

PEER REVIEW HISTORY

BMJ Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form (<http://bmjopen.bmj.com/site/about/resources/checklist.pdf>) and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Patients, Clinicians and Researchers Working Together to Improve Cardiovascular Health: a Qualitative study of Barriers and Priorities for Patient-oriented Research
AUTHORS	Santana, Maria-Jose; Zelinsky, Sandra; Ahmed, Sadia; Doktorchik, Chelsea; James, Matthew; Wilton, Stephen; Quan, Hude; Fernandez, Nicolas; Anderson, Todd; Butalia, Sonia

VERSION 1 – REVIEW

REVIEWER	Bev Holmes Michael Smith Foundation
REVIEW RETURNED	07-Jul-2019

GENERAL COMMENTS	<p>Thank you for the opportunity to review this paper, which deals with the increasingly important topic of patient oriented research. It sounds like the workshop was very productive in terms of setting priorities for cardiovascular research, and – critically – identifying barriers and facilitators to its successful conduct. In order for the paper to contribute to the field and the literature though, I believe there are some things to be addressed, per below:</p> <ul style="list-style-type: none"> - I like the fact that you have addressed not only priorities in your workshop, but barriers and facilitators. However I had a hard time understanding how they fit together in terms of the design and methods of the study. For example when you describe the design you say “this study used a participatory health approach to work with patients in eliciting their priorities.” Were the facilitators and barriers just an add-on? You also note that the goal of the study was to identify priorities, and that the study focused on “capacity building,” which has a large literature behind it also and would need to be unpacked with a view to where do barriers and facilitators fit in – and how those barriers and facilitators map onto those in the growing literature on various traditions of patient-engaged literature. What might help to tie this together – and I think it’s important anyway – to tell the reader what the patients understood the study to be about. You say the patient partner “discussed with potential participants their expectations...” – how and what came of that? - On the above-mentioned “participatory health research approach,” it is referenced a number of times and also so is participatory action research at one point (page 3, in the abstract). The latter is a decades-old practice with very specific goals and methodology and that’s not what you have done here so that would be a mistaken reference, but I’m not sure what a participatory health research approach is. At one point you reference three papers that are meant to be related but they’re
-------------------------	--

	<p>not. Unless this is an established tradition I would remove these references. It feels important though because you have set it up as the main approach in your theoretical framework on page 6.</p> <p>- Again on methodology, there is more information needed on the design of your study. There are many methodological approaches to priority setting; you describe yours as brainstorming to get to top priorities and eventually a “chosen priority” (page 8). It would be important to know how this (and the identification of barriers and facilitators) was facilitated and what were the dynamics. It sounds like you had a hugely successful day and people could learn from you about how to overcome the power imbalances that are inherent when getting patients, clinicians and researchers together. Importantly, what were the criteria used for both coming up with and ranking the priorities? How did you get away from “this is my priority and I’m voting for it”?</p> <p>- The other methodology piece that’s missing for me is your analytic approach. There is just a quick reference on page 9 that “the notes were collectively analyzed by themes.” What analytic tradition did you use and how did the themes ‘arise’ and who “arose” them? (when you say “comment made during the discussions were linked to the name of any individual” do you mean they were NOT linked?).</p> <p>- Why did patients receive a small honorarium and what drove that decision – that’s a hot topic and no doubt you have something to add here!</p> <p>- You are making an important point – and an important foundation for your study – when you note that “the experiences and outcomes from the patients’ perspective with cardiovascular disease remain to be optimized.” We would need to see references to back this claim up.</p> <p>- And on the note of references, there is a large and growing literature on patient engagement in research that would need to be drawn on throughout the paper. You note that your paper contributes to the current knowledge on POR (page 14) but you could reference more of it and demonstrate how.</p> <p>- Mistake in the name of the SPOR unit on page 6 – they are SUPPORT Units (an acronym), not SPOR Units.</p> <p>Again, thank you for this opportunity to read the paper and for all your efforts. I agree that there is an opportunity here to “gain an understanding of what matters to patients and to learn how patients’ priorities align with clinician-researchers in cardiovascular care [to] result in research and outcomes that are more relevant to both patients and clinicians.” We would just need to know and understand more about how you did this important work to benefit from it as a research community.</p>
REVIEWER	Davina Banner University of Northern British Columbia, Canada
REVIEW RETURNED	10-Jul-2019
GENERAL COMMENTS	Thank you for submitting this enjoyable and interesting manuscript.

	<p>The research topic is timely and the findings will add to a growing body of literature on patient-oriented research. The content is very specific to the Canadian patient-oriented research movement, for this reason, the manuscript will require some revisions to fit with the mandate of this journal and its international readership. For example, the UK has a rich tradition of patient and public involvement (INVOLVE), as well as the US (PCORI). Providing more context to patient-oriented research in Canada and how this aligns or conflicts with other international efforts on patient engagement in healthcare and research is necessary. For this reason, I would recommend a resubmission.</p> <p>Abstract: The abstract captures the key study processes and outcomes, however, this would benefit from further editing.</p> <p>Introduction: The potential contributions of patient-oriented research are clearly stated but greater attention to the international context of patient engagement (e.g. patient and public involvement) would have enhanced this section and would have aligned the content more clearly to the mandate of the journal and its readership. The authors provide a brief overview of the significance of cardiovascular disease. This section could have been enhanced through the inclusion of relevant epidemiological data. Furthermore, the authors refer to new therapies for the prevention and management of CVD (Pg 4 Line 22-24). Some examples would have enhanced this further. The background section lacked detailed and could have provided a more comprehensive overview of the importance of patient-oriented research/patient engagement in CVD care and research.</p> <p>Methods: The research approaches/methods are well suited the research goals, however, the theoretical foundations of the participatory approach and descriptive methods used could have been explained and justified further. For example, greater clarity on how the participatory health research approach/paradigm informed the work would be useful. In addition, the CIHR framework is used to inform the process, however, there are some inconsistencies in how this is referred to in the manuscript and greater clarity on how the framework informed the work would be helpful. The workshop methods were described in great detail and included the use of innovative and inclusive engagement techniques. It is unclear how the train-the-trainer content (Pg 5 Line 18-20) fits with the workshop.</p> <p>Recruitment: Good attention to sex and gender was demonstrated in the work, as well as a desire to engage a diverse range of participants in this research. Recruitment was undertaken using flyers in the clinical settings and SUPPORT Unit, however, it is unclear how a maximum variation sampling approach was undertaken. For example, were potential participants screened and recruited based on the stated demographic characteristics or was a convenience sampling approach used? Compensation of patient and family members was provided and a patient partner/leader co-led the work - excellent!</p> <p>Data analysis: The approaches used to analyze the data were not clear. Approaches to rigor were mentioned briefly (e.g. member checking) but this needs to be addressed in greater detail. Finally,</p>
--	---

	<p>ethical considerations are not addressed. For example, participants were video taped and detailed field notes were compiled. Was ethical approval obtained and did the participants provided their informed consent? Please clarify.</p> <p>Results: The study population was defined clearly. The barriers and facilitators of patient-oriented research were clearly delineated in the table. Clear priorities for CVD care were also identified and these were supported by a supplementary table and quotes (Appendix One). The findings are important and relevant to those engaged in patient-oriented research. A more streamlined and detailed description of the findings would have been beneficial and would have improved the flow of this section.</p> <p>Discussion: The early part of the discussion section was quite repetitive with the earlier sections and a greater focus on how these findings could extend existing knowledge or address critical gaps is needed. For example, the authors highlight some similarities and differences to other studies (e.g. 'the kidney study') but these discussions could have been more detailed and critical. The creation of a patient advisory group is a really great outcome of this work and will no doubt positively influence future planned research and healthcare initiatives. Overall, this is an important study that addresses current gaps. Further development of the international foundations of patient-oriented research, along with greater precision in the presentation of the methods and findings, would really strengthen this paper.</p>
--	--

REVIEWER	Axel Wolf Health and Care Sciences, Sahlgrenska academy
REVIEW RETURNED	17-Jul-2019

GENERAL COMMENTS	<p>Author Santana, Maria-Jose</p> <p>Thank you for inviting me to review this article that covers an important area for the future of healthcare service and research. The authors present research that aims to explore how patients and clinician-researchers can become involved in bidirectional patient-oriented research teams. In total, 23 participants meet during a two days workshop to create patient-oriented research areas of interested and also to discuss barriers toward patient participation in research.</p> <p>Overall, the article is easy to read and describes an innovative design toward patient involvement in research design. However, the article needs to be more transparent and hence make some major revisions regarding some critical aspects of its methodology.</p> <p>1: My first point wants to highlight that it would be good to describe what patient-oriented research is and how it differs from person-centred care. In particular, because the study aimed to explore the bidirectional aspects of co-creation. In person-centred care, the focus lies in the co-creation and discussion of goals. I do not understand the co-creation aspect of the study. What happened if the patient, clinicians or researchers weren't agreeing. Who defined the main topics/categories?</p> <p>2: Selection of participants: If I understand correctly, patients volunteered to participate, so I would like to know how the research team selected the patient, and if patients declined (if so</p>
-------------------------	---

	<p>how many) to participate. I also would like to see more demographic data on age (mean, median, etc.), social status, area of living etc. The same would go for the clinicians and researchers regarding the type of speciality if the clinicians were researchers also etc. To my understanding, the study invited around 20 clinicians, yet only seven participated. How come? Due to time management issues? Please clarify.</p> <p>3: The workshop and the five steps: While the steps and figure 1 help me to understand the process, I struggle to understand the data analysis of these steps:</p> <p>3a: Discussion of the barriers and facilitators as well as priorities: If I understand correctly, the authors used some sort of content analysis to group quotes and discussion into broader categories such as barriers, priorities (App 1), yet I don't understand who performed this and when. Did the different groups do this or the authors of the study? Did all participant discuss all content: for example, in table 1 the authors state that all participants discussed the barriers and solutions, yet I do not understand how this could be possible when you had five groups. Please clarify.</p> <p>ON page 9, line 14, you state the :</p> <p>"The notes were collectively analysed by themes to identify the top areas of prioritisation (SA, SZ, MS). Additionally, a summarised version of the top priorities was presented to the participants for members checking. We then reviewed the notes and flipcharts to ensure that no priorities were missed." If I understand this sentence correctly, you analysed and made the labelling, yet I still don't understand when you did this during the two days? Wasn't it hard to find patterns from the quotes and discussion and to form priorities during the short amount of time? Was there any disagreement about how to label the priorities and research questions?</p> <p>3b: Step 3 and 4: If I understand it correctly, you divided the participants into either patient groups or clinicians-researcher groups do to become more comfortable. What happened topics or questions where the participants disagreed? Or did you not perceive any disagreements between the participants. If not, maybe the lack of disagreement and discussion could be discussed more in the paper? For me, it would be interesting to get more feeling about the "climate" and sense of ease in the room concerning the level of comfort to talk, if all participants were active in the discussion etc.</p> <p>In particular, during step 4, I would like to know more about how congruent the topics were and the discussions around the topics, both regarding Figure 1 text about "is it doable" and the value discussion concerning experience, outcomes, cost etc.</p> <p>3c: Step 5: This step is for me still hard to understand, especially looking at figure 1. I lack the total amount of priorities set by all groups, how many were left after merging, and also the scoring of the priorities. The ten priorities are also in somewhat very broad (for example communication) in regards to the quotes and also the research questions. Hence it would be interesting to get more info regarding the creation and discussion of the priorities.</p> <p>In figure 1, the boxes (to be discussed, matched and merged) would need more clarification, as well as what is meant by top 5 priorities by patient and clinicians.</p>
--	---

	4: In the final part of the study, did the patient receive a chance to discuss the research questions that came out of the priorities? If not, this should be discussed in the limitations.
REVIEWER	Giovanni Biglino University of Bristol School of Clinical Science, Bristol Heart Institute
REVIEW RETURNED	28-Jul-2019
GENERAL COMMENTS	<p>Thank you for the opportunity to review this article, which discussed a 2-day workshop involving patients, clinicians and clinical researchers to identify priorities for patient-oriented research. This is a qualitative study, discussing the findings from the workshops (and the structure of the workshops themselves, which is of interest) and listing the top ten priorities identified at the end of the process.</p> <p>I also commend the authors for the PPIE approach and overall ethos.</p> <p>My major (constructive) criticism is that the novelty of the findings does not fully emerge in my opinion, or is not sufficiently highlighted. We are (or should be!) aware of the importance of communication (and communication facilitated by means of novel technologies), paying attention and honouring patient experience, engaging and involving patients in research etc So I suppose the what is really needed is the identification of HOW TO IMPLEMENT these priorities in practice, rather than identifying the priorities in themselves. This is obviously a huge question, but I wonder if there is more material from the workshop that could inform a discussion around implementation. On top of the theoretical vision for POR, I think the article would be of interest if it also suggested practical tips for how to implement a vision of POR, e.g. how to use the e-technologies? when and how to create a patient advisory group? I agree with the authors ("there is no one formula to conduct POR"). On top of the workshop methodology (which is well described) it would be also interesting to discuss if the priorities identifying with cardiovascular patients and clinicians are aligned (or not) to those identified in cancer, hypertension, DBM etc in other words, are there some overarching priorities and strategies? And what is specific to cardiovascular patients that would be beneficial to implement? The authors allude to some of these concepts (e.g. "participants began to plan future steps etc") and in my opinion these could be given more prominence and/or be discussed more in detail, as they represent potentially novel observations.</p> <p>I also felt that the theoretical framework section repeated concepts from the introduction</p> <p>Minor points:</p> <ul style="list-style-type: none"> - Abstract should mention more details (workshop) in the Setting - rare heart disease: how many participants had a rare condition? (CHD technically is not)

Reviewer 1

1. I like the fact that you have addressed not only priorities in your workshop, but barriers and facilitators. However I had a hard time understanding how they fit together in terms of the design and methods of the study. For example when you describe the design you say “this study used a participatory health approach to work with patients in eliciting their priorities.” Were the facilitators and barriers just an add-on? You also note that the goal of the study was to identify priorities, and that the study focused on “capacity building,” which has a large literature behind it also and would need to be unpacked with a view to where do barriers and facilitators fit in – and how those barriers and facilitators map onto those in the growing literature on various traditions of patient-engaged literature. What might help to tie this together – and I think it’s important anyway – to tell the reader what the patients understood the study to be about. You say the patient-partner “discussed with potential participants their expectations...” – how and what came of that?

Thanks for your questions, undoubtedly helped us to unpack the process. The identification of barriers and facilitators were not an add-on, on the contrary, through discussions with the patient-partner we decided that in order to break power differentials and start building capacity, gaining understanding on patient-oriented research and building partnership - how working together, we needed to start the workshop identifying potential barriers to working together. This activity was very productive and built partnership among participants.

We have added clarification around this point under the step 2 of the process section of the manuscript.

The patient-partner lead, SZ, called the interested participants to discuss the study, address consent and expectations related to time commitment.

2. On the above-mentioned “participatory health research approach,” it is referenced a number of times and also so is participatory action research at one point (page 3, in the abstract). The latter is a decades-old practice with very specific goals and methodology and that’s not what you have done here so that would be a mistaken reference, but I’m not sure what a participatory health research approach is. At one point you reference three papers that are meant to be related but they’re not. Unless this is an established tradition I would remove these references. It feels important though because you have set it up as the main approach in your theoretical framework on page 6.

Indeed, this is an important point, thanks for asking clarification. We refer to participatory health research as the paradigm that guides the research process. This is relevant to our study because participation means that people are involved in health research to improve the quality of the research.

We have reviewed the terminology, modified the methods section and addressed the references. See page 5, study context.

3. Again on methodology, there is more information needed on the design of your study. There are many methodological approaches to priority setting; you describe yours as brainstorming to get to top priorities and eventually a “chosen priority” (page 8). It would be important to know how this (and the identification of barriers and facilitators) was facilitated and what were the dynamics. It sounds like you had a hugely successful day and people could learn from you about how to overcome the power imbalances that are inherent when getting patients, clinicians and researchers together. Importantly, what were the criteria used for both coming up with and ranking the priorities? How did you get away from “this is my priority and I’m voting for it”?

In both cases, while addressing barriers and facilitators as well as priorities, the groups were separated (patients and clinician-researchers) to work on these tasks to later after their presentations to bring them together to work through consensus and ranking. During the workshop we didn't encounter any one who strongly felt like “this is my priority and I’m voting for it” a series of tools were used to avoid this. First, we set up the stage of the workshop with some rules about respect. We all reviewed these rules together. We focused on building partnership from the get go of the workshop, several strategies were used, including the following: a) during the table discussions we had volunteers to guide the discussions and another to present the results of their discussions to the rest of the group; b) involving all participants throughout the process by working together on the data from their flipcharts; c) the exercise of working together to defend their chosen priority using a ‘Dragon’s Den’ style pitch was a very engaging way to bring participants together and build partnership.

On the other point raised, you mentioned other priorities setting approaches, we have explain and contrasted these with dotmocracy. We have modified the content addressing your questions in the methods and discussion sections.

4. The other methodology piece that’s missing for me is your analytic approach. There is just a quick reference on page 9 that “the notes were collectively analyzed by themes.” What analytic tradition did you use and how did the themes ‘arise’ and who “arose” them? (when you say “comment made during the discussions were linked to the name of any individual” do you mean they were NOT linked?).

We have modified this accordingly to your comment. Please see Data collection and analysis sections.

5. Why did patients receive a small honorarium and what drove that decision – that’s a hot topic and no doubt you have something to add here!

During the initial invitation to patients, the patient-partner consulted the honoraria with them. As you mentioned, it’s a hot item, and an interesting one to study. Recently, CIHR has come up with some general guidelines, and also our provincial SPOR unit has developed guidelines for honoraria to guide decisions at provincial level. Recently, also, we have developed a sustainability plan for the advisory council established from this study and they get remunerated for their work.

6. You are making an important point – and an important foundation for your study – when you note that “the experiences and outcomes from the patients’ perspective with cardiovascular disease remain to be optimized.” We would need to see references to back this claim up.

Indeed, thanks for your suggestion. References have been added throughout the manuscript. For instance, we have included a paper from the European Society of Cardiology, describing the importance on patient engagement, see <https://www.escardio.org/The-ESC/What-we-do/esc-patient-engagement>

The other paper from the American Heart Association - Mark McClellan et al 2019, published in the Cardiology Journal, describes the challenges in cardiovascular disease. The authors worked with an advisory group of national health leaders (including patients, clinicians, payers, government officials, and manufacturers) to learn diverse perspectives on increasing access to and improving affordability to treatment for patients with cardiovascular health issues.

<https://www.ahajournals.org/doi/pdf/10.1161/CIR.0000000000000652>

7. And on the note of references, there is a large and growing literature on patient engagement in research that would need to be drawn on throughout the paper. You note that your paper contributes to the current knowledge on POR (page 14) but you could reference more of it and demonstrate how.

We have reviewed the references and added a few ones related to priority setting specifically, Manafò E, Petermann L, Vandall-Walker V, Mason-Lai P (2018) Patient and public engagement in priority setting: A systematic rapid review of the literature. PLoS ONE 13(3): e0193579.

<https://doi.org/10.1371/journal.pone.0193579>

8. Mistake in the name of the SPOR unit on page 6 – they are SUPPORT Units (an acronym), not SPOR Units.

We have address the minor modification on page 5.

Again, thank you for this opportunity to read the paper and for all your efforts. I agree that there is an opportunity here to “gain an understanding of what matters to patients and to learn how patients’ priorities align with clinician-researchers in cardiovascular care [to] result in research and outcomes that are more relevant to both patients and clinicians.” We would just need to know and understand more about how you did this important work to benefit from it as a research community.

We really appreciate your constructive reviews and hope that the modifications addressed your comments.

Reviewer 2

1. Abstract: The abstract captures the key study processes and outcomes, however, this would benefit from further editing.

Thanks for pointing this out. We have modified the text in the abstract following your advice.

2. Introduction: The potential contributions of patient-oriented research are clearly stated but greater attention to the international context of patient engagement (e.g. patient and public involvement) would have enhanced this section and would have aligned the content more clearly to the mandate of the journal and its readership. The authors provide a brief overview of the significance of cardiovascular disease. This section could have been enhanced through the inclusion of relevant epidemiological data. Furthermore, the authors refer to new therapies for the prevention and management of CVD (Pg 4 Line 22-24). Some examples would have enhanced this further. The

background section lacked detailed and could have provided a more comprehensive overview of the importance of patient-oriented research/patient engagement in CVD care and research.

Thanks for highlighting the lack of international context. We have review this and added a paragraph to the introduction (please see paragraph 2 in the introduction section).

Due to word limit, we have briefly addressed the additional information related to the relevance of patient engagement in CV health research and epidemiological data.

3. Methods: The research approaches/methods are well suited the research goals, however, the theoretical foundations of the participatory approach and descriptive methods used could have been explained and justified further. For example, greater clarity on how the participatory health research approach/paradigm informed the work would be useful. In addition, the CIHR framework is used to inform the process, however, there are some inconsistencies in how this is referred to in the manuscript and greater clarity on how the framework informed the work would be helpful. The workshop methods were described in great detail and included the use of innovative and inclusive engagement techniques. It is unclear how the train-the-trainer content (Pg 5 Line 18-20) fits with the workshop.

We have followed your suggestion and elaborated on participatory health research (see page 5, second paragraph) and addressed your comments related to the CIHR framework (page 5 last paragraph) and revised the description of train-the-trainer in page 6, first paragraph.

4. Recruitment: Good attention to sex and gender was demonstrated in the work, as well as a desire to engage a diverse range of participants in this research. Recruitment was undertaken using flyers in the clinical settings and SUPPORT Unit, however, it is unclear how a maximum variation sampling approach was undertaken. For example, were potential participants screened and recruited based on the stated demographic characteristics or was a convenience sampling approach used?

Compensation of patient and family members was provided and a patient partner/leader co-led the

work - excellent!

Thanks for your encouraging comments about our recruitment strategy. We strived to reach maximum variation, to do so, the research partner (SZ) called all the interested participants to briefly described the study, discuss expectations related to time commitment and honorarium. The selected participants were then invited to the workshop.

5. Data analysis: The approaches used to analyze the data were not clear. Approaches to rigor were mentioned briefly (e.g. member checking) but this needs to be addressed in greater detail. Finally, ethical considerations are not addressed. For example, participants were video taped and detailed field notes were compiled. Was ethical approval obtained and did the participants provided their informed consent? Please clarify.

We have clarified the data collection and analysis sections on page 10, first and second paragraphs. Permission was obtained from participants to video-taped their pitch presentations. Please see addition on page 6, last paragraph under sampling and participants.

6. Results: The study population was defined clearly. The barriers and facilitators of patient-oriented research were clearly delineated in the table. Clear priorities for CVD care were also identified and these were supported by a supplementary table and quotes (Appendix One). The findings are important and relevant to those engaged in patient-oriented research. A more streamlined and detailed description of the findings would have been beneficial and would have improved the flow of this section.

As the reviewer noted, due to word limitation, the findings are supported by the use of tables and appendix. We have review this section following your advice to improve the flow.

7. Discussion: The early part of the discussion section was quite repetitive with the earlier sections and a greater focus on how these findings could extend existing knowledge or address critical gaps is

needed. For example, the authors highlight some similarities and differences to other studies (e.g. 'the kidney study') but these discussions could have been more detailed and critical. The creation of a patient advisory group is a really great outcome of this work and will no doubt positively influence future planned research and healthcare initiatives. Overall, this is an important study that addresses current gaps. Further development of the international foundations of patient-oriented research, along with greater precision in the presentation of the methods and findings, would really strengthen this paper.

We have reviewed the discussion section and thank you for your constructive comments and helping us to improve the manuscript.

Reviewer 3

1. My first point wants to highlight that it would be good to describe what patient-oriented research is and how it differs from person-centred care. In particular, because the study aimed to explore the bidirectional aspects of co-creation. In person-centred care, the focus lies in the co-creation and discussion of goals. I do not understand the co-creation aspect of the study. What happened if the patient, clinicians or researchers weren't agreeing. Who defined the main topics/categories?

We have added a description on patient-oriented research in relation to the international initiatives including UK, USA and Canada. Patient-oriented research is included under the umbrella of person-centred care, focusing on engaging patients and public in health research that will ultimately inform improvement in health outcomes that are important to patients.

In order to deal with disagreement, the workshop started with a set of rules about respect, we focused on building partnership from the start of the workshop by working together identifying barriers and solution to work together. This collaborative approach was used throughout the two-day workshop, and conflict was not an issue as the priorities were identified in-group. The use of flip charts and

working through the data together strengthened the partnership. If disagreement had occurred, discussions continued until they was consensus on the priorities.

2: Selection of participants: If I understand correctly, patients volunteered to participate, so I would like to know how the research team selected the patient, and if patients declined (if so how many) to participate. I also would like to see more demographic data on age (mean, median, etc.), social status, area of living etc. The same would go for the clinicians and researchers regarding the type of speciality if the clinicians were researchers also etc. To my understanding, the study invited around 20 clinicians, yet only seven participated. How come? Due to time management issues? Please clarify.

Because this study focused on having patients as partners, we didn't ask for demographic details apart from the data reported. The clinician-researchers were invited to participate and due to their clinical work some of them couldn't make it to the workshop.

3: The workshop and the five steps: While the steps and figure 1 help me to understand the process, I struggle to understand the data analysis of these steps:

3a: Discussion of the barriers and facilitators as well as priorities: If I understand correctly, the authors used some sort of content analysis to group quotes and discussion into broader categories such as barriers, priorities (App 1), yet I don't understand who performed this and when. Did the different groups do this or the authors of the study? Did all participant discuss all content: for example, in table 1 the authors state that all participants discussed the barriers and solutions, yet I do not understand how this could be possible when you had five groups. Please clarify.

I still don't understand when you did this during the two days? Wasn't it hard to find patterns from the quotes and discussion and to form priorities during the short amount of time? Was there any disagreement about how to label the priorities and research questions?

Thank you for your comment. We have reviewed this section and added details about the process. We followed a participatory approach where we worked together to collect and analysed the data to identify first barriers and facilitators to then identify priorities.

To do so we grouped the patients and clinician-researchers in separate groups. At each table they had note pads and flip charts to collect their barriers and facilitators. Each table selected a chairperson to collect and another to present the data back to the larger group. Another individual annotated the information on a larger flip chart stuck to the wall and visible to the entire group. Then the entire group worked together to streamline the data, for instance grouping identical themes and merging similar ones. At the end of this exercise, the presented barriers and facilitators were identified. A similar process occurred with the priorities.

3b: Step 3 and 4: If I understand it correctly, you divided the participants into either patient groups or clinicians-researcher groups to become more comfortable. What happened topics or questions where the participants disagreed? Or did you not perceive any disagreements between the participants. If not, maybe the lack of disagreement and discussion could be discussed more in the paper? For me, it would be interesting to get more feeling about the "climate" and sense of ease in the room concerning the level of comfort to talk, if all participants were active in the discussion etc.

We have added brief description about this important point that you have highlighted in the methods section.

By participants working in groups, disagreements were sorted at each table during the discussions. During the presentation period to the larger group and discussions as a whole team, not disagreements were encountered. Discussions were focused on identifying the key priorities and merging the similar ones. For instance, two priorities (one from each group) were merged into one – the measurement of patient experience (patients) vs patient-reported experiences and outcome measures (clinician-researchers). We have added a modification to figure 2 describing this point.

In particular, during step 4, I would like to know more about how congruent the topics were and the discussions around the topics, both regarding Figure 1 text about "is it doable" and the value discussion concerning experience, outcomes, cost etc.

We appreciate your interest in knowing more details about the workshop. Given the word limit we are not able to provide the details. We are working on a dissemination strategy that includes the production of videos with all participants to unveil details of this process.

3c: Step 5: This step is for me still hard to understand, especially looking at figure 1. I lack the total amount of priorities set by all groups, how many were left after merging, and also the scoring of the priorities. The ten priorities are also in somewhat very broad (for example communication) in regards to the quotes and also the research questions. Hence it would be interesting to get more info regarding the creation and discussion of the priorities. In figure 1, the boxes (to be discussed, matched and merged) would need more clarification, as well as what is meant by top 5 priorities by patient and clinicians.

Thanks for this comment; we have addressed it in the results section by adding the following paragraph:

As depicted in Figure 1, participants worked independently in groups by patients and clinician-researchers to identify their top 10 priorities. Then all of these priorities were discussed and classified into three groups (displayed in Figure 2), including 5 priorities from patients, 5 common priorities to both groups (matched) and 3 priorities unique to clinician-researchers. Then out of the other 8 priorities, three were merged. Merged priorities included: patient experiences (from patients) merged with patient-reported experiences and outcomes (from clinician-researchers) and three priorities (tool to support decision-making, technology to predict and prevent cardiovascular disease, and patient and caregivers' perceptions on the role of technology in patient engagement) merged into priority 4 – e-health technology.

4: In the final part of the study, did the patient receive a chance to discuss the research questions that

came out of the priorities? If not, this should be discussed in the limitations. The patients have discussed the questions and at the moment they are working in research proposal to address some of these priorities. The patient advisory council is active and recently we managed to secure funding from the Libin Institute to ensure sustainability for the next three years.

We appreciate your thoughtful comments and thank you for making our manuscript better.

VERSION 2 – REVIEW

REVIEWER	Davina Banner University of Northern British Columbia, Canada
REVIEW RETURNED	22-Oct-2019

GENERAL COMMENTS	Thank you for submitting this revised manuscript. Based on my review of the article and the author response, I am satisfied that all of the major theoretical and methodological concerns have been addressed and the revised version has greater clarity and precision as a result.
-------------------------	--

REVIEWER	Giovanni Biglino Bristol Medical School, Bristol, UK
REVIEW RETURNED	08-Nov-2019

GENERAL COMMENTS	Overall the manuscript has improved. Just to note: - Good to mention INVOLVE in the UK, just note involve.org.uk is a leading participation charity, but also INVOLVE can refer to invo.org.uk, which is part of, and funded by, the National Institute for Health Research, to support active public involvement in the National Health Service (NHS), public health and social care research. It is one of the few government funded programmes of its kind in the world. Maybe mention both? - I would still include a mention about implementation, e.g. will a participatory approach also be adopted to discuss implementation and not just identification of the priorities? The article is very good in detailing the methodology that was used, which is of interest, just the priorities in themselves are not new. Perhaps the first step is indeed the creation of the patient advisory council mentioned in the conclusion and just a note could be added on exploring the implementation as being the next step.
-------------------------	---

VERSION 2 – AUTHOR RESPONSE

Reviewer 1, Bev Holmes:

4. The other methodology piece that's missing for me is your analytic approach. There is just a quick reference on page 9 that "the notes were collectively analyzed by themes."

What analytic tradition did you use and how did the themes 'arise' and who "arose" them? (when you say "comment made during the discussions were linked to the name of any individual" do you mean they were NOT linked?)

Response: Thank you for this comment. We have clarified our analytic approach in the methods section (page 10, paragraph 3). We took a deductive approach in identifying top research priorities, and two researchers and one patient partner in the research team worked to summarize these priorities, followed by peer debriefing. Additionally, a summarized version of the top priorities was presented to the participants for members checking and further refinement (page 10).

Reviewer 3, Axel Wolf:

3b ...In particular, during step 4, I would like to know more about how congruent the topics were and the discussions around the topics, both regarding Figure 1 text about "is it doable" and the value discussion concerning experience, outcomes, cost etc.

Response: Thank you for this comment. The topics that you referred to are the identified priorities. These quotes presented in Appendix 1 reflect the discussions on the priorities. A criteria including feasibility and 'is it doable' was used by the groups to depict their priority. The process is discussed in step 4 under the methods section.

Editor: We felt that a brief summary of your response to the following comments should be included in the discussion section of your manuscript.

Response: We have addressed these comments in the discussion (see page 16) and now we address each individual review.

Reviewer 3, Axel Wolf:

2: Selection of participants: If I understand correctly, patients volunteered to participate, so I would like to know how the research team selected the patient, and if patients declined (if so how many) to participate. I also would like to see more demographic data on age (mean, median, etc.), social status, area of living etc. The same would go for the clinicians and researchers regarding the type of specialty if the clinicians were researchers also etc. To my understanding, the study invited around 20 clinicians, yet only seven participated. How come? Due to time management issues? Please clarify.

Response: Thank you for this comment and appreciate this viewpoint of reporting standard demographics of patients. Patients recruitment is described on page 11. Only two patients weren't able to make it to the workshop.

In patient engagement as it is a new and evolving area, we viewed patients not as participants per say, rather active patient partners in the co-creation of research priorities. As such we did not collect demographic information on them and similarly did not collect it on clinician-researchers other than the information presented in the manuscript. We acknowledge this as a possible limitation in our study.

The core team of P2 clinician-researchers, nine individuals with diverse specialties, were invited and only two were not able to participate due to previous work commitment. The clinician-research specialties are reported on page 11 in the beginning of the results section.

4: In the final part of the study, did the patient receive a chance to discuss the research questions that came out of the priorities? If not, this should be discussed in the limitations.

Response: Thank you for this comment. The patients have discussed the questions and at the moment they are working in research proposals to address some of these priorities which we have noted on page 13, paragraph 1. The patient advisory council is active and recently we managed to secure funding from the Libin Institute to ensure sustainability for the next three years.

Editor - Please revise the 4th point in your Strengths and Limitations section of your manuscript (after the Abstract). This section should contain five short bullet points, no longer than one sentence each, that relate specifically to the methods.

Response: Thank you for this comment. We have revised the strengths and limitations section of the manuscript (page 3)

Editor - Please complete a thorough proofread of the text and correct any spelling and grammar errors that you identify. e.g. Page 10: "pitches on priorities"

Response: Thank you and we have proofread the text.

Reviewer(s)' Comments to Author:

Reviewer: 2. Reviewer Name: Davina Banner

Institution and Country: University of Northern British Columbia, Canada

Please state any competing interests or state 'None declared': None

Please leave your comments for the authors below

Thank you for submitting this revised manuscript.

Based on my review of the article and the author response, I am satisfied that all of the major theoretical and methodological concerns have been addressed and the revised version has greater clarity and precision as a result.

Response: We would like to thank the reviewer for giving us their feedback and accepting our changes.

Reviewer: 4

Reviewer Name: Giovanni Biglino

Institution and Country: Bristol Medical School, Bristol, UK

Please state any competing interests or state 'None declared': None to declare

Please leave your comments for the authors below

Overall the manuscript has improved.

Just to note:

- Good to mention INVOLVE in the UK, just note involve.org.uk is a leading participation charity, but also INVOLVE can refer to invo.org.uk, which is part of, and funded by, the National Institute for Health Research, to support active public involvement in the National Health Service (NHS), public health and social care research. It is one of the few government funded programmes of its kind in the world. Maybe mention both?

- I would still include a mention about implementation, e.g. will a participatory approach also be adopted to discuss implementation and not just identification of the priorities? The article is very good in detailing the methodology that was used, which is of interest, just the priorities in themselves are not new. Perhaps the first step is indeed the creation of the patient advisory council mentioned in the conclusion and just a note could be added on exploring the implementation as being the next step.

Response: We would like to thank the reviewer for their comments. We have included a sentence about invo.org.uk in the introduction section (page 4). In regards to implementation, the patient partner advisory council is currently working on research proposals, and is involved in a few research projects that address the research priorities. We have included this information in the discussion section (page 16, last paragraph of discussion).