Author’s response to reviews

Title: Ethical aspects of diagnosis and interventions for children with Fetal Alcohol Spectrum Disorder (FASD) and their families

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Author’s response to reviews:

We are grateful for this opportunity to revise the manuscript and for the constructive criticism and suggestions from the reviewers and the editor. Below we present our response for each of the comments made.

EDITORIAL COMMENTS

1. Abstract: The 'background' section poses the issue that “The value for individuals being identified as having FASD remains controversial” but this issue is not addressed in the abstract's conclusion.
Our response: We have made some minor edits to the conclusions which we hope make this connection more clear.

2. Abstract: The methods section is vague - please be more specific, e.g. what type of literature review? Were the discussions formal qualitative research or deliberative engagement?

Our response: We have rewritten the methods part of the abstract. It does not solve all the description issues, but we suggest it is acceptable that we handle this properly in the methods section in the paper.

3. Introduction: Page 3: The three questions posed at the bottom of p3 need to be explicitly addressed in the conclusion

Our response: We have rewritten the conclusion to make sure all three questions are addressed.

4. Methods: Page 4: Reference [8] is an important reference, but is in Swedish. Could an English summary be included as a supplementary file to this submission?

Our response: We suggest the official SBU summary in English is included as a supplementary file; attached.

5. Methods: Lines 12-30, Page 4: The methodology of the literature search needs to be described in more detail. Was this a scoping review? A systematic review? Something else? Please state and justify your methodology.

Our response: We have now clarified and justified the methodology used.

6. Methods: Lines 31-50, Page 4: It is not clear whether the discussions described here were formal research, or something more akin to expert peer-review prior to submission. Please clarify and justify. Either way, how did you analyse the themes that emerged?

Our response: We have now clarified how the work has proceeded and how themes have been analyzed.

7. Body of discussion: These sections are largely descriptive, without enough by way of conceptual analysis or normative ethical reflection. The sections also contain empirical claims (e.g. dot points at bottom of p7). Orienting this content to show why it is explicitly ethical is required.

Our response: The main point of this paper is to identify aspects of ethical relevance, and to some extent to show how they are interrelated. We think this is a valuable contribution to this area. The aim is however more limited when it comes to conceptual analysis, and even to normative analysis. However, we have made some minor additions and reformulations in order to be more explicit about ‘the ethics’ of what we bring up.
8. Discussion: Line 57, bottom p6: reference is made here to 'need' but this concept is not defined nor is it discussed how such prioritisation might take place.

Our response: We are referring to need in a broad general sense, and have now made some minor edits to the sentence to make this point clear. We have also made an addition in the following paragraph relating (very broadly) to normal functioning.


Our response: This has been provided.

10. Limitations section: You note that you don’t weigh the different aspects presented; however you could do more to frame these issues as ethical issues – why are they normatively relevant or significant?

Our response: Point well taken, and relates to comment 7 above. In the limitations section we have added that an answer on how to weigh the different aspects presented does not only require more empirical input, but also further normative analysis.

REVIEWER 1 (JB)

I. Re: ‘ethical analyses relating to FASD had been carried out during the time period relevant to our literature search, then our search would have identified them’ I think there are a couple of papers/reports missing, one highlights something barely touched on in this this review - the role of state in prevention of FASD, and the corollary - what is the role of professionals in surveillance/state notification/policing things:

1) Larcher V, Brierley J. Fetal alcohol syndrome (FAS) and fetal alcohol spectrum disorder (FASD)-diagnosis and moral policing; an ethical dilemma for paediatricians. Arch Dis Child. 2014 Nov;99(11):969-70


Our response: We may have been overconfident, or just expressed poorly what we meant. We have made some minor edits to the sentence referred to in order to slightly modify our claim.
We have read and considered the suggested sources, but have decided not to include them in the manuscript since we have already intentionally avoided getting further into the prevention issue.

II. Given the importance given here to the benefits/risks of identifying the 'social' aetiology of the problem and the impact if this on care provision and negative connotations I wonder whether the authors ought to acknowledge the wealth of literature in similar but distinct areas - stigma in neonatal abstinence syndrome due to heroin addiction, congenital HIV etc and very speculative IVF.

Our response: We now acknowledge the parallels in the text when bringing up stigmatization as a potential problem with getting a diagnosis (page 8, under the heading Stigmatization, blame, and guilt).

REVIEWER 2 (MS)

1. This research is described as 'part of a health technology assessment'. What does that mean and what does it entail? It is said to be based on published literature as well as discussion with experts in the field and a reference group of medical ethicists'. It would be good to have some basic details about the experts in the field and the medical ethicists and the numbers taking part? What role did these groups have and what form did the discussions take? Was this formal research or informal discussions? Or, are the groups described actually the authors of the paper? Please clarify.

Our response: We have rewritten the methods section to be clearer about this. The members of the reference group are listed in the acknowledgment.

2. Given the lack of detail, it is problematic that the discussion does not distinguish between what is in the literature and what comes from discussions with the expert group or the reference group e.g. p. 5 lists 'potential advantages of FASD-related diagnostics', citing a group of four references covering eight dot points that follow. Individual dot points are not referenced.

Our response: We have changed the referencing style here to increase precision, and updated some of the references.

3. It would be good to have some information about the inclusion and exclusion criteria resulting in 39 articles from the 430 potentially relevant abstracts that were included in the ethical analysis.

Our response: We have now made a brief addition, recognizing that there was no formal procedure for inclusion and exclusion (other than what is now mentioned).

4. What is the SBU council?

Our response: This has now been explained.
5. What is the FASD project group? What is its connection to this paper?

Our response: What the FASD project group is has now been explained indirectly by the rewritten beginning of the methods section. The members of the FASD project group who contributed substantially to this work are also co-authors of the paper. We have now clarified this under Authors’ contributions.

6. Please explain the 'guidance for systematic identification of ethical aspects of healthcare technologies' that was 'used as an additional support'.

Our response: We have changed “guidance” to “check-list” and hope that is more self-explanatory. A reference is provided to the relevant document, which is published.

7. The paper's conclusion is somewhat imprecise. It could be more explicit as it is in the abstract.

Our response: Thanks for pointing this out. We have modified the conclusion, also in an attempt to return to the original three questions stated early in the paper.