PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	What matters most to patients about primary health care: Mixed methods patient priority setting exercises within the PREFeR (PRioritiEs For Research) Project
AUTHORS	Edwards, Louisa; Monro, Melody; Butterfield, Yaron; Johl, Ravin; Loftsgard, Kent; Pelletier, Hayley; McGavin, Colleen; Lavergne, M.

VERSION 1 - REVIEW

REVIEWER	Barbara J Turner University of Texas Health San Antonio, USA
REVIEW RETURNED	31-Aug-2018

GENERAL COMMENTS	This manuscript describes a timely and important initiative to engage patients in the process of developing priorities for improving primary care. The paper is mostly descriptive about the components of that initiative while reporting the results of a small convenience sample of patients who participated in one aspect of this process. Although this is the “first known” patient priority setting for primary care that is not specific to a disease process, it has relatively limited results to report at this point because 10 patients were recruited to participate in an online nominal group technique approach but only 8 actually participated. Another 2 were interviewed individually which is a fundamentally different approach. Although the results are supposedly novel, most of the topics generated by this small sample of patients prioritized appear to be fairly standard as issues that patients have raised about primary care. Some of these are said to be a surprise and not previously identified by the authors but it is not clear which ones these were. One of the strengths of the approach is that it is structured by the Dialogue Model. According to that model (based on the website cited in Reference 1) the first phase is exploration with an interdisciplinary team. It involves the ideas of patients and other stakeholders. One of the weaknesses of this model is it does not specify adequately the sample needed to gain a relatively representative view of ideas from stakeholders to then build on. Most mixed methods research would not expect to reach saturation of ideas from 8 individuals. Another concern is that, for some reason, patient stakeholders who are recruited needed some previous research experience (page 9 line 13). This is not explained in the document and it have biased the types of patients who participated. Another concern is that the sample is said to be diverse but they are described only as “mixed

	or any other racial-ethnic background”. In terms of demographics, would be more helpful to specifically know whether they are of African origin or Indian origin etc. Because this is an Internet administered process, all of the individuals had to have access to the web and have an email. Not surprisingly, this is a relatively educated group of individuals with most having a BA or higher. It was an interesting approach to use the NGT in an online manner. It would be helpful to cite other studies that use this approach with individuals from the public. (Rice DB, Cañedo-Ayala M, Turner KA, Gumuchian ST, Malcarne VL, Hagedoorn M, Thombs BD; Scleroderma Caregiver Advisory Team. Use of the nominal group technique to identify stakeholder priorities and inform survey development: an example with informal caregivers of people with scleroderma. BMJ Open. 2018 Mar 2;8(3):e019726). The paper describes having done a follow-up survey of patients about the issues raised by this select group of individuals but fails to present these results. The focus on access to the EMR is a bit of a surprise. From their literature review – has this emerged as a top patient priority? Or is it an interest of only this small subset of individuals. It would have been useful for the discussion to be less repetitive of the processes and put the results in context. Another option for this paper is to take the approach of a clinical trial and simply describe the design. Then when more data are available (such as from the patient survey), these results should be published together. As it is, the paper does not achieve either the goal of the description of the approach or the results. Instead of rehashing the introduction and methods, the authors could comment more on other studies of patient health care priorities (non disease specific) such as https://www.pcori.org/sites/default/files/NYU2870-White-Paper.pdf.

REVIEWER	Joanna Crocker Nuffield Department of Primary Care Health Sciences, University of Oxford, United Kingdom
REVIEW RETURNED	04-Sep-2018

GENERAL COMMENTS	This is an interesting and well-written paper reporting a regional research priority setting exercise within primary health care in British Colombia, Canada. To my knowledge it is the first patient-centred priority setting exercise to be conducted within general primary care, rather than focusing on a specific condition or aspect of primary care. It therefore has the potential be very useful to people internationally wishing to set research priorities within similarly broad, non-disease specific areas of care. The authors’ approach was informed the Dialogue Model, which is a sound methodology specifically developed (in the Netherlands) for research agenda setting. However, I have some concerns about the actual methods used, interim reporting and limitations which I think need to be addressed before the paper is published (see my comments listed below). Major comments: 1) I am a little confused as to why the findings of only the first 4 steps of an 8-step prioritisation process (Figure 1) are being reported here. The topics are presented as patient priorities for future research, but in fact they are just interim priorities which will likely change during the subsequent steps. And there is no

information about which ones have already been fully or partially addressed by research. Why not report on the final prioritisation findings, within the context of past and ongoing research? If the authors are able to justify publishing these interim findings, I think the fact they are interim needs to be made explicit in the abstract, and there should be more focus on the methodological findings / lessons learned (for others wanting to carry out similar processes elsewhere) and less on the order of priorities (which is likely to change anyway).

2) Related to the above point, it's not clear to me why the interim prioritisation was conducted at all. It is not (as I understand it) part of the Dialogue Model which the authors claim to be using (Abma & Broerse, 2010). Why not take all 18 topics generated during the first step forward to the subsequent steps, rather than only the top 10 topics (ranked by a small, unrepresentative sample of patients – see my next point)?

3) The first 3 steps in the priority-setting process (including the interim prioritisation) seem to me to be rather flawed, because they involved only 9 people who were originally selected for a broader purpose (a patient advisory group to the British Colombia Primary Health Care Research Network) and who seem not to reflect the diversity of the wider population (they had a high level of education and almost all had prior experience of involvement in research; I'm not convinced by the authors' claim that this was a 'sociodemographically diverse' group of patients). This sampling seems to be counter to the Dialogue Model, which advises that for the Consultation phase (gathering topic ideas), care should be taken to contact and get meaningful input from a diverse patient population and that the Prioritisation phase should be done via questionnaire to ensure wide reach (Abma & Boerse, 2010). I think this modification needs adequate justification, especially because all of the subsequent steps depend on the findings of this one. Why did the authors not start from scratch and choose a more diverse sample of patients according to the specific objective of this project? It seems that much more effort was put into recruiting diverse patients for the later Prioritisation and Integration phases compared to this first (and arguably most crucial) one. This seems to me to be an important limitation which needs to be highlighted in the paper.

4) Related to the above points, my understanding is that there are several quite major deviations from the Dialogue Model, including those the authors report on page 10, the interim prioritisation mentioned above, and the methods used in the Exploration and Consultation phases. Given differences, I don't think the authors can really claim that they are using the Dialogue Model, but rather that the design was informed by the Dialogue Model framework. It would be helpful if all substantial differences were reported and explained. Perhaps the authors could also briefly report on how they did (or did not) adhere to the 'underlying notions' of the Dialogue Model (such as 'good social conditions', 'process facilitation' and 'dialogue between stakeholders') (Abma & Boerse, 2010)?

5) Related to the above points, it would help if the authors could clarify whether (and if so, how) patients recruited for subsequent steps were able to introduce new topics of importance to them. If this was not possible, it is another limitation which needs to be

discussed. Patients from lower educational backgrounds (for example) might have had different ideas to the advisory group members, and it is not clear how these would be captured by the process. Another commonly used research priority setting process, the James Lind Alliance, uses public surveys at the first step to enable as many people as possible to suggest topics and questions, which might be a better way to maximise opportunity for all voices to be heard? Perhaps the authors could briefly compare and contrast their methods to this one? I do think there are some relative strengths of the authors' approach, such as the Integration phase which brings in researchers and policymakers.

6) The objective was to generate topics for future primary care research. However, many of the topics actually generated seem to be about changing health service delivery / policy (e.g. 'No universal drug plan' and 'Unable to find a doctor') rather than conventional research questions or 'neutral' topics. This might be because patients were asked what stood out for them about primary care and there was no research input into the stages presented in this paper. But I think readers would benefit from some discussion about this and the differences/boundaries/overlap between priorities for health service delivery/policy and priorities for health service research. Is the problem an absence of research or a gap between the translation of research into policy and practice? The Discussion (page 25-26) suggests that regional government already knows that many of these topics are important; they just aren't being fully or successfully implemented yet. It is interesting that one of the most obvious research-related topics, 'Knowledge gaps in clinical evidence' was actually ranked very low and did not even make it into the Top 10 – what does this say about what patients want researchers to be doing? Would action research (i.e. research to effect specific change) be a more appropriate fit? On a related note, in the Discussion the authors refer several times to 'priorities for primary health care / primary care' which I think should be 'priorities for health care research' (unless I have misunderstood the objective).

7) The abstract conclusion states 'The project activities to date demonstrate the feasibility of including patients in priority setting exercises for primary health care in general, rather than focusing on a condition-specific population or disease area'. I think this needs to be reworded because there are examples of non condition-specific prioritisation in primary care, for example the 'healthy behaviours' studies cited in the introduction, and a recent James Lind Alliance Priority Setting Partnership which focused on patient safety in primary care (Morris et al. 2018). This paper does take an even broader perspective by focusing on primary care in general, and I think it would be very helpful if the authors could reflect on and discuss the particular experiences and challenges of doing this in the paper (with reference to the range of health conditions / medical history of the included patients), rather than just stating that they have demonstrated feasibility. Do they have any recommendations for others wishing to set priorities within similarly broad areas of care?

8) It is not clear why health care providers were included in the prioritisation and integration phases, when they were excluded from the exploration, consultation and interim prioritisation phases. I think this needs more explanation/justification. Involving any stakeholder group at such a late stage might reduce their level of

engagement and feeling of ownership if they don't feel their views are adequately reflected in the list of topics presented to them. My understanding is that the Dialogue Model explicitly avoids shifting control/power from one stakeholder group to another, instead emphasising that all stakeholders have a unique and relevant perspective and that dialogical exchanges will result in mutual understanding and shared action agendas.

9) Exploration phase – Why did the literature review include only research conducted within Canada? Could research conducted in other countries also be useful and help avoid duplication/waste? (Many of the prioritised topics are concerns in the UK too, for example.) If this can't be changed I think it needs to be explained and/or acknowledged as a limitation. Also, could the authors clarify how the findings of the literature review and horizon scanning survey were used to inform the next steps of the prioritisation process? The authors state that 'A reference library of provincial papers and all citations from the included papers in the two searches was compiled for used by the Patient Advisory' – how did the patients use them, and did they have the necessary skills to understand and interpret academic papers?

Minor comments:

10) The reference to 'Patient Advisory' throughout the paper feels strange/incomplete to me; adding the word 'Board', 'Group' or 'Panel' would make it easier to read.

11) Introduction page 7 – I find the reference to 'specific, structured' frameworks and 'non-specific tools' confusing – what is the meaning of 'specific' here? Perhaps the sentence could be reworded.

12) Page 9 – 'The Patient Advisory were research partners in PREFeR and, therefore, contributed to planning and shaping the research activities' – it is not usually the case that research participants are involved in the research design. Could the authors provide a bit more detail about why and how they were involved in this way (as opposed to involving other patients)? Might their involvement in planning have had any impact on the findings?

13) It would be good to know more about the selection process for recruiting patients to the advisory group. What proportion of those who expressed interest were selected, and how were they chosen?

14) Did the patient advisory group members already know each other or had worked together before this prioritisation exercise? If so, how might this have influenced the results? Is 'group think' a potential limitation?

15) What relevant skills and/or qualifications did the facilitator of the patient discussions have to ensure that all participants were given sufficient opportunity to voice their views?

16) Page 10 – 'We wanted the rapid literature reviews to inform the scope of what has already been researched within BC and Canada' – I'm not sure how the reviews can inform something which has already happened? I think this needs rewording.

17) Page 11 - Data Collection – it would be helpful to report the number of patients who were invited to take part. Also, ‘patients were asked to first briefly describe their primary care experiences’ seems not quite consistent with being asked ‘what stood out for you in primary care?’ – I would be more specific.

18) Page 12 - Priority Setting Workshop – I assume this was a face-to-face meeting but I think this should be made explicit, especially as an earlier step used teleconferencing.

19) Page 13 – Were patients given as long as they needed to rank the priorities, or was there a time limit?

20) Page 13 – I think that creating a sum of scores for each topic was appropriate for the purposes of prioritisation, as was resolving ties by counting the number of votes, but I don’t understand why the ‘relative importance of each topic, or the proportion (%) of scores ranked within the top 10 topics’ was calculated. Unless I have misunderstood, the results in terms of rankings should be identical to the sum of scores (as shown in Table 3), so I’m not sure why this step was necessary? I find the sum of scores easier to interpret.

21) On a related note, what happened to topics not chosen as patients’ top 10? Were they scored ‘0’?

22) Why were the participants in the Prioritisation stage asked to individually rate (rather than rank) the topics, while in the interim prioritisation phase they were ranked not rated? This could do with a brief explanation.

23) Table 1 – Age would be easier to interpret if range rather than (or as well as) standard deviation was reported. Previous research involvement – what does this mean – as a participant and/or in the design etc. of research? What do ‘moderate’ and ‘experienced’ equate to? Also, did the authors collect any information about patients’ health conditions / medical history? Since one of the objectives was to demonstrate the feasibility of involving patients in non-condition specific prioritisation, it would be really helpful to see how diverse a group they were in terms of their health issues / medical history – how might this have influenced the findings?

24) Page 19 – The patients provided ‘82 experiences and examples of what stood out for them in primary care in BC (data not shown)’ – could these be provided as a supplementary file?

25) Table 3 – As mentioned above, I am confused by the inclusion of the ‘relative importance (%)’ and ‘ranked priority (via %)’ columns. The data are identical to the ‘sum of scores’ and ‘ranked priority (via scores)’ columns; I don’t think both are needed?

26) Could the authors discuss any challenges they encountered and lessons learned? This could be very helpful for others wanting to carry out similar prioritisation exercises.

Thank you for the opportunity to review this paper. I hope these comments are helpful and look forward to reading the published version.

References:

	Abma TA, Broerse JE. Patient participation as dialogue: setting research agendas. Health Expect 2010, 13(2):160-173. Morris RL, Stocks SJ, Alam R, Taylor S, Rolfe C, Glover SW, Whitcombe J, Campbell SM. Identifying primary care patient safety research priorities in the UK: a James Lind Alliance Priority Setting Partnership. BMJ Open 2018, 8(2).

VERSION 1 – AUTHOR RESPONSE

Reviewers' Comments to Author:

Reviewer: 1

Reviewer Name: Barbara J Turner

Institution and Country: University of Texas Health San Antonio, USA

Please state any competing interests or state ‘None declared’: none

This manuscript describes a timely and important initiative to engage patients in the process of developing priorities for improving primary care. The paper is mostly descriptive about the components of that initiative while reporting the results of a small convenience sample of patients who participated in one aspect of this process. Although this is the “first known” patient priority setting for primary care that is not specific to a disease process, it has relatively limited results to report at this point because 10 patients were recruited to participate in an online nominal group technique approach but only 8 actually participated. Another 2 were interviewed individually which is a fundamentally different approach. Although the results are supposedly novel, most of the topics generated by this small sample of patients prioritized appear to be fairly standard as issues that patients have raised about primary care. Some of these are said to be a surprise and not previously identified by the authors but it is not clear which ones these were.

Thank you for your very constructive comments. We agree that the results as initially presented were limited. We have, therefore, edited the paper to include results of the subsequent province-wide survey, which included 464 patients and 173 primary care providers.

We have also expanded the discussion about how results compare to previously identified priorities as follows (pages 26-27):

“The 10 priorities originally identified by the Patient Advisory align fairly well with some recent priority setting initiatives in the province. The BC Ministry of Health recently identified eight service delivery priorities for the province’s health system in 2014[28]. These were re-confirmed at the primary care level by the BC-PHCRN research, clinical, and policy leads in 2015[20], but no patients were involved in this process. There is considerable overlap between the previous BC-PHCRN priorities and those reported herein, though there were two uniquely-identified Patient Advisory priorities that were not advanced by either the BC Ministry of Health or BC-PHCRN: Mental health resources (albeit, access to specialist treatment, in general, was mentioned by both organizations, and reducing demand on hospitals by improving care for those with mental health and substance use issues was one of five strategic priorities for the BC Ministry of Health in 2017[29]) and Improve and strengthen patient-provider communication. There were also examples where patient perspectives added depth and context. While both sets of priorities spoke to the need to address challenges specific to rural and remote settings, patients highlighted that transportation needs to be considered, and privacy and stigma may impact those in rural areas. The previous BC-PHCRN priorities included the need to enhance access, utilization, and continuity of patient information with other providers and levels of care, but patients spoke more directly to the need for electronic health records and the ability to access their medical records themselves. Finally, while the previous priorities noted issues around workforce planning, patients framed this as being unable to find a regular doctor or other primary health care provider. This highlights the value of including patients directly in setting priorities for primary care, and is another demonstration of how the priorities of patients and other stakeholders might differ."

One of the strengths of the approach is that it is structured by the Dialogue Model. According to that model (based on the website cited in Reference 1) the first phase is exploration with an interdisciplinary team. It involves the ideas of patients and other stakeholders. One of the weaknesses of this model is it does not specify adequately the sample needed to gain a relatively representative view of ideas from stakeholders to then build on. Most mixed methods research would not expect to reach saturation of ideas from 8 individuals.

In the first phase, patients were not recruited to be representative of the whole province, but rather to achieve maximum diversity with respect to health care needs, geographic context, ethnicity (including First Nations), age, and gender. We appreciate that this was not as clearly explained as it could have been, so we have added clarification about this on page 9. It is likely that we may not have reached saturation within the patient advisory, but many of the top 10 priorities align with other priority setting initiatives (see pages 26-27). Furthermore, our understanding of the Dialogue Model and the Nominal Group Technique is that they are intended to generate a wide variety of ideas, rather than achieve saturation. Nevertheless, we now also report results of a province-wide survey that has much broader input. We thank the reviewer for bringing these important issues to our attention and we have now discussed these among the limitations on page 29.

Another concern is that, for some reason, patient stakeholders who are recruited needed some previous research experience (page 9 line 13). This is not explained in the document and it have biased the types of patients who participated. Another concern is that the sample is said to be diverse but they are described only as “mixed or any other racial-ethnic background”. In terms of demographics, would be more helpful to specifically know whether they are of African origin or Indian origin etc. Because this is an Internet administered process, all of the individuals had to have access to the web and have an email. Not surprisingly, this is a relatively educated group of individuals with most having a BA or higher.

We sought to recruit patients to the Patient Advisory group both with and without previous research experience – we now clarify this on page 9. However, given the channels through which we recruited, in the end, the majority had some degree of research experience.

The Patient Advisory is diverse across a number of socio-demographic variables, rather than ethnicity alone (e.g., age, sex, education levels, health conditions, research experience). Regarding ethnicity, Indigenous people (First Nations and Metis) in British Columbia face health disparities and structural racism within the health care system. Therefore, we now explicitly report demographics for Patient Advisory members who self-identified as Indigenous.

We have expanded discussion of issues of diversity and representation in both the Results and limitations. However, the survey was not limited to those with internet and email access; it could have been completed over the telephone (only 1 respondent did so), and we advertised in local newspapers to try to broaden our reach.

It was an interesting approach to use the NGT in an online manner. It would be helpful to cite other studies that use this approach with individuals from the public. (Rice DB, Cañedo-Ayala M, Turner KA, Gumuchian ST, Malcarne VL, Hagedoorn M, Thombs BD; Scleroderma Caregiver Advisory Team. Use of the nominal group technique to identify stakeholder priorities and inform survey development: an example with informal caregivers of people with scleroderma. BMJ Open. 2018 Mar 2;8(3):e019726).

Thank you for this very helpful reference. We have now included this within the Methods section (page 12).

The paper describes having done a follow-up survey of patients about the issues raised by this select group of individuals but fails to present these results. The focus on access to the EMR is a bit of a surprise. From their literature review – has this emerged as a top patient priority? Or is it an interest of only this small subset of individuals. It would have been useful for the discussion to be less repetitive of the processes and put the results in context. Another option for this paper is to take the approach of a clinical trial and simply describe the design. Then when more data are available (such as from the patient survey), these results should be published together. As it is, the paper does not achieve either the goal of the description of the approach or the results. Instead of rehashing the introduction and methods, the authors could comment more on other studies of patient health care priorities (non disease specific) such as https://www.pcori.org/sites/default/files/NYU2870-White-Paper.pdf.

We welcome this feedback and have now substantially revised the paper to include importance ratings from the survey as well.

Interestingly, in the survey results, the “Information sharing, including electronic medical records” topic was within the top three most highly rated priorities for the group of 464 patients (it ranked sixth in the

173 primary care provider results). We now discuss this, as well as how this corresponds with an existing provincial priority related to health information on page 27.

Finally, we have now also cited the mentioned study within the Introduction. Once again, thank you for drawing our attention to this highly relevant reference.

Reviewer: 2

Reviewer Name: Joanna Crocker

Institution and Country: Nuffield Department of Primary Care Health Sciences, University of Oxford, United Kingdom

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

This is an interesting and well-written paper reporting a regional research priority setting exercise within primary health care in British Colombia, Canada. To my knowledge it is the first patient-centred priority setting exercise to be conducted within general primary care, rather than focusing on a specific condition or aspect of primary care. It therefore has the potential be very useful to people internationally wishing to set research priorities within similarly broad, non-disease specific areas of care. The authors' approach was informed the Dialogue Model, which is a sound methodology specifically developed (in the Netherlands) for research agenda setting. However, I have some concerns about the actual methods used, interim reporting and limitations which I think need to be addressed before the paper is published (see my comments listed below).

Major comments:

1) I am a little confused as to why the findings of only the first 4 steps of an 8-step prioritisation process (Figure 1) are being reported here. The topics are presented as patient priorities for future research, but in fact they are just interim priorities which will likely change during the subsequent steps. And there is no information about which ones have already been fully or partially addressed by research. Why not report on the final prioritisation findings, within the context of past and ongoing research? If the authors are able to justify publishing these interim findings, I think the fact they are interim needs to be made explicit in the abstract, and there should be more focus on the methodological findings / lessons learned (for others wanting to carry out similar processes elsewhere) and less on the order of priorities (which is likely to change anyway).

Thank you for this feedback. We have now included results of the province-wide survey and place these in the context of the published literature. We agree that focusing on the survey results, which indicate the final order of the priorities, are of more significance.

2) Related to the above point, it's not clear to me why the interim prioritisation was conducted at all. It is not (as I understand it) part of the Dialogue Model which the authors claim to be using (Abma

& Broerse, 2010). Why not take all 18 topics generated during the first step forward to the subsequent steps, rather than only the top 10 topics (ranked by a small, unrepresentative sample of patients – see my next point)?

Similar to the methods used by Broerse, Zweekhorst, van Rensen, et al. (2010), we decided in advance that 10 topics would be a manageable number to carry forward for an online survey. We did not know how many topics would be generated by the patient advisory at the outset, but our sense was that the general public would not be willing to rate more than 10 topics, as well as several socio-demographic questions. Therefore, we used the rankings from the patient advisory as the first step in determining a smaller, more manageable list of priorities for patients and primary care providers to rate in terms of importance in a survey. A publication stemming from a health research priority setting workshop ("Mind the gap! Multi-stakeholder dialogue for priority setting in health research"), which Broerse contributed to, also depicted an interim prioritization phase within the Dialogue Model. We appreciate that this could be made more clear in the manuscript, which we have now done on page 13. We also note that the decision to retain 10 was somewhat arbitrary in the limitations listed on page 29.

3) The first 3 steps in the priority-setting process (including the interim prioritisation) seem to me to be rather flawed, because they involved only 9 people who were originally selected for a broader purpose (a patient advisory group to the British Colombia Primary Health Care Research Network) and who seem not to reflect the diversity of the wider population (they had a high level of education and almost all had prior experience of involvement in research; I'm not convinced by the authors' claim that this was a 'sociodemographically diverse' group of patients). This sampling seems to be counter to the Dialogue Model, which advises that for the Consultation phase (gathering topic ideas), care should be taken to contact and get meaningful input from a diverse patient population and that the Prioritisation phase should be done via questionnaire to ensure wide reach (Abma & Boerse, 2010). I think this modification needs adequate justification, especially because all of the subsequent steps depend on the findings of this one. Why did the authors not start from scratch and choose a more diverse sample of patients according to the specific objective of this project? It seems that much more effort was put into recruiting diverse patients for the later Prioritisation and Integration phases compared to this first (and arguably most crucial) one. This seems to me to be an important limitation which needs to be highlighted in the paper.

The Patient Advisory was recruited to maximize diversity with respect to geographic context and health care needs. They were not intended to be a representative sample of the province, but rather to bring diversity in relevant expertise and experience of primary care. The objectives in recruiting the Advisory were, therefore, aligned with the objectives of the research project and, in fact, this was the first project they took on as a group.

We have now added detail on recruitment of the Patient Advisory to the Methods and Results sections (pages 9 and 18, respectively). We also now report results of the province-wide survey. However, survey respondents were also not representative of the province as a whole. We now discuss limitations pertaining to recruitment of both Advisory members and survey respondents on page 29.

4) Related to the above points, my understanding is that there are several quite major deviations from the Dialogue Model, including those the authors report on page 10, the interim prioritisation mentioned above, and the methods used in the Exploration and Consultation phases. Given differences, I don't think the authors can really claim that they are using the Dialogue Model, but rather that the design was informed by the Dialogue Model framework. It would be helpful if all substantial differences were reported and explained. Perhaps the authors could also briefly report on how they did (or did not) adhere to the 'underlying notions' of the Dialogue Model (such as 'good social conditions', 'process facilitation' and 'dialogue between stakeholders') (Abma & Boerse, 2010)?

This is an excellent point. We now better explain our rationale for diverging from the model within the Introduction (page 8):

"The PREFeR Project is informed by the Dialogue Model, which is a multi-stage priority setting framework involving multiple stakeholders, but modified this model to focus solely on patient perspectives for initial topic generation[1]. Here we report the resulting patient-identified priorities and their importance for patients and primary care providers."

We expand on this in the Discussion (pages 27-28).

"That said, the five phases of the Dialogue Model were modified substantially to address the aim of PREFeR, which explicitly focuses on patient priorities. Consistent with Canada's Strategy for Patient Oriented Research, the Patient Advisory members were research partners in PREFeR and contributed to planning and shaping the research activities. Their dual role as research participants and team members means that to some degree we shifted control and power to patients within the process, which is inconsistent with the Dialogue Model. This was implemented in a context where policymakers, clinicians, and researchers had previously worked to identify priorities in the absence of patients. Perspectives from these other stakeholder groups were, however, gathered and compared to patient priorities, albeit later in the project. In the end, these aligned quite closely, though patients identified some additional priorities and added additional context and depth to existing ones."

We have also modified all language as suggested.

5) Related to the above points, it would help if the authors could clarify whether (and if so, how) patients recruited for subsequent steps were able to introduce new topics of importance to them. If this was not possible, it is another limitation which needs to be discussed. Patients from lower educational backgrounds (for example) might have had different ideas to the advisory group members, and it is not clear how these would be captured by the process. Another commonly used research priority setting process, the James Lind Alliance, uses public surveys at the first step to enable as many people as possible to suggest topics and questions, which might be a better way to maximise opportunity for all voices to be heard? Perhaps the authors could briefly compare and contrast their methods to this one? I do think there are some relative strengths of the authors' approach, such as the Integration phase which brings in researchers and policymakers.

Once again, we thank the reviewer for making this point. Immediately following the 10 topics, there was an open-ended textbox with the instruction, “Please use this space if you have any comments you’d like to share.” Few suggested new topics, but of course we did not solicit new topics directly. We have noted this aspect of the survey methods on page 15, but also noted the importance of listing this as a limitation (page 29).

The James Lind Alliance is indeed another excellent priority setting framework. However, it is a lengthy process that seeks to identify research uncertainties, which was not the focus of the current project. We now note this in the Introduction (page 7).

6) The objective was to generate topics for future primary care research. However, many of the topics actually generated seem to be about changing health service delivery / policy (e.g. ‘No universal drug plan’ and ‘Unable to find a doctor’) rather than conventional research questions or ‘neutral’ topics. This might be because patients were asked what stood out for them about primary care and there was no research input into the stages presented in this paper. But I think readers would benefit from some discussion about this and the differences/boundaries/overlap between priorities for health service delivery/policy and priorities for health service research. Is the problem an absence of research or a gap between the translation of research into policy and practice? The Discussion (page 25-26) suggests that regional government already knows that many of these topics are important; they just aren’t being fully or successfully implemented yet. It is interesting that one of the most obvious research-related topics, ‘Knowledge gaps in clinical evidence’ was actually ranked very low and did not even make it into the Top 10 – what does this say about what patients want researchers to be doing? Would action research (i.e. research to effect specific change) be a more appropriate fit? On a related note, in the Discussion the authors refer several times to ‘priorities for primary health care / primary care’ which I think should be ‘priorities for health care <i>research</i>’ (unless I have misunderstood the objective).

As stated within the Introduction (pages 7-8), the aim of PREFeR was not to develop research questions directly. Instead, the project sought to identify patient-generated topics of importance that future research and researchers might develop into more specific questions. Therefore, it is by design that some of the topics are stated in general, overarching or even policy-oriented language. However, we agree that more discussion of this would be beneficial to readers, and so we have expanded on this in the Discussion section. Indeed, some topics have been extensively researched. These topics may, therefore, suggest a need for strengthened knowledge translation or implementation science to inform changes to policies or service delivery (page 28). We have corrected the inconsistencies in language, but do still contend that this is a primary care priority setting project, which could, of course, include more general aspects of health care/service delivery. Once again, we maintain that it is important to consider not just the overarching and more general topic labels, which are necessarily broadly stated, but also the more illustrative bullet points (see Supplementary file 1 and file 2). It is only when considering the two together that the full nature of each topic can be appreciated.

7) The abstract conclusion states ‘The project activities to date demonstrate the feasibility of including patients in priority setting exercises for primary health care in general, rather than focusing on a condition-specific population or disease area’. I think this needs to be reworded because there are examples of non condition-specific prioritisation in primary care, for example the ‘healthy behaviours’

studies cited in the introduction, and a recent James Lind Alliance Priority Setting Partnership which focused on patient safety in primary care (Morris et al. 2018). This paper does take an even broader perspective by focusing on primary care in general, and I think it would be very helpful if the authors could reflect on and discuss the particular experiences and challenges of doing this in the paper (with reference to the range of health conditions / medical history of the included patients), rather than just stating that they have demonstrated feasibility. Do they have any recommendations for others wishing to set priorities within similarly broad areas of care?

We have reworded the Abstract and expanded discussion about the challenges of priority setting within such a broad area on page 28:

“Another notable divergence is that we identified topics for future research, rather than specific research questions. The top-rated topics are broad, and many reflect priorities for health system change that may be informed by research, but are not themselves research questions. Results from rapid literature reviews of the 10 topics, along with results from the ‘horizon scanning’ survey with BC researchers relating to completed, current or upcoming research on the 10 topics, will provide valuable information about the nature and extent of research on the topics. In some cases, the topics may suggest a need for strengthened knowledge translation or implementation science to inform changes to policies or service delivery rather than additional research. Also, as the searches covered broad topics, but were limited to BC and Canada, we recommend that more thorough reviews should be conducted that are specific to any future research objectives.”

8) It is not clear why health care providers were included in the prioritisation and integration phases, when they were excluded from the exploration, consultation and interim prioritisation phases. I think this needs more explanation/justification. Involving any stakeholder group at such a late stage might reduce their level of engagement and feeling of ownership if they don’t feel their views are adequately reflected in the list of topics presented to them. My understanding is that the Dialogue Model explicitly avoids shifting control/power from one stakeholder group to another, instead emphasising that all stakeholders have a unique and relevant perspective and that dialogical exchanges will result in mutual understanding and shared action agendas.

This is an excellent point. We expand on our rationale for focusing on patient priorities within the Introduction, including the context of Canada’s Strategy for Patient Oriented Research, and the fact that priorities had previously been identified through a process that included only policymakers, clinicians, and researchers (page 8). Therefore, in PREFeR, we began with patient-identified priorities, but included perspectives of primary care providers during the survey, as well as through the final dialogue event, which did serve to facilitate dialogue between patients and providers. We now discuss this more fully in the context of the Dialogue Model on pages 27-28.

9) Exploration phase – Why did the literature review include only research conducted within Canada? Could research conducted in other countries also be useful and help avoid duplication/waste? (Many of the prioritised topics are concerns in the UK too, for example.) If this can’t be changed I think it needs to be explained and/or acknowledged as a limitation. Also, could the authors clarify how the

findings of the literature review and horizon scanning survey were used to inform the next steps of the prioritisation process? The authors state that ‘A reference library of provincial papers and all citations from the included papers in the two searches was compiled for used by the Patient Advisory’ – how did the patients use them, and did they have the necessary skills to understand and interpret academic papers?

We had limited resources to conduct the review process and could only feasibly examine Canadian literature for such broad areas of inquiry. This is a limitation of our study, but nevertheless pertinent to identifying future avenues of research within Canada (in line with the aims of PREFeR). We now explain that the reference library informed discussion at the concluding dialogue event between all stakeholders, but we’d expect that researchers taking on questions within the topics identified would conduct more thorough reviews specific to their research objectives (page 28).

Minor comments:

10) The reference to ‘Patient Advisory’ throughout the paper feels strange/incomplete to me; adding the word ‘Board’, ‘Group’ or ‘Panel’ would make it easier to read.

The patient group discussed their role and drafted the Terms of Reference. “Patient Advisory” was the language they chose, and so we have honored that preference in the manuscript.

11) Introduction page 7 – I find the reference to ‘specific, structured’ frameworks and ‘non-specific tools’ confusing – what is the meaning of ‘specific’ here? Perhaps the sentence could be reworded.

We have reworded this to read: “There are a host of tools and techniques for priority setting. One recent methodological review found roughly equal numbers of studies that used published, structured frameworks and those that used newly-developed tools (e.g., surveys specific to the project) [12].”

12) Page 9 – ‘The Patient Advisory were research partners in PREFeR and, therefore, contributed to planning and shaping the research activities’ – it is not usually the case that research participants are involved in the research design. Could the authors provide a bit more detail about why and how they were involved in this way (as opposed to involving other patients)? Might their involvement in planning have had any impact on the findings?

Under Canada’s Strategy for Patient Oriented Research, patients are included as full team members. We now comment on this within the Discussion on pages 27-28. To clarify, however, the Patient Advisory contributed solely as research participants at the outset of the project (i.e., in terms of identifying priorities for primary health care), and then contributed to the research design thereafter. We, therefore, do not believe this impacted on the study findings in any way.

13) It would be good to know more about the selection process for recruiting patients to the advisory group. What proportion of those who expressed interest were selected, and how were they chosen?

This is an important point, so thank you for inviting us to provide more information around this. We now include this detail within the Methods (page 9) and Results (page 18) sections.

“In total, 11 members (8 female, 3 male) were recruited to the Patient Advisory between March and May 2017. Of these, 10 were recruited through recruitment advertisements placed with Patient Voices Network BC. A total of 3 males and 12 females replied to these ads and we chose 3 males and 7 females. An eighth female member was recruited from another study to ensure equal representation across the five regional health authorities in the province.”

14) Did the patient advisory group members already know each other or had worked together before this prioritisation exercise? If so, how might this have influenced the results? Is ‘group think’ a potential limitation?

They had not previously worked together. This was the first activity within the Patient Advisory. We now specify this on page 18. Moreover, one of the advantages of using the Nominal Group Technique is that it encourages equal contribution of all members because all ideas are shared in a round-robin fashion and the ranking of priorities is done individually and privately, without group discussion. While groupthink is always a possibility in any collaborative decision making activity, we are quite confident that this was not influential in the prioritization process.

15) What relevant skills and/or qualifications did the facilitator of the patient discussions have to ensure that all participants were given sufficient opportunity to voice their views?

The facilitator has a PhD in psychology and nearly a decade of experience working within large, multi-stakeholder primary care research teams that all included patient and public involvement members. She has also run patient and public involvement forums in previous roles. Additionally, the other research team members were present during the discussions, and co-facilitated to further ensure patient voices were given equal opportunity. We recognize that this information was not included in the manuscript and is important consideration for reflexivity. Therefore, we have now included this additional information on pages 11-12.

16) Page 10 – ‘We wanted the rapid literature reviews to inform the scope of what has already been researched within BC and Canada’ – I’m not sure how the reviews can inform something which has already happened? I think this needs rewording.

We have clarified the purpose of the rapid literature reviews (page 14):

“This information was used to inform the Integration phase and future development of research questions, but we do not report results herein. This reflects a divergence from the Dialogue Model, in which we carried out rapid literature reviews after the Consultation phase, instead of within the Exploration phase.”

17) Page 11 - Data Collection – it would be helpful to report the number of patients who were invited to take part. Also, ‘patients were asked to first briefly describe their primary care experiences’ seems not quite consistent with being asked ‘what stood out for you in primary care?’ – I would be more specific.

We have now clarified this on page 12:

“In line with NGT, patients were asked to first briefly describe the ideas they had in response to the question, “what things really stand out for you in primary care?”.”

18) Page 12 - Priority Setting Workshop – I assume this was a face-to-face meeting but I think this should be made explicit, especially as an earlier step used teleconferencing.

Yes, that is correct – it was a face-to-face meeting. We have now specified this on page 13.

19) Page 13 – Were patients given as long as they needed to rank the priorities, or was there a time limit?

Patients were given as long as they needed – now noted on page 14.

20) Page 13 – I think that creating a sum of scores for each topic was appropriate for the purposes of prioritisation, as was resolving ties by counting the number of votes, but I don’t understand why the ‘relative importance of each topic, or the proportion (%) of scores ranked within the top 10 topics’ was calculated. Unless I have misunderstood, the results in terms of rankings should be identical to the sum of scores (as shown in Table 3), so I’m not sure why this step was necessary? I find the sum of scores easier to interpret.

It is true the sum of scores is easiest to interpret. In revising the manuscript to include survey results, we have removed this detail from the manuscript itself and include former Table 3 only as an online appendix (Supplementary file 3).

21) On a related note, what happened to topics not chosen as patients’ top 10? Were they scored ‘0’?

These were simply not retained in the online survey, but can be seen in Supplementary file 3. No final ranked priority score is attributed to topics not retained in the top 10, in line with McMillan et al. However, if they were given ranked scores, they would fall within the range of 11-18.

22) Why were the participants in the Prioritisation stage asked to individually rate (rather than rank) the topics, while in the interim prioritisation phase they were ranked not rated? This could do with a brief explanation.

In the first stage (i.e., interim prioritization phase), our goal was to reduce the list to a manageable number for the online surveys. We ranked the topics in line with the scoring techniques described in McMillan et al., as already noted in the paper. In the second stage (i.e., prioritization via the surveys), our goal was to identify relative importance. Rating at the second stage generated more useful information as we were able to look not only at the rank order, but also understand which topics were closely clustered with respect to importance. We now explain this more fully on page 15, and agree that the distinction is worth making clear in the manuscript.

23) Table 1 – Age would be easier to interpret if range rather than (or as well as) standard deviation was reported. Previous research involvement – what does this mean – as a participant and/or in the design etc. of research? What do ‘moderate’ and ‘experienced’ equate to? Also, did the authors collect any information about patients’ health conditions / medical history? Since one of the objectives was to demonstrate the feasibility of involving patients in non-condition specific prioritisation, it would be really helpful to see how diverse a group they were in terms of their health issues / medical history – how might this have influenced the findings?

We have now added the age range, as well as described the definitions around research experience and types of health issues on page 16 and/or as footnotes to Table 1.

24) Page 19 – The patients provided ‘82 experiences and examples of what stood out for them in primary care in BC (data not shown)’ – could these be provided as a supplementary file?

In fact, all examples are listed in Supplementary file 2, grouped under corresponding topic headings. We now clarify this in the manuscript on page 15.

25) Table 3 – As mentioned above, I am confused by the inclusion of the ‘relative importance (%)’ and ‘ranked priority (via %)’ columns. The data are identical to the ‘sum of scores’ and ‘ranked priority (via scores)’ columns; I don’t think both are needed?

We have presented these results in line with McMillan et al., but now include this as a supplementary file (file 3).

26) Could the authors discuss any challenges they encountered and lessons learned? This could be very helpful for others wanting to carry out similar prioritisation exercises.

Many of the challenges and lessons learned correspond to your previous comments. In particular, how to balance a focus on patient priorities with other stakeholders, and how to tackle as broad a topic area as primary care. The Discussion has been substantially modified to include this content.

Thank you for the opportunity to review this paper. I hope these comments are helpful and look forward to reading the published version.

Thank you for your very constructive comments. By addressing and responding to these comments, we believe we have greatly strengthened the manuscript.

References:

Abma TA, Broerse JE. Patient participation as dialogue: setting research agendas. Health Expect 2010, 13(2):160-173.

Morris RL, Stocks SJ, Alam R, Taylor S, Rolfe C, Glover SW, Whitcombe J, Campbell SM. Identifying primary care patient safety research priorities in the UK: a James Lind Alliance Priority Setting Partnership. BMJ Open 2018, 8(2).

VERSION 2 – REVIEW

REVIEWER	Joanna Crocker University of Oxford, United Kingdom
REVIEW RETURNED	14-Feb-2019

GENERAL COMMENTS	Thank you for the opportunity to review this revised paper, which I think is much improved, and more useful now that it includes the online survey results too. The authors have addressed all my previous major concerns. I have a few minor comments/queries related to the revisions, listed below. 1) Abstract results – the mean ranges and SDs given in brackets are meaningless without an explanation of the scale used. I would replace with something like ‘all 10 topics were rated important or highly important, on average, by both groups’.

	2) Page 5 Summary of strengths and limitations of the study – It would be helpful to include detail of the characteristics referred to in the last point e.g. add in brackets ‘high educational background and prior experience of involvement in research’. 3) Page 8 – ‘We have only identified two recent publications…’ You could also include the priorities identified by the JLA PSP in primary care patient safety (Morris et al 2018). Reference: Morris RL, Stocks SJ, Alam R, et al. Identifying primary care patient safety research priorities in the UK: a James Lind Alliance Priority Setting Partnership. BMJ Open. 2018;8(2). 5) Page 10 – Thank you for providing further information about recruitment methods. I still think these methods target a small and biased sample of patients already engaged with research or health care initiatives (no effort was made to recruit ‘new’ patients / those with no prior experience of involvement or lower educational background)… In my view this is the main limitation of the paper and should be highlighted. 6) Page 17 – Rapid literature reviews. Can you briefly explain why you diverged from the Dialogue Model in this way? 7) Literature reviews, horizon scanning survey and dialogue event – You could perhaps remove or reduce these detailed methods sections as you don’t report on the results – I think they make the paper a longer and more complicated read whereas you could just focus on the NGT and online survey with patients and providers. You have said you will publish the findings of the dialogue event elsewhere and I think the detailed methods might sit better in that paper too. The overview in Figure 1 is helpful though. 8) Table 1 – ‘Employed’ is missing a ‘No’ category (or is the data missing)? ‘Country of birth’ is missing one person? How many Regional Health Authorities are there in BC / did you cover all of them? (it might be helpful to include a footnote). 9) Page 26 Interim workshop / Supplementary File 3 – As I previously noted, the rankings for sum of scores and relative importance are identical because they are based on the same raw data. Therefore only one ‘Ranked priority’ column (via sum of scores / relative importance) is necessary and would be less confusing. Also, the footnote says that the relative importance was calculated using a ‘maximum points for the group’ of 9 patients x 55 points = 495. I’m not sure where the 55 points has come from – is this definitely correct? 90 (9 patients x 10 points) would be the maximum possible sum of scores for each topic, but maybe I have misunderstood. 10) Table 3 needs a footnote to explain that importance was rated from 1 to 5 (with appropriate labelling), otherwise the mean importance is meaningless. 11) Page 18 – ‘A freetext box was provided for any comments respodnents wished to submit’ – were any of these comments suggested topics not already identified by the Patient Advisory? Although you rightly point out that you did not solicit additional topics in the survey, I think it would be worth briefly listing any suggested new topics/themes and mentioning them in the Discussion, as perhaps if they had been included in the survey,

	they would also have been rated as highly important (and could be explored by other groups in future). 12) Page 29 limitations – 'Although the initial priorities were developed by a group of patients purposively recruited to maximize diversity' – I don't think you can really say this given the targeting of existing groups only and the overall characteristics of the sample. You could say something like 'to maximise diversity with regard to gender, ethnicity, region and health issues' and then highlight the lack of diversity with regard to educational background and prior research experience. 13) Page 40 Ethical approval - 'Harmonized ethical approval' – it's not clear to me what this is or which part of the project it relates to – please could you clarify? 14) Figure 1 – A visual overview of the process is very helpful, but it is not clear how the different stages are related to each other. I would advise either restructuring it to show how the stages actually informed each other / fed into each other (which I personally would find more helpful), or else make it clear in the Figure caption that the phases are shown in chronological order (which I assume is the case based on my reading of the main paper). Thank you and best of luck!

VERSION 2 – AUTHOR RESPONSE

Reviewer: 2

Reviewer Name: Joanna Crocker

Institution and Country: Nuffield Department of Primary Care Health Sciences, University of Oxford, United Kingdom

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

1) Abstract results – the mean ranges and SDs given in brackets are meaningless without an explanation of the scale used. I would replace with something like 'all 10 topics were rated important or highly important, on average, by both groups'.

We thank the reviewer for the suggested change, which we have adopted (page 3).

2) Page 5 Summary of strengths and limitations of the study – It would be helpful to include detail of the characteristics referred to in the last point e.g. add in brackets 'high educational background and prior experience of involvement in research'.

We agree that this would provide helpful context for readers. We have amended this final bullet point on page 5 as follows: “Generalizability of the current priority topics might be limited, given the characteristics of the Patient Advisory (e.g., well educated, previous experience with research) and survey respondents (e.g., predominantly female, White).”

3) Page 8 – ‘We have only identified two recent publications…’ You could also include the priorities identified by the JLA PSP in primary care patient safety (Morris et al 2018).

Reference: Morris RL, Stocks SJ, Alam R, et al. Identifying primary care patient safety research priorities in the UK: a James Lind Alliance Priority Setting Partnership. BMJ Open. 2018;8(2).

Thank you for highlighting this additional reference. We have now included this as well, noting that Morris et al. were specifically looking to identify patient safety priorities in primary care. Again, this is not as broad-focused as our approach, but we agree that it is a relevant citation to include. This is listed as reference 12 in our list.

5) Page 10 – Thank you for providing further information about recruitment methods. I still think these methods target a small and biased sample of patients already engaged with research or health care initiatives (no effort was made to recruit ‘new’ patients / those with no prior experience of involvement or lower educational background)… In my view this is the main limitation of the paper and should be highlighted.

We recognized the importance of including patients without prior research experience and did seek patients with no or little prior research experience at the time of recruitment. This was stated in the additional recruitment details provided in the previously revised manuscript on page 9 (“We also sought to include both patients with and without previous research experience.”).

We have now included details about the Patient Advisory characteristics as a further limitation on page 30 (“Although the initial priorities were developed by a group of patients purposively recruited to maximize diversity with regard to gender, ethnicity, prior research experience, health region and health issues, they were mostly well educated and had previous experience with research, and we cannot claim to have reached saturation in possible topics.”), and this is already listed as one of the main limitations in the ‘Summary of strengths and limitations’ bullet points for the manuscript (page 5). We believe this now adequately highlights the sampling limitation raised by the reviewer.

6) Page 17 – Rapid literature reviews. Can you briefly explain why you diverged from the Dialogue Model in this way?

This was explained in the original submission, but we removed it from the revision in order to reduce the word count. We have now briefly stated why this was the case, once again, given it is important point of justification. However, deviations such as this are permitted within the Dialogue Model. We have inserted the following text to explain more about this on page 15: “This modification was made to provide input to contextualize what research had already been conducted within Canada. Within the Dialogue Model, literature reviews may be undertaken for scoping purposes, and to directly shape subsequent activities within the model, such as we chose to do.”

7) Literature reviews, horizon scanning survey and dialogue event – You could perhaps remove or reduce these detailed methods sections as you don’t report on the results – I think they make the paper a longer and more complicated read whereas you could just focus on the NGT and online survey with patients and providers. You have said you will publish the findings of the dialogue event elsewhere and I think the detailed methods might sit better in that paper too. The overview in Figure 1 is helpful though.

We thank the reviewer for this suggestion and have reduced these sections in this revision (text was deleted from the relevant sections within pages 14-18). We chose not to remove these sections completely, though, since it is important to the aim in this manuscript of overviewing the entire PREFeR Project activities. Some text is required in order to provide enough detail to understand what is presented in Figure 1.

8) Table 1 – ‘Employed’ is missing a ‘No’ category (or is the data missing)? ‘Country of birth’ is missing one person? How many Regional Health Authorities are there in BC / did you cover all of them? (it might be helpful to include a footnote).

Both of these variables were dichotomized, but we appreciate this was not clearly interpretable in the original table. Therefore, we have added the data for those not employed and the one member who was born outside of Canada. We thank the reviewer for the great suggestion of adding a footnote about the number of regional Health Authorities in BC (there are five), which we have now added in as footnote 2 in both Tables 2 and 3.

9) Page 26 Interim workshop / Supplementary File 3 – As I previously noted, the rankings for sum of scores and relative importance are identical because they are based on the same raw data. Therefore only one ‘Ranked priority’ column (via sum of scores / relative importance) is necessary and would be less confusing. Also, the footnote says that the relative importance was calculated using a ‘maximum points for the group’ of 9 patients x 55 points = 495. I’m not sure where the 55 points has come from – is this definitely correct? 90 (9 patients x 10 points) would be the maximum possible sum of scores for each topic, but maybe I have misunderstood.

We have now removed the ‘Ranked priority (via %)’ column that the reviewer noted was redundant with the earlier appearing ‘Ranked priority’ column. We have not highlighted this with track changes, so as to avoid confusion about the final supplementary file we are submitting.

In the revised manuscript, we removed some text that provided more detail about the tabulation of these scores. In an effort to keep the manuscript text as concise as possible, we have provided additional detail in the footnote of this revised supplementary file. Once again, we have not highlighted this with track changes, so as to avoid confusion about the final supplementary file we are submitting. The text we have added is as follows: “The total of 55 points is derived from the sum of the 10 ranks (1+2+3+4+5+6+7+8+9+10).”

10) Table 3 needs a footnote to explain that importance was rated from 1 to 5 (with appropriate labelling), otherwise the mean importance is meaningless.

Thank you for pointing out this accidental omission. We have now added this as a footnote to the table (page 26).

11) Page 18 – ‘A freetext box was provided for any comments respodnents wished to submit’ – were any of these comments suggested topics not already identified by the Patient Advisory? Although you rightly point out that you did not solicit additional topics in the survey, I think it would be worth briefly listing any suggested new topics/themes and mentioning them in the Discussion, as perhaps if they had been included in the survey, they would also have been rated as highly important (and could be explored by other groups in future).

We have not analyzed the freetext comments in detail and think it is beyond the scope of the current paper to do so. The freetext box instructions simply informed respondents that they could provide any comments they’d like to share (see Supplementary file 1). As we mentioned in our previous response about this aspect of the survey, very few respondents suggested new topics, since they were not specifically asked to do so. We contend that it would be slightly biased to report details for those who did, since no respondents were instructed to do so.

12) Page 29 limitations – ‘Although the initial priorities were developed by a group of patients purposively recruited to maximize diversity’ – I don’t think you can really say this given the targeting of existing groups only and the overall characteristics of the sample. You could say something like ‘to maximise diversity with regard to gender, ethnicity, region and health issues’ and then highlight the lack of diversity with regard to educational background and prior research experience.

We thank the reviewer for this suggested change to this limitation. The modified sentence on page 29 now states: “Although the initial priorities were developed by a group of patients purposively recruited to maximize diversity with regard to gender, ethnicity, prior research experience, health region and health issues, they were mostly well educated and had previous experience with research, and we cannot claim to have reached saturation in possible topics.”

13) Page 40 Ethical approval - ‘Harmonized ethical approval’ – it’s not clear to me what this is or which part of the project it relates to – please could you clarify?

We have now clarified this further within the ‘Ethical approval’ section on page 33. The ethical approval was ‘harmonized’ between two institutions, since research team members were based at two locations. We have additionally noted that this was granted for the topic importance surveys.

14) Figure 1 – A visual overview of the process is very helpful, but it is not clear how the different stages are related to each other. I would advise either restructuring it to show how the stages actually informed each other / fed into each other (which I personally would find more helpful), or else make it clear in the Figure caption that the phases are shown in chronological order (which I assume is the case based on my reading of the main paper).

The figure includes a yellow arrow running down the back of each of the key activities to indicate the flow of activities and how they were ordered, in line with the stages of the Dialogue Model. The reviewer is correct that these were presented in chronological order. To make this more apparent, we have amended the figure caption to noted that it is a flowchart (which implies one activity flows on to another).

Thank you and best of luck!

Once again, we’d really like to thank this reviewer for carrying out such a careful re-read of our manuscript. We agree that a number of areas pointed out by this reviewer have now benefited from additional points of clarification, and there was some text that could be reduced as suggested.