PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Top 10 (plus 1) research priorities for expectant families and those with children to age 24 months in Alberta, Canada: Results from the Family Research Agenda Initiative Setting (FRAISE) priority setting partnership project.
AUTHORS	Brockway, Meredith; Keys, Elizabeth; Bright, Katherine; Ginn, Carla; Conlon, Leslie; Doane, Stephanie; Wilson, Jacqueline; Tomfohr-Madsen, Lianne; Benzies, Karen

VERSION 1 – REVIEW

REVIEWER	Gronlund, Toto NIHR, James Lind Alliance
REVIEW RETURNED	25-Feb-2021

GENERAL COMMENTS	Thank you for submitting this manuscript and congratulations on doing the work. This is an important and interesting contribution to the field. A few minor suggestions for the manuscript follow below, mainly about structure, clarity and consistency. Abstract The Design section in the Abstract would benefit from a bit of proof-editing, to help make each phase and activity clear. It may be helpful to include numbers of people reached through the online survey and through in-person meeting. There is a reference to Phase One, Phase Two etc in the Abstract. These terms are not used elsewhere in the manuscript. The Methods section in the body of the manuscript mentions iterative phases A to E. Figure 1 describes the process in terms of the flow of Steering Committee meetings. Perhaps Figure 1 could also reference the phases A to E, and the Abstract amended accordingly? It would be really helpful for the reader to have more consistency in referencing the flow of the process. In the results section of the abstract, the current list of top 10(plus 1) is a rephrasing of the actual priorities. It would be perhaps more helpful to either list the actual priorities (not rephrased) OR list the topic areas of the priorities, as per Table 2, and write indicate "three of the priorities are on Mental Health/Relationships, two priorities on each of Access to Information,Immunity, Child development, and one priority on each of Sleep and Feeding" . Article Summary section: The strengths and limitations are a fair assessment. An additional strength is that this type of approach has revealed different kinds

of priorities to those already established and mainly determined by researchers, funders and industry.

Methods section:
It would be helpful to be clear how the FRAISE project differs from a standard JLA approach, and in particular, how the FRAISE project was less resource intensive and more practical for vulnerable groups. The author should be aware that the JLA process is flexible and adaptable, and is for example currently being used in lower income settings in Africa. Any learning that can be shared from the FRAISE approach would be invaluable for other priority setting partnerships.

The iterative phases A to E omit to mention the evidence checking (level of research) phase (which is mentioned in Figure 1).

The author is commended in undertaking in-person approaches to reach vulnerable groups. It would be helpful to understand how this was done.

Steering Committee section:
Could the author please clarify what was the final constitution of the Steering Committee, as compared to the aims. Did the core group of 15 include a spread of diversity?

How large was the wider online group of Steering Committee members, and what was the approximate constitution of this group, accepting that it varied during the project. What were the differences in the roles of the two steering committees ? Were these roles formalised in a "terms of reference" or similar ?

Online survey development section:
The references to "Steering Committee" - is this the core or the wider SC?

Priority setting workshops and consensus building section, and Figure 1:
The manuscript text refers to workshops and Figure 1 refers to SC Meetings. I assume they are synonymous, but for clarity use one term. There is also the question whether these involved the wider SC or the core group of 15?

The text for this section appears not to describe the first workshop, which was the development of the 12 themes and survey. It would be helpful if all 4 workshops were described and referenced, even if briefly.

Data analysis
The expert assessment stage appears to have reduced the number from 202 "codes" to 34. It is not clear how the criteria A,B,C,D were used, and whether the expert assessment is backed up be a reference base of evidence.

Inconsistency between Figure 1 and text: "34" questions in Figure 1, "30" mentioned in the text.

Results section:
Text states that "479" completed the survey, the Table 1 indicates N=480

	After Figure 2, the manuscript mentions that "Survey participants ranked stress, emotional, and mental health; sleep; and infant feeding as the three most important priorities." Was it somehow possible for survey participants to rank their submitted priorities in the first survey, or is this simply about the count of priorities submitted ? If it is a count (as per figure 3), then the term "ranked" should not be used. Finally, this manuscript reports on a competent, inclusive and transparent priority setting process. It appears to be based on the James Lind Alliance consensus process with minor variations. It would be helpful for other priority setting projects if this manuscript shared explicitly the modifications, and the rationale for the modifications, that have been made to the standard process.

REVIEWER	Theologis, Tim Oxford University Hospitals NHS Foundation Trust, Paediatric Orthopaedic Surgery
REVIEW RETURNED	30-Jun-2021

GENERAL COMMENTS	This is an interesting study, which applies research priority setting principles to a specific population. The research area (research priorities for expectant families and those with children to age 24 months) is interesting. The novelty is that the project was based on a specific community. The Authors have named the methodology a "modified James Lind Alliance" process based on a previously published paper. However, reading through the paper, the methodology appears fundamentally different to the JLA one. The quality assurance of the JLA is the supervision of the project by JLA representatives who encourage "fair play" and ensure that bias is not introduced by any of the participating stakeholders. Further to that, the JLA priority setting partnerships aim to produce generalisable results by involving all possible stakeholders with particular emphasis in under-represented minorities. The JLA representatives ensure that these voices are heard during the process. The other fundamental difference is that in JLA priority setting projects the research questions are suggested by the widest number of participants and not by the steering group. I understand that in this paper, the themes were produced by the steering group and participants expanded the number of specific questions. There is no doubt that setting research priorities with the involvement of all relevant stakeholders is an important first step in any area of research. The procedure that has been developed by the JLA to ensure that the research priorities are decided in a fair way that reflects opinions without bias has been tried and tested. I would question the reliability of modifications to this procedure as these may allow researcher bias and probably limit the generalisability of the results

VERSION 1 – AUTHOR RESPONSE

Reviewer: 1 **Ms. Toto Gronlund, NIHR**

Comments to the Author: Thank you for submitting this manuscript and congratulations on doing the work. This is an important and interesting contribution to the field. A few minor suggestions for the manuscript follow below, mainly about structure, clarity and consistency.	Thank you to Reviewer 1 for their positive feedback and constructive comments and suggestions.
Abstract The Design section in the Abstract would benefit from a bit of proof-editing, to help make each phase and activity clear. It may be helpful to include numbers of people reached through the online survey and through in-person meeting. There is a reference to Phase One, Phase Two etc in the Abstract. These terms are not used elsewhere in the manuscript. The Methods section in the body of the manuscript mentions iterative phases A to E. Figure 1 describes the process in terms of the flow of Steering Committee meetings. Perhaps Figure 1 could also reference the phases A to E, and the Abstract amended accordingly? It would be really helpful for the reader to have more consistency in referencing the flow of the process. In the results section of the abstract, the current list of top 10 (plus 1) is a rephrasing of the actual priorities. It would be perhaps more helpful to either list the actual priorities (not rephrased) OR list the topic areas of the priorities, as per Table 2, and write indicate "three of the priorities are on Mental	Thank you for this feedback, we have completed additional editing to improve clarity in this section. We have included numbers of people reached through the online survey and in-person meeting. The use of the term "phases" has been eliminated to focus more on the process. The wording is now consistent to what was written in the body of manuscript as well as the flow diagram.

Health/Relationships, two priorities on each of Access to Information, Immunity, Child development, and one priority on each of Sleep and Feeding".	Thank you for this suggestion. We have rephrased the results section as per your suggested wording. This now reads: *“The final top 10 (plus 1) research priorities included three priorities on Mental Health/Relationships, two priorities on each of Access to Information, Immunity, and Child Development, and one priority on each of Sleep, Pregnancy/Labour, and Feeding.”*
Article Summary section: The strengths and limitations are a fair assessment. An additional strength is that this type of approach has revealed different kinds of priorities to those already established and mainly determined by researchers, funders and industry.	Thank you for identifying this additional strength. We have revised this section and included this additional strength, which reads: “*By creating a welcoming and family-friendly atmosphere and including alternate engagement strategies, parents were meaningfully engaged throughout the partnership process, resulting in the identification of parent-oriented research priorities, many of which differed than priorities previously established by researchers, funders, and industry*.”
Methods section: It would be helpful to be clear how the FRAISE project differs from a standard JLA approach, and in particular, how the FRAISE project was less resource intensive and more practical for vulnerable groups. The author should be aware that the JLA process is flexible and adaptable, and is for example currently being used in lower income settings in Africa. Any learning that can be shared from the FRAISE approach would be invaluable for other priority setting partnerships.	We have added more details regarding the specific modifications that we have made in the methods section on page 8. This now reads: *“These modifications included the facilitation and expert coaching of the initial steering committee workshop by subject matter experts in patient engagement and experienced in priority setting partnerships from one of Canada’s regional Strategy for Patient-Oriented Research Support Unit (AbSPOR), rather than a JLA advisor. After this initial meeting, the lead authors, who are trained in group facilitation and community engagement, facilitated the remaining in-person meetings using consensus-building and shared-decision making strategies. During the analysis and prioritization of the potential research priorities, we used collective sensemaking to narrow down and rank the research priorities. Rather than conduct rapid literature reviews to determine the level of evidence available for the research uncertainties, we asked*

	practice and research subject matter experts to rate the availability of evidence on research uncertainties. These rankings were then used by the steering committee members to help them prioritize the top 30 priorities. "
The iterative phases A to E omit to mention the evidence checking (level of research) phase (which is mentioned in Figure 1).	Thank you for this feedback. We have added details regarding the evidence checking phase to the text as step 4 on page 9: *"(4) consult with research and practice experts to determine which codes were already well researched,"*
The author is commended in undertaking in-person approaches to reach vulnerable groups. It would be helpful to understand how this was done.	Thank you for this commendation. We have provided additional details on the strategies that we used in the methods section on page 9: *"These techniques included research assistants physically attending programs and services that provided targeted care to populations who were under-represented in the survey to recruit potential participants. To facilitate participation at these sites, research assistants carried electronic tablets that participants could use to complete the online survey before or after their programming or appointments."*
Steering Committee section: Could the author please clarify what was the final constitution of the Steering Committee, as compared to the aims. Did the core group of 15 include a spread of diversity?	Thank you for noting this omission. In response, we have added a sentence on page 10 to read: *"While we did not meet our target for Indigenous representation, we exceeded our aim for a* diverse group, *with 5 of the 15 parents identifying as a visible minority, 2 parents identifying as fathers, and 2 parents identifying as new to Canada."*
How large was the wider online group of Steering Committee members, and what was the approximate constitution of this group, accepting that it varied during the project. What were the differences in the roles of the two steering committees? Were these roles formalised in a "terms of reference" or similar?	We agree that additional details on the steering committee are needed and have added a more detailed explanation of online steering committee composition on page 10. We added: *"To accommodate the regionality and required flexibility of steering committee members, we provided opportunities for a larger online group of additional steering committee members. These members consisted of parents and providers who attended some, but not all workshops in person, as well as healthcare providers from other sites in Alberta, and directors and managerial staff who were unable to attend in-person workshops due to time constraints. The size of this group fluctuated throughout the study timeline, with a maximum of 10 members in addition to the core steering committee. The role of the larger steering committee was to provide online feedback and consultation throughout the priority setting process."*

Online survey development section: The references to "Steering Committee" - is this the core or the wider SC?	The reference in this section should have been to the core steering committee. We have revised this section on page 10 this to state “core steering committee” when applicable.
Priority setting workshops and consensus building section, and Figure 1: The manuscript text refers to workshops and Figure 1 refers to SC Meetings. I assume they are synonymous, but for clarity use one term. There is also the question whether these involved the wider SC or the core group of 15?	Figure 1 now uses the term ‘workshop’ for consistency. Also, use of *meeting* has been replaced with *workshop* throughout text. The numbers of steering committee members and ‘core’ or ‘wider’ have been added throughout the manuscript for clarity.
Data analysis The text for this section appears not to describe the first workshop, which was the development of the 12 themes and survey. It would be helpful if all 4 workshops were described and referenced, even if briefly.	We have added a description of the 1st steering committee workshop to the *Online survey development to identify research questions* Section as this appeared to be a better fit.
The expert assessment stage appears to have reduced the number from 202 "codes" to 34. It is not clear how the criteria A,B,C,D were used, and whether the expert assessment is backed up be a reference base of evidence.	We have edited this section for clarity so the reader may now understand that the steering committee reduced the number of priorities, not the experts. On page 13, this now reads: *“Steering committee members were instructed to review and consider expert feedback when building consensus about which of the 30 priorities to move forward”* We hope this revision helps to clarify how the experts used the criteria of the availability of evidence to make recommendations on the level of evidence available for each research uncertainty, as outline on page 12, where we have outlined that experts were asked to rate the level of evidence according to whether there were systematic reviews and/or meta-analyses, single studies and/or inconsistencies, or there were no research studies: “experts were asked to indicate if each potential research priority was: (1) well researched (systematic reviews and meta-analysis available), (2) somewhat researched (single studies, some inconsistent evidence on topic), (3) not researched (no studies), or (4) unsure.” We hope our previous clarification

	The modification to JLA of using experts instead of rapid literature review was added to the limitations section on page 22 and now reads: *However, these modifications (e.g., using external group facilitators with expertise in patient engagement to establish the steering committee rather than a paid JLA advisor; consulting with experts on the level of evidence, rather than conducting rapid literature reviews on each potential research uncertainty) greatly reduced the cost of the project, thereby increasing feasibility.*
Inconsistency between Figure 1 and text: "34" questions in Figure 1, "30" mentioned in the text.	Thank you for noting this inconsistency as confusing. While our original procedure specified that the priorities would be narrowed down to 30, at the time of prioritization, the steering committee felt that 34 was appropriate for this initial stage of reducing the research questions. Thus, while it was our original intent to have a top 30 questions, in practice, we had a list of the top 34 questions. We have clarified this by providing additional details in the text on page 13: "*While the original intent was to narrow down the priorities to a top 30 list, at the time of prioritization, the steering committee reached consensus that the list should include 34 priorities*)." We have also revised Figure 1 to help clarify this distinction.
Results section: Text states that "479" completed the survey, the Table 1 indicates N=480	Thank you for noting this discrepancy. We have corrected this error on page 14 to ensure that the N = 480 in both the text and the Table.
After Figure 2, the manuscript mentions that "Survey participants ranked stress, emotional, and mental health; sleep; and infant feeding as the three most important priorities." Was it somehow possible for survey participants to rank their submitted priorities in the first survey, or is this simply about the count of priorities submitted? If it is a count (as per figure 3), then the term "ranked" should not be used.	We included an item in the first survey that asked parents to rank the 12 parenting topics of interest that were identified by the steering committee. This has been clarified and this sentence on page 16 now reads: "*When asked to rank the 12 broad parenting topics of interest, survey participants ranked stress, emotional, and menta l health; sleep; and infant feeding as the three most important parenting topics*."

Finally, this manuscript reports on a competent, inclusive and transparent priority setting process. It appears to be based on the James Lind Alliance consensus process with minor variations. It would be helpful for other priority setting projects if this manuscript shared explicitly the modifications, and the rationale for the modifications, that have been made to the standard process.	Thank you for this feedback. We have added details about this when discussing the strengths and limitations on page 22, which now reads: *“Additionally, while this research priority setting partnership used a process based on a previously reported modified JLA approach,*[13] *these modifications have not been validated against the standardized JLA method. These modifications (e.g., using external group facilitators with expertise in patient engagement to establish the steering committee rather than a paid JLA advisor; consulting with experts on the level of evidence, rather than conducting rapid literature reviews on each potential research uncertainty) greatly reduced the cost of the project, thereby increasing feasibility. Additionally, the current approach incorporated principles of participatory action research (e.g., engaging with a community who have self-identified in reciprocal relationships, recognition of local knowledge, and incorporating processes of co-learning to take actions that will improve community member’s wellbeing), which are widely recognized as critical in facilitating meaningful participant engagement and ensuring that participants’ voices are represented throughout the research process.”*
Reviewer: 2 Mr. Tim Theologis, Oxford University Hospitals NHS Foundation Trust	
Comments to the Author: This is an interesting study, which applies research priority setting principles to a specific population. The research area (research priorities for expectant families and those with children to age 24 months) is interesting. The novelty is that the project was based on a specific community. The Authors have named the methodology a "modified James Lind Alliance" process based on a previously published paper. However, reading through the paper, the methodology appears fundamentally different to the JLA one. The quality assurance of the JLA is the supervision of the project by JLA representatives who encourage "fair play" and ensure that bias is not introduced by any of the participating stakeholders. Further to that, the JLA priority setting partnerships aim to	Thank you to Reviewer 2 for sharing their feedback on our manuscript. In response to the concerns outlined, and as requested by Reviewer 1, we have provided additional details regarding the methodology and modifications made to the JLA process. We hope that our revisions in the methods section, in response to feedback provided by Reviewer 1, have more clearly outline the process and how the researchers’ encouraged “fair play” among steering committee members to ensure that all members of the diverse steering committee had opportunities for their voices to be reflected in the top research priorities. We believe our strategies to engage representatives from communities who are typically under-represented in maternal child health research (i.e., fathers; diverse ethnicities) were relatively successful in both the larger survey and the steering committee. We have added additional details about the external facilitation and coaching that we received from patient engagement experts at one of Canada’s regional Strategy for Patient-Oriented Research Support Units (AbSPOR) for the initial steering committee, as well as the lead authors’ experience in using group facilitation techniques. We acknowledge the concern that the validity of modifications has not been tested and that this may limit generalizability. However, our innovative, community-based participatory action

produce generalisable results by involving all possible stakeholders with particular emphasis in under-represented minorities. The JLA representatives ensure that these voices are heard during the process. The other fundamental difference is that in JLA priority setting projects the research questions are suggested by the widest number of participants and not by the steering group. I understand that in this paper, the themes were produced by the steering group and participants expanded the number of specific questions. There is no doubt that setting research priorities with the involvement of all relevant stakeholders is an important first step in any area of research. The procedure that has been developed by the JLA to ensure that the research priorities are decided in a fair way that reflects opinions without bias has been tried and tested. I would question the reliability of modifications to this procedure as these may allow researcher bias and probably limit the generalisability of the results	approach presents much potential for the population being researched. With the additional details provided in this revised version, we hope that other researchers will be able to replicate our method and demonstrate validity of these modifications. We have revised the strengths and limitations in the Study Summary section, as well as in the discussion, to more explicitly acknowledge these limitations.

VERSION 2 – REVIEW

REVIEWER	Gronlund, Toto NIHR, James Lind Alliance
REVIEW RETURNED	15-Aug-2021

GENERAL COMMENTS	Thank you for revising the paper and addressing the review questions and points. There are two items I would still wish to be addressed. The first is: On page 7 of the revised manuscript it is claimed that the JLA process is "resource intensive". Please consider alternative, more transparent and meaningful wording here, or indeed just remove this. This statement seems mainly to refer to the fact that the project did not employ an accredited JLA adviser, and used steering committee expertise for evidence checking,

	rather than a literature search of systematic reviews and guidelines. In not employing a JLA adviser, costs to the project may have been reduced, though the opportunity cost of the researcher time should be considered in any resource estimation. It is not clear how the methodology adopted in this project is less "resource intensive" in terms of time committed and overall project costs, including researcher time and costs. In fact, the community engagement approach used must have been resource intensive, as this requires in-person time. My second point is more cosmetic, and is about the multiple different references to 'parents' 'patients' 'families' 'people' 'caregivers' 'providers' 'knowledge users' 'community agency representatives' 'researchers' 'clinicians'. Clearly many of these terms are relevant and appropriate where they are used in the manuscript, but at times the terms used seems somewhat random. In particular is not clear whether "caregivers" includes both informal supporters (family friends) and professional caregivers such as clinicians. This is important because the study objective / participant definition includes only 'expectant parents and caregivers'. Please review the use of these terms, and ensure they are appropriate throughout the manuscript. Is the term patient relevant ? (other than in relation to describing the JLA process, or in discussion of the results of project in more general terms). The term 'knowledge user' could perhaps be an useful overall collective name, and is defined on page 5 as including parents, clinicians and community agency representatives. Does 'knowledge users' also include caregivers? All these stakeholder types are participants in the process as per Table 1. I know this is tricky, having to repeat the stakeholder types time and again!

REVIEWER	Theologis, Tim Oxford University Hospitals NHS Foundation Trust, Paediatric Orthopaedic Surgery
REVIEW RETURNED	24-Aug-2021

GENERAL COMMENTS	My main concern with the previous edit of this paper related to the definition of its methodology: the Authors defined their methodology as a “modified James Lind Alliance” priorities setting partnership (JLA PSP). The revised version of the paper clarified where this study deviated from the JLA PSP. It did not employ trained JLA facilitators but external experts. It did not involve rapid literature searches but was based on expert opinions. The deviations from the JLA PSP methodology would affect the validity of the study. The modified methodology led to cost reduction in this project but did not allow for the rigorous supervision and “fair play” of the JLA led projects. The Authors have otherwise addressed the issues raised in the previous review.

VERSION 2 – AUTHOR RESPONSE

Reviewer 1: Ms. Toto Gronlund, NIHR

Comments to the Author:	Author Response
Thank you for revising the paper and addressing the review questions and points. There are two items I would still wish to be addressed. The first is: On page 7 of the revised manuscript it is claimed that the JLA process is "resource intensive". Please consider alternative, more transparent and meaningful wording here, or indeed just remove this. This statement seems mainly to refer to the fact that the project did not employ an accredited JLA adviser, and used steering committee expertise for evidence checking, rather than a literature search of systematic reviews and guidelines. In not employing a JLA adviser, costs to the project may have been reduced, though the opportunity cost of the researcher time should be considered in any resource estimation. It is not clear how the methodology adopted in this project is less "resource intensive" in terms of time committed and overall project costs, including researcher time and costs. In fact, the community engagement approach used must have been resource intensive, as this requires in-person time.	Thank you to Reviewer 1 for their time and insight in completing the review. Thank you for this feedback. We have revised this section of the manuscript by omitting the claim that the JLA is resource intensive. The corresponding section (1st paragraph in the methods section) now reads: "Many PSPs use the JLA approach;[6] however, employing a trained JLA facilitator may be impractical for lower resourced research studies."
My second point is more cosmetic, and is about the multiple different references to 'parents' 'patients' 'families' 'people' 'caregivers' 'providers' 'knowledge users' 'community agency representatives' 'researchers' 'clinicians'. Clearly many of these terms are relevant and appropriate where they are used in the manuscript, but at times the terms used seems somewhat random. In particular is not clear whether "caregivers" includes both informal supporters (family friends) and professional caregivers such as clinicians. This is important because the study objective / participant definition includes only 'expectant parents and caregivers'. Please review the use of these terms, and ensure they are appropriate throughout the manuscript.	Thank you for this feedback. We have reviewed the manuscript in detail and made edits throughout to try to use these terms more precisely. We have tried to clarify that caregivers includes both informal caregivers (i.e., those providing daily care to children but who may not identify as their parent) and professional caregivers. We hope the added text on page 7 clarifies this: "Involving parents and caregivers (i.e., those who provide direct care for children and who may or may not self-identify as parents, as well as professional caregivers who provide care to families with children)."

Is the term patient relevant ? (other than in relation to describing the JLA process, or in discussion of the results of project in more general terms). The term 'knowledge user' could perhaps be an useful overall collective name, and is defined on page 5 as including parents, clinicians and community agency representatives. Does 'knowledge users' also include caregivers? All these stakeholder types are participants in the process as per Table 1. I know this is tricky, having to repeat the stakeholder types time and again!	Thank you for this important feedback. We have revised the text throughout to more precisely use the terms patient, caregiver, and knowledge user. We now only include the term patient when referring to "patientoriented research" and other more general processes. We hope our edits, including the definition of caregivers, have added clarity to the manuscript.

Reviewer 2: Mr. Tim Theologis, Oxford University Hospitals NHS Foundation Trust	
Comments to the Author:	Author Response

Comments to the Author: My main concern with the previous edit of this paper related to the definition of its methodology: the Authors defined their methodology as a "modified James Lind Alliance" priorities setting partnership (JLA PSP). The revised version of the paper clarified where this study deviated from the JLA PSP. It did not employ trained JLA facilitators but external experts. It did not involve rapid literature searches but was based on expert opinions. The deviations from the JLA PSP methodology would affect the validity of the study. The modified methodology led to cost reduction in this project but did not allow for the rigorous supervision and "fair play" of the JLA led projects. The Authors have otherwise addressed the issues raised in the previous review.	Thank you to Reviewer 2 for their time and insight in providing their review. We have added the limitation that our modified methodology did not include the rigorous external supervision of a JLA-led project. The paragraph in the discussion about the study limitations now reads: "Additionally, while this research priority setting partnership used a process based on a previously reported modified JLA approach,13 these modifications have not been validated against the standardized JLA method. Not employing a trained JLA facilitator means that the study did not have the rigorous external supervision that other JLA-led studies benefit from.“