Author’s response to reviews

Title: Values and value conflicts in implementation and use of preconception expanded carrier screening - an expert interview study

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Author’s response to reviews:

Dear Editor:

We thank you and the reviewers for the feedback on the article. The input and the changes that have been made to the article has improved it in content and quality. Below, we have replied to all the reviewers’ comments and questions. The responses, in the letter, are in dark red. In the manuscript, the alterations that have been made are highlighted in yellow and tracked changed.

Best regards

Rebecca Bennett (Reviewer 1):

This paper aims to ‘investigate values that experts recounted in relation to implementation and use of preconception ECS’. It does this with a number of interviews from members of Swedish ethics committees. I cannot comment on the validity of the empirical work done which I presume another reviewer will verify.

While there has clearly be a great deal of work that has gone into this paper at the moment it lacks a clear purpose that would be needed to provide value to these interviews.

The main problem with the paper as it stands is it is not clear why the authors wanted to know what values influenced these experts in their decisions in this area. The authors need to be clearer from the outset what question they wanted to answer by doing this research.
We agree with the reviewer, the background has been changed to better explain the purpose of examining value and value conflicts in relation to preconception ECS. The changes are kept in track changes and highlighted in yellow. Briefly, preconception ECS is defined as a health technology warranting assessment, part of which is addressing values and in the background we explain why that is important. Please refer to lines 94-112.

A further problem with this paper as it stands is that it is likely that these results would be similar to most people's comments on an ethical issue and it is therefore not clear why it is useful to know that these 10 people think about autonomy/respect for persons, nonmaleficence/do no harm, justice etc etc.

If the purpose of the paper is to identify the sort of values that are shaping discussions of this issue in Swedish ethics committees then we need to know why doing so is important.

This comment relates to the previous one. We have addressed this in the background, where we motivate the study in terms of dissemination of the preconception ECS as a new HT in Sweden.

The paper starts, in a number of places to do more than just reflect what these 10 people see as the pertinent values here, it starts to argue about these values and the ethical arguments in this area but only in fits and starts as space won't allow a full discussion of this.

The paper now focuses on reporting of the empirical results and discussing them in the light of literature on values in the bioethical literature. We have removed the theoretical framework as we agree with the reviewers that it was too ambitious to do both (report empirical results and conduct an ethical analysis where we discuss value theory) in one paper. Please refer to altered background and methods sections in the manuscript.

This paper has potential and I can see the work that has gone into the interviews it reports, however, to make the most of this hard work the authors need to be much clearer a) what the question was that they were attempting to answer b) why this is an important question to answer c) what their answer to this question was as a result of these interviews and d) what this work adds to the debate.

In the new version of the manuscript, we have covered the 4 questions as follows

a- The study is investigating the value and value conflicts in relation to implementation and use of preconception ECS in Sweden, and if any of these values conflict with the societal values.

b- It is important to address such a question with any new health technology that may be implemented. One reason is to overcome the dissemination problem and uptake by politicians and within Swedish healthcare system.
c- The experts harbored several concerns regarding moral values and principles associated with preconception ECS, some of which were contradicting with Swedish healthcare moral values of solidarity, human dignity and cost effectiveness.

d- The results are pertinent to Swedish context primarily but the work gives insight on how new HT can challenge local moral norms. In addition, the results could be pertinent to other Scandinavian countries where its healthcare systems are founded on certain moral values.

If this can be done then the work done in the interviews can really have some impact. However, it would be advisable to focus on reporting these interviews and the values expressed rather than trying to do more and start analysing these values and arguments as this is too much for one paper to do.

We agree with the reviewer and we have removed the theoretical framework. We have followed the advice of the reviewer and the study now reports only the empirical findings and discuss them in the light of some of the bioethical literature on values.

Detailed comments

P. 2 This is a bit of an over simplified view of this debate. There is a great deal of debate around how useful principles such as dignity are when they are so ambiguous for instance. It is not clear that there is any consensus on these principles any part of the world.

In the background we attempted to give an overview of values as used in some policy and regulation documents. We have not attempted to engage in the debate of definitions or descriptions of what those values mean. This was addressed briefly in the discussion of the results of the paper. As advised, we concentrated on the results and did not deliberate on a discussion of a more theoretical nature.

Page 4 - I'm not sure what point you are making here and this ends up being difficult to follow.
Page 5 - first full sentence needs a reference.

This section has been removed and some parts incorporated into the background just to give an overview of what moral values mean.

Page 5 - para 2 But why do you want to know this? What question are you trying to answer with this study? Your aim later in this page needs to come earlier in the paper so that the reader can understand what the paper wants to do.
We have reformulated the aim in terms of HTA. Please refer to the background and methods sections.

Jenny Krutzinna (Reviewer 2):

Thank you for this interesting article on an important topic. Your research addresses a very relevant and timely issue of the different values influencing health policy-making. However, I have some major comments and questions about some aspects of the paper, which may need to be addressed before publication.

General comments:
- The structure of the paper could be improved by guiding the reader to the next sections. At present, individual sections appear disconnected from each other and it is not clear where the article is going.
- The findings could be more effectively presented, for instance, with a clearer structure and explanation of the categorisation at the outset.
- It is sometimes not clear if a statement came from a study participant or if it is the authors' view. Please check that all responses are clearly attributed to study respondents or indicate where you are commenting as authors. - My main concern is the lack of clarity with regard to the conceptual framework (see below).

The paper has been changed, some sections deleted and explanations added to better communicate the findings of the study. We have explained the analysis process and the results and restructured them to show the themes, categories and subcategories.

The theoretical framework has been removed and we focused on presenting the results and analyzing them with clearly demarcated themes categories and sub categories. We also explained what the different findings mean. All that has been presented in the result section are respondents views, not the authors’ nor the literature. We have rephrased the text to reflect this in the results section.

Background

This section provides a brief historical overview of some developments in bioethics with regard to values. However, no mention is made of the relevance of this to the current research question / study. The topic of ECS or similar modern health technologies that raise particular ethical
questions is not mentioned or discussed in the background, although in the abstract it is referred to under the heading "background".

It may be a good idea to providing some information on the motivation for the study in this section, as well as an explanation of ECS and its relevance for ethics. Maybe the "Rationale for the study" could be moved up into this section to inform the reader at the outset what this article is about.

The background section has been revamped and the theoretical framework deleted. The paper now reports the empirical findings of secondary analysis of transcripts of experts to identify value and value conflicts associated with preconception ECS. Please refer to the background and methods sections pages 3-8.

Theoretical background

This starts out of the blue - please link to the first part of the background section and explain to the reader where you are going (e.g. provide some guidance on the structure of the article).

This sub-section is not very clear. Under the heading, value theory and theories of rights are mentioned as the main branches of ethical theory, followed by a brief description of the two. It is not clear to me what this section is supposed to achieve, as there is no conclusion as to the theoretical framework to be applied in this study and the text is merely descriptive of some theory. A very limited selection of views on values are discussed superficially with no connection to the present context. To strengthen the overall article, this section would need to be re-written, to explain to the reader which theoretical framework was used to inform the study and why.

We agree with the reviewer. This section has been deleted and some parts incorporated in the background. Please refer to the background section pages 3 & 4.

Methods

In the methods sections, I am missing information on how the theory was used to inform the study. While there is sufficient information on participant selection, no explanation is provided about the role of theory in the study. The reference to Bogner and Menz is not sufficient to explain the methodological approach to the study. A brief explanation of what theory-generating expert interviews are, and why this approach has been chosen, would be helpful.

The methods section now explains in more details how data collection was performed. We have utilized systemizing expert interviews not theory generating type. This has been clarified in the methods sections under subheading Expert interviews page 5 lines 127 to 136.
Table 1 seems unnecessary, given that the same information is provided in the main text.

This has been deleted.

Could you provide the Swedish names of the organisations in parentheses, so they can be more easily identified (esp. the National Board for Health and Welfare) or provide links to their respective websites?

The Swedish names have been provided in the manuscript now page 6 lines 145-150.

Analysis

Please provide information on the set of values obtained after a thorough literature review. How was the literature review conducted (e.g. discipline, search terms, etc.), what were the results?

We have not conducted a systematic literature review, we have just referred to literature to identify the definitions of common values such as respects for persons, solidarity in order to identify relevant texts in the transcripts and discuss our results in the discussion section. For example, respect of persons include privacy, autonomy and integrity. Please refer to the analysis section, where we explained it better. Page 7 lines 168 – 176.

Results

How were the seven themes identified? Are these based on coding or on the literature review? How were the sub-themes identified - based on the authors' own views or on some theory?

Respondents stated the values by name such as autonomy, human dignity, solidarity, integrity, privacy, justice. The themes and categories and subcategories have been identified by coding. The categorization was deductively inspired from literature as explained in the analysis section. It did not depend on some theory. In the results sections we have delineated the major themes, categories and subcategories by titles and subtitles now pages 8 -14.

This section would benefit from more structure, which may of course be challenging, given that values are not always clear and may overlap. Given that one key aim of the paper is to discuss the value conflicts that may arise in ECS, it might be an idea to have a dedicated section on value conflicts.

The results have been changed into values and value conflicts as major themes, then categories and subcategories. Under the major theme values encompassed the values and its subgroups. Please refer to result section and the revised figure one, which show the results.
One issue I have with this section is that the values are not explained or defined, either based on a theoretical approach or with an explanation from the authors on why they chose to group certain sub-themes together under one theme. The approach is also not consistent - e.g. under "solidarity" a comment on "human dignity" is provided, which is merely equated with equality. Can justice, equality and social care really be subsumed under solidarity - if we don't even know what is meant by solidarity in the first place? To make sense of these findings, it is important to understand the rationale behind the themes / sub-themes. Concerns for the disabled are also mentioned under solidarity and human dignity, and it is not clear how the categorisation was made.

Health as a value seems puzzling, as several things are subsumed under the heading, all of which would also fit at least under one other value.

The results section has been revamped and we have tried to justify the selection of the categories and sub categories, one justification is, they were mentioned by respondents and the other relied on literature describing values and what it constitutes, thirdly some values and text, according to our views, seemed to make sense to be grouped together per the coding process. We explain that in the analysis and results section.

Discussion

This first part of this section is helpful by setting out the Swedish context, which helps to explain the findings in the study. What I would like to see, however, is a more structured approach to the rest of this section. In particular, some signposting might help, e.g. an explanation of the challenges associated with trying to identify distinct values in such a study. This section is largely descriptive and some of the information would probably be better suited to the background section. Also, no critical literature is cited - notions such as "human dignity" and "solidarity" are very controversial in bioethics, but this is not even acknowledge. While a full literature review is not necessary for present purposes, it would be appropriate to start off by saying that the value conflicts expressed by the interviewees is echoed by the academic debate on the relevance / existence of certain values. No link is made to the theoretical framework in the earlier part of the article. Value theory is mentioned in the context of health, but is not applied to any other values - why? Finally, what is the analysis with regard to ECS? Going back to the aim of the study (values, weights, conflicts) - what did you find? The first point is addressed well, but the discussion of weighing between values and conflicts which may arise could be made stronger.

We have mentioned in the discussion how defining values such as solidarity and human dignity has been difficult and we presented literature that explained the different contexts and meaning some values hold and how no one meaning was sufficient. We related this to how respondents addressed these values…the value conflicts we mention, transpires from preconception ECS or
its use, it does not reflect the debate of what these values mean or do not mean in the bioethical literature. We are aware of the debate but it is outside the scope of the paper. We are not discussing the debate about the definitions of values we are describing. We have pointed out the various ways experts expressed some of the values.

The theoretical framework has been deleted and the paper is an empirical one that communicates and discusses participants’ findings.

We have stated at the onset of the discussion “Solidarity and its subcategories of equality, justice and social care, were discussed by all experts, whereas respect of persons with reference to autonomy, integrity and privacy constituted the lion’s share of reference, even if it was not mentioned by all interviewees. It can be established that both these categories were of paramount significance to our experts in addressing preconception ECS” this reflects the values and their weights.

In the last section of the discussion new text was added regarding value conflicts. kindly refer to page 19 lines 476 -485.

Conclusions

The values listed here are mentioned as the ones referred to by the experts - however, earlier it said that these arose from literature review. Is this list exhaustive or were there any others?

Please refer above to the replies regarding the literature review and the analysis of the transcripts. Hopefully it has been made clearer now in the text.