PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Moving Forward Through Consensus – Protocol for a Modified Delphi Approach to Determine the Top Research Priorities in the Field of Orthopaedic Oncology
AUTHORS	Schneider, Patricia; Evaniew, Nathan; Rendon, Juan; McKay, Paula; Randall, R.; Turcotte, Robert; Vélez, Roberto; Bhandari, Mohit; Ghert, Michelle; Investigators, PARITY

VERSION 1 - REVIEW

REVIEWER	Prof. Ajay Puri Tata Memorial Hospital Mumbai, India
REVIEW RETURNED	26-Mar-2016

GENERAL COMMENTS	Excellent initiative and should be given the go ahead. A few suggestions and comments; • The authors expect to invite approximately 125 clinician-scientists who would include participants from North and South America, Europe, Asia, Africa and Australia with substantial variation in prior research experience, clinical focus, and career stage. Their initial “net” would include members participating in the PARITY trial, Musculoskeletal Tumor Society (MSTS) members, and Orthopaedic Research and Education Foundation (OREF) partners. I believe this would essentially have a North American bias and would not be truly representative of the “global” orthopaedic oncology fraternity. They may need to cast their net wider by soliciting more participants (one strategy could be to include authors/researchers who have published original articles in high impact journals relevant to this field). • In the discussion the authors mention that “The validity of several consensus methods has been questioned due to manipulation of the processes by organizers”. I think the proviso that “Any research questions that the reviewers feel are missing will be added to the list of questions that progress further” leaves the authors open to this bias. It is highly unlikely that 125 participants would miss a clinically relevant research question. The authors thus risk biasing the study with the introduction of “such missing” questions. • The authors risk a similar “individual researcher” bias in Phase III by bringing forward for final ranking a research question which at least two respondents score as a nine. As the number of total participants is undefined it may be better to use a percentage of total participants scoring a question as nine rather than a “fixed” number 2.

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	One of the main drawbacks of the "Delphi" technique is the lack / absence of response from adequate participants. It would be nice if the authors have a "fall back"plan in case the initial response to their mailings is inadequate. Overall an excellent proposal that should identify relevant questions on which to focus research.

REVIEWER	Jacob D. Hill Cancer Treatment Centers of America Midwestern Regional Medical Center Zion, Illinois, United States of America
REVIEW RETURNED	11-Apr-2016

GENERAL COMMENTS	Overall, this is a creative and well-described application of a modified Delphi panel for use in orthopedic oncology research. I expect the method described in this protocol will be successful in producing the desired outcomes of establishing clarification and censuses to critical research questions in orthopedic oncology. I applaud the authors and research team for the well developed protocol and recommend the manuscript be accepted for publication. A few questions for clarification: 1. In phase II-Ranking Evaluation, you mention if none of the research questions meet the inclusion or non-consensus thresholds, then the thresholds will be lowered until a critical mass of research questions are able to be brought forward for the next phase (phase III). What will be this critical mass of questions? How will this be determined? From prior experience with Delphi panels, it is important to balance how many questions are discussed among a panel of experts (for the in-person discussion). If there are numerous experts on the panel, there will be constraints on the amount of time or the amount of questions that can be fully discussed. If the panel is smaller more time can be dedicated per question, or a greater number of questions can be discussed. 2. In phase III-In-Person Expert Panel Consensus Meeting, you mention if too many individuals express interest in participation on the expert panel, then you will select a group with diverse geographical backgrounds, and a balanced configuration of stakeholders and career stages. Do you have a more systematic approach to selecting this panel? How many individuals will be selected for the panel? How do you decide someone is qualified to represent the field of orthopedic oncology research on the international level? 3. In phase III-In-Person Expert Panel Consensus Meeting, you mention there will be an experienced facilitator. However, is there a predetermined structure to this discussion to ensure everyone has a fair and equal chance to express their opinion? For example, there could be a period of uninterrupted answers for each expert, followed by open discussion, followed by final individual thoughts. From prior experience with an in-person Delphi discussion, there are certain personality types that will hesitate to speak up unless provided an opportunity to speak. These individuals may not have the chance to share important points and experience unless a semi-structured format is implemented.

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	These questions are provided to further support this manuscript, and are no means required to be addressed. As previously stated, I support this manuscript for publication and look forward to final result of this modified Delphi panel.

VERSION 1 – AUTHOR RESPONSE

Reviewer 1

COMMENT:
Excellent initiative and should be given the go ahead.

RESPONSE:
Thank you for your thoughtful review of our manuscript.

COMMENT:
The authors expect to invite approximately 125 clinician-scientists who would include participants from North and South America, Europe, Asia, Africa and Australia with substantial variation in prior research experience, clinical focus, and career stage. Their initial "net" would include members participating in the PARITY trial, Musculoskeletal Tumor Society (MSTS) members, and Orthopaedic Research and Education Foundation (OREF) partners. I believe this would essentially have a North American bias and would not be truly representative of the "global" orthopaedic oncology fraternity. They may need to cast their net wider by soliciting more participants (one strategy could be to include authors/researchers who have published original articles in high impact journals relevant to this field).

RESPONSE:
We agree that only including participants from the PARITY trial, Musculoskeletal Tumor Society (MSTS) members, and the Orthopaedic Research and Education Foundation (OREF) would not be truly representative of the global orthopaedic oncology community. As such, we have decided to also invite members from the Connective Tissue Oncology Society (CTOS) and the International Society of Limb Salvage (ISOLS). Moreover, given that the CTOS membership also includes physicians other than orthopaedic oncologists, it is likely that inviting CTOS members to participate in this initiative will also increase the number of stakeholders represented.

CHANGE:
We have made the following changes to the methods (page 6, third paragraph):
We will invite approximately 200 clinician-scientists who are interested or actively participating in the PARITY trial, Musculoskeletal Tumor Society (MSTS) members, Connective Tissue Oncology Society (CTOS) members, International Society of Limb Salvage (ISOLS) members and Orthopaedic Research and Education Foundation (OREF) partners to participate.

COMMENT:
In the discussion the authors mention that "The validity of several consensus methods has been questioned due to manipulation of the processes by organizers". I think the proviso that "Any research questions that the reviewers feel are missing will be added to the list of questions that progress further" leaves the authors open to this bias. It is highly unlikely that 125 participants would miss a clinically relevant research question. The authors thus risk biasing the study with the introduction of "such missing" questions.

RESPONSE:
We agree that the addition of any missing research questions to the list of questions that progress to the next phase may introduce bias.

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CHANGE:
We have removed from the methods (page 7, second paragraph):
Any research questions that the reviewers feel are missing will be added to the list of research questions that progress to Phase II.

COMMENT:
The authors risk a similar “individual researcher” bias in Phase III by bringing forward for final ranking a research question which at least two respondents score as a nine. As the number of total participants is undefined it may be better to use a percentage of total participants scoring a question as nine rather than a “fixed” number 2.

RESPONSE:
We agree that since the number of total participants is currently undefined, it would be better to use a percentage of total participants scoring a question as nine rather than a fixed number of participants.

CHANGE:
We have made the following changes to the methods (page 9, second paragraph):
At least 10% of respondents scored the research question as a nine.

COMMENT:
One of the main drawbacks of the "Delphi" technique is the lack / absence of response from adequate participants. It would be nice if the authors have a "fall back" plan in case the initial response to their mailings is inadequate.

RESPONSE:
We agree that we may have received an inadequate number of responses had we only invited clinician-scientists who are interested or actively participating in the PARITY trial, Musculoskeletal Tumor Society (MSTS) members and Orthopaedic Research and Education Foundation (OREF) partners to participate. Since expanding our target population to also include members of the Connective Tissue Oncology Society (CTOS) and the International Society of Limb Salvage (ISOLS), we believe that we will receive an adequate number of responses to generate a comprehensive list of possible research questions.

Reviewer 2

COMMENT:
Overall, this is a creative and well-described application of a modified Delphi panel for use in orthopedic oncology research. I expect the method described in this protocol will be successful in producing the desired outcomes of establishing clarification and censuses to critical research questions in orthopedic oncology. I applaud the authors and research team for the well developed protocol and recommend the manuscript be accepted for publication.

RESPONSE:
Thank you for this comment and your thoughtful review of our manuscript.

COMMENT:
In phase II-Ranking Evaluation, you mention if none of the research questions meet the inclusion or non-consensus thresholds, then the thresholds will be lowered until a critical mass of research questions are able to be brought forward for the next phase (phase III). What will be this critical mass of questions? How will this be determined? From prior experience with Delphi panels, it is important to balance how many questions are discussed among a panel of experts (for the in-person discussion).

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If there are numerous experts on the panel, there will be constraints on the amount of time or the amount of questions that can be fully discussed. If the panel is smaller more time can be dedicated per question, or a greater number of questions can be discussed.

RESPONSE:
We will consider ten research questions a critical mass. However, in our experience with Delphi studies, we have found that most issues/options progress through to the consensus meeting. Therefore, it is likely that most questions will be brought forward to the consensus meeting, but will be tiered for discussion according to priority.

CHANGE:
We have added the methods (page 8, first paragraph):
We consider a priori that a critical mass of ten priority research questions will suffice. However, Delphi processes tend to be flexible and therefore it is more likely that there will be a large number of questions brought forward for discussion.17 If this is the case, we will rank the questions in tiers and give priority to discussion of the highest ranking tier.

Reference:
Fink A, Kosecoff J, Chassin M, and Brook R. Consensus methods: characteristics and guidelines for use. Am J Public Health 1984; 74(9): 979-83.

COMMENT:
In phase III-In-Person Expert Panel Consensus Meeting, you mention if too many individuals express interest in participation on the expert panel, then you will select a group with diverse geographical backgrounds, and a balanced configuration of stakeholders and career stages. Do you have a more systematic approach to selecting this panel? How many individuals will be selected for the panel? How do you decide someone is qualified to represent the field of orthopedic oncology research on the international level?

RESPONSE:
We agree that a more systematic approach to selecting the expert panel may ensure a more balanced configuration of geographical backgrounds, stakeholders and career stages. We will select a group of 20 individuals and will ensure that there is at least one representative from each geographical background, stakeholder group and career stage.

CHANGE:
We have made the following changes to the methods (page 8, second paragraph):
Should too many individuals express their interest in participating on the expert panel, we will select a diverse and balanced group comprised of 20 individuals, with at least one individual from each of the geographical, stakeholder and career stage groups.

COMMENT:
In phase III-In-Person Expert Panel Consensus Meeting, you mention there will be an experienced facilitator. However, is there a predetermined structure to this discussion to ensure everyone has a fair and equal chance to express their opinion? For example, there could be a period of uninterrupted answers for each expert, followed by open discussion, followed by final individual thoughts. From prior experience with an in-person Delphi discussion, there are certain personality types that will hesitate to speak up unless provided an opportunity to speak. These individuals may not have the chance to share important points and experience unless a semi-structured format is implemented.

RESPONSE:
We agree that some structure to the consensus meeting will ensure that all participants have an

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opportunity to voice their opinions. We have developed a semi-structured agenda [see Figure Two] to provide participants in order to minimize time constraints on the day of the consensus meeting and to ensure that all participants are allotted the time required to express their ideas/opinions.

CHANGE:
We have added to the methods (page 8, third paragraph):
A semi-structured agenda will be provided to minimize time constraints and to ensure that all individual participants are allowed a period of uninterrupted time to voice their opinions for each topic discussed [see Figure Two].

Once again, we would like to thank you and the reviewers for your careful review and constructive feedback of our manuscript. We look forward to your reply.

VERSION 2 – REVIEW

REVIEWER	Prof. Ajay Puri Tata Memorial Hospital, India
REVIEW RETURNED	29-Apr-2016

GENERAL COMMENTS	All reviewer comments have been adequately addressed in the revised manuscript.

REVIEWER	Jacob Hill ND, MS Cancer Treatment Centers of America Midwestern Regional Medical Center Zion, Illinois, USA
REVIEW RETURNED	30-Apr-2016

GENERAL COMMENTS	The revisions made to the original submitted manuscript are acceptable and clarify any questions or concerns. I recommended the manuscript be accepted for publication.

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