the grammar of the title so that the words "patient" and "caregiver" could be pluralized.

Authors' response

Thank you for pointing out this ambiguity. We have updated the title in line with your suggestion. It now reads, "Themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making in healthcare systems: a scoping review". Further to this change, we have updated "patient and family caregiver" in other parts of the manuscript in line with your comments. Please see lines 118, 130-131, 132-133, 230, 454, and 495.

Reviewer comment

2. The sentence on page 6, line 146 seems to be missing a verb.

Authors' response

Line 146 now reads, "ALK is a Caucasian female in her late 30s with a disability..."

Reviewer comment

3. On page 8 we're told that only studies from upper middle and high income countries were selected. It might be helpful to explicitly state in the text how much this reduced the number of studies included. My guess is that it may not have resulted in much of a reduction but this would be helpful to know.

Authors' response

Studies were excluded for being about low and middle-income countries ahead of selecting reviews. Figure 1 shows that 86 studies were excluded for being on low and middle income countries. A further look at those excluded studies indicates that 5 of those 86 studies were reviews of the literature.

In the result section, lines 260 to 261 now reads, "86 excluded articles, including 5 reviews, were on low- or middle-income countries."

Reviewer comment

4. In Table 2, under outcome, number 6, "improve" should presumably read "improved" in both appearances.

Authors' response

We have corrected these typos in Table 2, and corrected other minor typos such as changing "PFC" to the plural form "PFCs". Table 2 is now replaced by an infographic, Figure 2, to enhance the presentation of the themes. Figure 2 include the corrections made to Table 2. The content of the infographic (i.e., Figure 2) is the same as would be in Table 2. The title of Figure 2 reflects your suggestion for refining the title of our paper.

Reviewer comment

5. On page 20, the word "parameters" is used. I don't think it's used again and I'm therefore not sure if it's meant in the scientific sense or in the colloquial sense which has come to mean "limits." If the latter is intended, it might be better in a scientific paper to use a clearer word such as "limits."

Authors' response

We have change the word "parameters" to "limits" on line 280.

Reviewer comment

6. On page 23, line 350, "allow" should read "allows."

Overall, this paper may prove helpful to future researchers as well as to groups wishing to promote involvement of patients and caregivers in healthcare planning and decision-making.

Authors' response

The word "allow" has been change to "allows". Please see line 362.

Reviewer: 2

Dr. Mohamad Al-Tannir, King Fahad Medical City Faculty of Medicine Comments to the Author:

none

Reviewer: 3

Reviewer comment

Dr. Haske van Veenendaal , Erasmus University Rotterdam Comments to the Author:

This report is a comprehensive review aiming to contribute to evaluation of the extent to which policy-driven patient and family caregiver engagement initiatives are effective. Key themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making across the organization and system domains were gathered. Patient and family caregiver partners were engaged as research team members in this study. The subject of this review is current and the results are valuable for research and practice. However, especially the methods section needs to report additional information.

Authors' response

Thank you for your positive overall assessment of our paper. Please see our response below to your further comments.

Reviewer comment

- The research question aims at evaluation of a provincial engagement initiative? In many countries health care systems are predominantly organized at the national and/or local level. What does this mean for the generalizability of the results?

Authors' response

While our scoping review was done to inform evaluation of provincial engagement initiatives, the included studies are literature reviews that span health care systems organized at local to national levels. While our research question addresses the provincial context, users of our paper might find that the evaluation themes are more widely applicable.

Reviewer comment

- Although information is available in the protocol that has been published previously, I would like to understand how the predetermined categories were determined? The same accounts for only using reviews in the review. One could argue that valuable information is available in original research.

Authors' response

Thank you for this question. The pre-determined categories were determined by adopting the Donabedian structure-process-outcome conceptual framework. We reasoned that it fits well with the overarching categories of a logic model. Lines 219 to 222 of our paper now reads, "The Donabedian conceptual framework was used because it is widely accepted as a standard for guiding systematic evaluation of the quality of healthcare and fits well with the overarching categories of a logic model."

We had read all of the 366 relevant articles, and recognized that many of the non-review articles were synthesized and presented in the review articles. To create the "themes for evaluation" as a useful tool, we anticipated that the higher order knowledge presented in the reviews were credible and sufficient to identify the themes. We recognized, however, that some of the non-review articles were not captured in the reviews or provided insights that were not sufficiently captured for themes we identified in our study. Therefore, we selected the 37 non-review articles that provided additional and unique information to supplement each theme's description .

Reviewer comment

- Please explain why and how 37 original articles are added to the selection of 42 reviews?

Authors' response

Thank you for this question. We would like to reiterate our response above, which is applicable here. We had read all of the 366 relevant articles, and recognized that many of the non-review articles were synthesized and presented in the review articles. To create the “themes for evaluation” as a useful tool, we anticipated that the higher order knowledge presented in the reviews were credible and sufficient to identify the themes. We recognized, however, that some of the non-review articles were not captured in the reviews or provided insights that were not sufficiently captured for themes we identified in our study. Therefore, we selected the 37 non-review articles that provided additional and unique information to supplement each theme’s description.

Lines 250 to 254 of our paper now reads, “The summaries were primarily based on literature reviews as higher-order knowledge to create “themes for evaluation” as a useful tool, and were supplemented by non-reviews for richer descriptions. After reading all of the relevant articles, 37 non-review studies (see supplementary file) were used when they provided additional and unique information for each theme’s description.”

Reviewer comment

- The coding process is not clear to me. In the abstract, the number of 5 coders is mentioned. In the methods section, I can only find 4 (CBH, HM, ML, and MD). Next, it is not clear what % is independently coded and what part was checked by a second coder and/or CBH

Authors' response

We have corrected the typo in the abstract to state 4 coders instead of 5 coders.

All of the data were independently coded by the coders. Initially all of the coders independently coded the same portion of data, and then after training on the data they independently coded separate portion of data. So as not to confuse readers into thinking that two or more people coded the excerpts for each category after the training, lines 233 to 235 now reads, “After this training on open-coding and creating an initial coding scheme, HM, ML, and MD each open-coded the excerpts for structure, process, and outcome, respectively.”

Reviewer comment

- Results: Implementation/impact is not one of the outcome themes? Or is Cost-effectiveness almost similar to implementation?

Authors' response

The term impact is use synonymously with the term outcome. Each of the 8 themes in the “outcome” category reflects ways in which engagement impacts aspects of healthcare systems.

Reviewer comment

- The research team of multiple stakeholders met and discussed the preliminary themes identified and their corresponding excerpts. At what time, how many times, who was in the team?

Authors' response

Our research team had six team meetings that included all members of the research team throughout the scoping review study. In addition, team members provided feedback to written documents by email and 1 on 1 phone calls. This is highlighted throughout the paper, where we refer to the activities and contributions of the “research team” such as in lines 254 to 256, where our paper reads “The research

team members further refined the descriptions of the themes through discussions by email, and team meetings, and 1-on-1 phone calls”.

To clarify who was on the team, lines 240 to 243 now reads, “Our research team of multiple stakeholders, listed as the authors in this article, met and discussed the preliminary themes identified and their corresponding excerpts. The discussions led to combining some themes within their overarching categories, renaming some themes, and adding one new theme.”

Reviewer comment

- Discussion/conclusion: Of course, it is very relevant to determine initiatives that are effective and investigate their effectiveness. But even so, effectiveness might depend on the context as well. Is it possible that we need to tailor approaches to get them more effective?

Authors' response

Yes, it is possible to tailor approaches to get them more effective. This is reflected in the conclusion where we state “the themes can be used to investigate the mechanisms through which relevant initiative are effective”. The mechanism, and thus the context, are captured in the categories of “Structure” and “Process”. We have also added the definition “Structure” to lines 276-277, which reads “Structure comprises the settings or contexts within which engagement occur, such as organisational structure, materials and human resources.”

Reviewer comment

- Is there an explanation for the fact that so few process themes were identified? One would expect that the most information would be available on the process of engaging?

Authors' response

This is a very good question. The themes within the “Process” category are broad concepts, such as “Engagement approach” which further studies would demonstrate have multiple sub-themes. The number of themes is not reflective of number of possible subthemes. Further to this, the number of themes under each category does not in anyway reflects the level of importance of a category.

Reviewer comment

- Limitations: I can only find 1 limitation (relevant (grey) literature and input from experts) that might be missed, but I think there are more. E.g. only studies conducted in upper middle- and high-income countries were selected. Is the context of engagement initiatives that sensitive or might interesting research be missed this way? The authors did not assess the methodological quality of the included studies, what could be the effect of this approach.

Authors' response

We have added to lines 486-488 of the discussion section, “By limiting included studies to those about upper middle and high income countries, our findings need to be confirmed for generalizability to low and middle income countries.”

You are right that not assessing the methodological quality of the included studies was as an effect of the established scoping review approach. We have updated lines 163 to 164 to now read, “The methodology consists of six stages and does not require assessment of the methodology quality of included studies.”

Reviewer: 4

Mrs. Lina Heier, University Hospital Bonn, University Hospital Bonn Comments to the Author:

Reviewer comment

Dear authors

Thank you very much for the opportunity to read and comment on your manuscript. This is a relevant and interesting topic addressing key themes for evaluating the quality of initiatives to engage patients and family caregivers in decision-making across the organization and system domains of healthcare systems. You have created a very advanced manuscript and I have only very few comments for you, which I have addressed below:

Authors' response

Thank you for your positive feedback on our manuscript.

Reviewer comment

- Abstract: Please add a sentence that the categories structure, process and outcome are adapted from the Donabedian conceptual framework.

Authors' response

Lines 40 to 42 now reads, "We extracted text under three pre-determined categories: structure, process, and outcomes that were adapted from the Donabedian conceptual framework."

Review comment

- Introduction, lines 110 & 111: Although the terms are described in more detail in the study protocol, naming them here would be helpful to the reader.

Authors' response

Thank you for this suggestion. We considered this suggestion, and believe that in stating that "patients and family caregivers" simply describes the many categories of individuals and groups served by healthcare systems" provides adequate detail rather than defining "patient" and "family caregiver" separately. We have removed the previously included text that states "as defined in our published protocol" because it implies that the reader needs to read the protocol, which is not the case.

Reviewer comment

- Methods, lines 142 & 143: Please explain why you decided on \$35/hour (is it a common hourly wage for external experts/patient partners?)

Authors' response

To address your question, the sentence in lines 141 to 143 now reads "Patient partners (AK, ALK, and VB) joined individually when recruited from a provincial community of volunteers and were offered a \$35/hour honorarium as reasonable compensation in line with guidelines by regional health research organizations."

Reviewer comment

- Methods, lines 149 & 150: Please clarify "very active in patient engagement" or give an example.

Authors' response

To clarify, the sentence in lines 149 to 151 now reads, "He is very active in patient engagement such as making conference presentations and being a community advocate for physical and mental health."

Reviewer comment

- Methods, line 180: Please explain why the search strategy is so broad (in terms of time of publication) (one publication is published over 19 years ago and seems a bit old).

Authors' response

The sentence in lines 182 to 185 now indicates that the search strategy is broad to capture any foundational study. The sentence now reads, "A university librarian with expertise in systematic reviews helped to develop the search strategy for seven databases: CINAHL, Cochrane Library, EMBASE, MEDLINE, PsycINFO, Social Work Abstracts, and Web of Science from their inception (to capture any foundational study) through to 14 June 2019"

Reviewer comment

- Methods, lines 230 – 232: Please clarify the step "renaming the codes as themes". What is the exact difference between codes and themes? Maybe you can give an example.

Authors' response

Renaming codes to themes involved changing from the terms use in the articles to terms that align with concept that fits with the objective of our study.

Lines 235 to 238 now reads, "CBH met one-on-one with HM, ML, and MD to discuss and finalize their codes. They combined and renamed codes as themes by changing terms from the articles to align with concepts that fit the study objective and redistributing some excerpts to more appropriate themes."

Reviewer comment

- Results (in general): I would suggest to add a brief description of what the following categories and themes are about. E.g., After "Selection themes: Structure comprises the settings or context within which engagement occur, such as organisational structure, materials and human resources".

Authors' response

In line with your suggestion, we have added a brief description for each of the categories in the results section. Please see lines 276-277, 338, and 382.

Reviewer comment

- Results (in general): Please check citation again, e.g. citation in line 277 differs from citation in line 279.

Authors' response

I can confirm that they are different and correspond to the two cited studies provided as examples for "best practices, models, and frameworks to use in an engagement plan". Please see lines 285 to 291.

Reviewer comment

- Discussion: Please discuss the involvement and influence of the patient partners on your results.

Authors' response

In the methods section, at the end of the paragraph for the Patient and Public Involvement section, to lines 157 to 159 we have added, "For example, they influenced the results by suggesting names and descriptions for themes, ways to combine themes, and identifying a missing theme."

Reviewer comment

- Discussion & conclusion: Although you selected studies only from upper middle and high-income countries, however, healthcare systems differ greatly. Please discuss if and what impact the differences can have on the identified themes and your conclusion.

Authors' response

We have added to lines 490-492 of the discussion section, "By limiting included studies to those about upper middle and high income countries, our findings need to be confirmed for generalizability to low and middle income countries."

VERSION 2 – REVIEW

REVIEWER	Kirk, Andrew University of Saskatchewan, Medicine
REVIEW RETURNED	24-Aug-2021
GENERAL COMMENTS	I feel that the authors have addressed the reviewers' concerns.
REVIEWER	van Veenendaal , Haske Erasmus University Rotterdam, ESHPM
REVIEW RETURNED	05-Sep-2021
GENERAL COMMENTS	The authors have adressed the comments of the reviewers adequately. I have no further comments.
REVIEWER	Heier, Lina University Hospital Bonn, Institute for Patient Safety
REVIEW RETURNED	01-Sep-2021
GENERAL COMMENTS	Dear authors Thank you very much for addressing all of my comments and revising of your manuscript.