Author's response to reviews

Title: Fetal alcohol spectrum disorder: development of consensus referral criteria for specialist diagnostic assessment in Australia

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Author's response to reviews: see over
Dear Editor,

Re ‘Fetal alcohol spectrum disorder: development of consensus referral criteria for specialist diagnostic assessment in Australia’

Thank you for the constructive reviews of our manuscript. We have revised the manuscript based on the reviewer’s comments as detailed below and have uploaded a revised version.

Reviewer: Ju Lee Oei

Minor Revisions

1. How were respondents contacted prior to the questionnaire being sent? If they had agreed initially to answering the questionnaire, it is disappointing that only 75% of them responded.

We have expanded our description of the study methods under the recruitment subheading to indicate that panel members who were recruited based on their previous report of a case of FAS to the Australian Paediatric Surveillance Unit (APSU) were passively recruited into the study and were least likely to respond to the survey: “Individuals passively enrolled via the APSU were advised of the study prior to survey administration, and were only removed from the panel if they declined to participate (17/57). In contrast, participants who were recruited via the steering group or professional organisations had all actively indicated their intention to participate. Consistent with these differences in recruitment, individuals who were passively recruited via the APSU were less likely to respond to the survey (67.5%) compared with participants who were recruited through professional bodies (71.0%) or the study steering group (79.4%).” (p6, last para)

We have also expanded our discussion of limitations to acknowledge the potential for response bias: “Response to the survey was lowest among participants who were passively recruited through the APSU. Although the overall level of response exceeded the 70% recommended level (Sumison 1998) and is comparable to that reported among similar Delphi studies of health professionals (Myers 2010, Downs 2009), non-response may have influenced the survey findings.” (page 18 para 2).

2. How were the international respondents chosen as this was a paper designed to test Australian opinions?

International respondents were included in the survey based on specific recommendations from consumer and community participants to include assessment of the opinions of recognised international experts in the field. We have added further detail about recruitment of international respondents in the methods section: “ … international experts in the field based on the recommendation of consumer and community participants…” (page 6, last para).
International participant responses were retained in this analysis as we found no substantial differences in perceptions between the international and Australian respondents. The survey evidence based on Australian and international expert opinion was then considered by panel members at the workshop. We have added a statement that the survey findings did not differ substantially between the Australian and international respondents: “Responses of the 7 international participants did not differ substantially from those of the 83 Australian participants, and due to the small number of international participants, findings are reported collectively.” (page 9 para 3).

3. This paper assesses "expert opinion". Alcohol is very much ingrained into Australian culture. Would a survey of coal-face workers, i.e. people who do not have recognised expertise in FAS be appropriate as children can usually only get referred to “experts” if people on the ground recognise the problem in the first place.

We agree that the perceptions of health professionals who do not have recognised expertise in FAS are critical to successful service provision. Surveys on the knowledge, attitudes and practice of health professionals in Western Australia with respect to alcohol and pregnancy and FAS have been completed (Payne et al. 2005, Australian and New Zealand Journal of Public Health, 29(6), 558-564) as well as evaluation of an intervention to improve awareness (Payne et al. 2011, Paediatric and Perinatal Epidemiology, 25(4), 316-27). This remains an issue which is highly relevant to successful implementation. We have added the following text to the discussion to highlight the importance of this issue and the need for a comprehensive implementation strategy: “The successful implementation of referral criteria will depend upon the ability of services providers who do not have specific expertise in FASD to recognise the issue, be aware of the specialist services available for diagnosis and management, and of the potential benefits of referral.” (page 19 para 2).

Reviewer: Liza Edmonds

1. In the methods section, under recruitment, 139 people agreed to participate. How many people were asked to participate? I could not see this in the results section either. Also 13 out of 17 people attended the consensus workshop – what happened to the other 4 did they only attend part of it? Again I could not see this in the results section.

We have added information on the number of individuals invited to participate in the study on page 6, last paragraph, and page 7 first paragraph (n=57, n=128, n=35) in addition to the following: “Of the 220 individuals invited to participate in the survey, 81 either did not respond to the email invitation or declined to participate prior to the survey administration and were excluded from the study.”

Four panel members were unable to attend the face-to-face consensus workshop, and we have added further information on the participation of panel members in recommendation development on page 8 para 4: “Panel members met monthly by teleconference prior to the face-to-face workshop to provide input on study design, and to review and oversee the collection and evaluation of evidence to be considered in recommendation development. Only 13 of the 17 panel members were able to attend the 2-day workshop at which consensus criteria for referral were developed; however, all panel members participated in the review of workshop outcomes and subsequent final recommendation development.”

2. In the results section, under criteria for prenatal alcohol exposure in paragraph 2 it was noted that "9 respondents indicated support." out of how many respondents were the 9? was it 9/17 or 9/139? I think it would be useful for the readers to know this so that they can interpret your findings.
We have added further information about the denominators for respondents commenting on criteria for prenatal alcohol exposure on page 10 para 3: Among the 29 survey participants who commented on the referral criteria for prenatal alcohol exposure, 9 indicated support …”. We also added the following to page 11 para 6: “Among the 14 participants who commented on the other criteria for referral assessed, …”.

3. Also in the workshop findings results section - can I suggest that it be reviewed as I found it hard to read. While I realise it is hard to condense information on a large amount of material, it might be more reader friendly to review this section.

We have reviewed the workshop recommendations section and edited the presentation in several places (pages 12 through 13) in an attempt to improve readability while retaining the coverage of findings.

4. In the discussion section I could not the discussion around the strengths and limitations of this study. While I realise that the response rate was reasonable for surveys - it was still relatively low. This might be acceptable in this situation but I think it is still worthy of discussion. Other potential discussion points include that not all of the consensus panel were able to attend the full 2 days, that it was a select group of individuals who attended this work shop etc (potential selection bias).

We have added further discussion on the limitations of the study, including response on page 18 para 2 as detailed for reviewer Oei points 1 and 3. In addition, we also acknowledged that not all panel members were able to attend for the 2-day face-to-face consensus development workshop; however these respondents were engaged in reviewing evidence and outputs prior to and following the workshop and in formulating the final recommendations: “Although not all panel members were able to attend the 2-day face-to-face consensus development workshop, all panel members were engaged in reviewing evidence and outputs prior to and following the workshop and in formulating the final recommendations.” (page 19 para 1).

5. I am sure that the authors would have ensured that indigenous peoples were included on the panel but this was not mentioned within the paper or methodology. I think if acknowledgement of such input is important and would demonstrate the authors’ cultural awareness of such important issues for most first nation communities internationally.

Thank you - we have reviewed the text on page 8 para 4 to include the text “… and three of the panel members were Indigenous.”

Reviewer: Michela Jane Angela Morleo

Major compulsory revisions

1. The abstract needs revisiting. It should standalone, i.e. make sense in isolation. The workshop participants are mentioned in the results but not the methodology. The conclusion does not lead on from the rest of the abstract. “Statements that described other criteria for referral…” is too vague. “...similar to those recommended in Canada” but Canadian guidelines have not been previously mentioned in the abstract. Also additional details are needed, how many health professionals completed the survey? All %s should be out of the number who completed the survey, not the number who were invited to do so. Use exact percentages rather than more than/almost.

We have revised the abstract to address the identified issues through the additions/modifications listed below. Percentages are reported relative to the number who returned the survey.

Methods: “… reviewed by a panel of 14 investigators at a consensus …”

“… and FASD based on published recommendations for referral in North America, was sent …”

Conclusion: “… similar to those recommended in North America.”
Results: “described criteria for referral other than prenatal alcohol exposure.”
“Among the 139 health professionals who were sent the survey, 103 (74%) responded, and 90 …”
“Over 80% of respondents agreed that referral … significant prenatal alcohol exposure, defined as 7 or more standard drinks per week and at least 3 standard drinks on any one day, …”

2. I think the authors need to be clearer about why this study is needed and why it should be published in this international journal. Are the authors seeking to specifically test the applicability/validity of the guidelines for use in Australia? Why do they think the guidelines not directly applicable - this needs to be much clearer. Also, why would an international audience be interested? Would an Australian journal be more relevant?

We have highlighted in the introduction that FASD is under-recognised in Australia, that there is no formal guidance to support clinicians’ decision making in the absence of a suitable screening test, and that the development of standard criteria for referral may improve recognition and diagnosis of this disorder. In the discussion we have recommended that further studies are required to evaluate the feasibility and performance of the recommended criteria for referral.

We believe that an international audience may be interested in these findings as there is currently no international consensus on recommended referral criteria, this work is based on recommendations used internationally, and there is little published empirical evidence to guide recommendation development or the design of other strategies to improve recognition of these disorders.

3. You should include some discussion of the numbers/percentages who selected "no comment" in order that we can have some assessment of the level of expertise of the individuals involved, as a measure of validity of the survey.

We have added the following text to the discussion:
“Although only 77% of respondents reported experience in FASD screening or diagnosis, expertise relevant to the development of criteria for referral is not confined to individuals with practical experience in screening or diagnosis, and includes individuals who have non-clinical roles. The established under-recognition of FASD in Australia (Elliott, 2008) is also consistent with Australian health professionals’ limited experience in screening and diagnosis. Approximately 70% or more of survey respondents completed the questions on referral criteria, suggesting that most believed they had expertise relevant to the development of criteria for referral.” (page 18 para 3)

4. There is no obvious limitation section in the discussion. I would include some mention of no precise measure of how experienced or expert these individuals were.

We have expanded on the limitations of the study as described above under Oei point 1 and Edmonds point 4.

Minor essential revisions
5. Page 4, bottom of para 1. Who are the "high risk subgroups"?

We have added the following text on page 4 para 1: “including Indigenous populations and those in correctional and child care settings.”

6. Page 4, top of para 2. "The varied nature of FASD presentation..." Please provide more details for an audience who may not be familiar with FASD.

We have added the following text to page 4 para 2: “… with respect to both dysmorphology and neuropsychological profile.”
7. Top of page 5, how are you defining the term "drinks"?

We have added and referenced the term ‘standard’ to the text on page 5 para 1.

8. Page 5, bottom of para 1. What are the additional criteria?

We have added the following text to page 5 para 1: “… including the presence of a facial anomaly and growth deficit or central nervous system deficit, and parent or caregiver concern that their child might have FAS.”

9. Top of page 7, should this read four published criteria.

We have retained reference to 3 published criteria, as the two Canadian publications are based on the same source material. We have revised the subsequent text on page 7 para 2 to make this clearer: “These included the Canadian guidelines for the diagnosis of FASD [12, 29], the CDC guidelines for the diagnosis of FAS [14], and referral criteria used by the Washington State Fetal Alcohol Syndrome Diagnostic and Prevention Network (WA FAS DPN) [15].”

10. Page 7, second paragraph, when mentioned the 23 statements, should you refer to table 2?

We have added reference to Table 2 on page 7 para 2.

11. Page 9, first para in results. Individuals are only participants once they participate in the study. The 139 people are who were invited to the study. All percentages should be out of the people who participate, not out of the potential pool of participants.

We have clarified our description of the complex recruitment process used on page 6 last para as described above under Oei point 1 and included information on the number of individuals approached to participate prior to distribution of the survey (n=220). We have also revised our description of response at the beginning of the results section: “Of the 139 individuals who were sent the survey, 103…”


We have inserted reference to the Australian standard drink definition of 10g alcohol on page 9 para 3: “… defined as containing 10g of alcohol (Australian guidelines to reduce health risks from drinking alcohol, National Health and Medical Research Council, 2009).”

13. Table 1, avoid abbreviations in the table title, and define FAS in the footnotes as per other abbreviations.

We have made the suggested revisions to Table 1.

14. Table 2, please define standard drinks in a footnote.

We have made the suggested addition to Table 2.

Reviewer: Alison Tigg

Discretionary revisions:
1. The recruitment of the health professionals was designed to be those with expertise or experience in FASD screening or diagnosis. However, in the results only 77% of participants reported experience in FASD screening or diagnosis. Could this perhaps be clarified further?

We have added text to the discussion to clarify that direct experience in screening or diagnosis is only one form of expertise relevant to recommendation development, as described above at Morleo point 3.

2. The authors have mentioned the difference between screening and diagnosis but seem to be using the criteria for diagnosis in the screening criteria. Could they please clarify what the next step for children sent for screening would entail, i.e., details of the diagnostic process once referred.

Recommendations on the criteria used for FASD diagnosis in Australia were published in 2013 and national implementation of these recommendations, which will provide further detail on the assessment process, is planned. We have added the following text to the discussion on page 16 para 2 to clarify the recommended process: “Individuals meeting the criteria for referral should undergo comprehensive assessment by a relevant specialist medical practitioner using a multidisciplinary assessment approach (Watkins et al., 2013).

We thank all four reviewers for their insightful comments and believe that these revisions have improved the quality of the manuscript. Thank you for your consideration.

Yours sincerely,
Rochelle Watkins