Author's response to reviews

Title: High burden of otitis media in Aboriginal and non-Aboriginal children and otitis media in Aboriginal children predicted by failed otoacoustic emissions: a birth cohort study in an arid zone of Western Australia

Authors:

Deborah Lehmann (deborahl@ichr.uwa.edu.au)
Sharon Weeks (mbra6582@bigpond.net.au)
Peter Jacoby (peterj@ichr.uwa.edu.au)
Dimity Elsbury (dimityelsbury@yahoo.com.au)
Janine Finucane (janine.finucane@health.wa.gov.au)
Annette Stokes (annette_stokes@hotmail.com)
Ruth Monck (clrmonck@emerge.net.au)
Harvey Coates (harveyc@cyllene.uwa.edu.au)

Version: 2 Date: 17 April 2008

Author's response to reviews: see over
17 April 2008

To the Editor
BMC-series journals

Re MS: 1664895199174690
High burden of otitis media in Aboriginal and non-Aboriginal children and otitis media in Aboriginal children predicted by failed otoacoustic emissions: a birth cohort study in an arid zone of Western Australia Deborah Lehmann, Sharon Weeks, Peter Jacoby, Dimity Elsbury, Janine Finucane, Annette Stokes, Ruth Monck and Harvey Coates

Thank you for sending the comments from 2 reviewers. We have now addressed these below and revised the manuscript which is attached. As requested by Reviewer 2 we have also changed the title. The paper is now entitled: Absent otoacoustic emissions predict otitis media in young Aboriginal children: a birth cohort study in Aboriginal and non-Aboriginal children in an arid zone of Western Australia

Below I have addressed point-by-point the comments from reviewers.

We look forward to hearing from you soon.

Yours sincerely,

[Signature]

Principal Research Fellow
Division of Population Sciences
Telethon Institute for Child health Research

Response to reviewers’ comments:

**Reviewer 1**

Major Compulsory revisions
1. The inclusion and exclusion criteria have been stated in the first paragraph of the method.
2. A sentence describing socioeconomic, environmental and demographic differences between Aboriginal and non-Aboriginal groups has been included in the last paragraph of the introduction and in the first paragraph of the methods section we have now indicated that the socioeconomic and demographic characteristics of the study population are described elsewhere.

Minor essential revisions
3. We have now indicated in the first paragraph of the methods section that follow-up rates are reported elsewhere. It is not particularly relevant to indicate here the overall follow-up rates since the Echocheck and tympanometer became available at different times during the study. Table 1 provides useful information on the number of successful tympanometry readings (i.e. types A + B + C) undertaken in the field (by research assistants) and in the clinic (by audiologists) at different ages. The issue of
limited follow-up at the clinic is discussed in the last paragraph of section entitled ‘Strengths and limitations of the study’ in the discussion.
4. In the TEOAE section of the methods the text has been amended as follows: “Results of the test required no interpretation and were recorded as pass, fail or not valid, the last usually due to excessive environmental or subject noise.”

Discretionary revisions:
5. This is covered under limitations of the study (see 3. above) where we indicate that the limited follow-up of Aboriginal children in the clinic was a limitation to the study, resulting in fewer outcome measurements and hence reduced statistical power. We also note in the same section that the limited number of follow-up visits meant we could not follow the natural history of the disease (though this was not the primary aim of our study). But we do point out in the same section that tympanometry in the field provided important supplementary information.
6. We have referred to the limitations of low-frequency tympanometry in the methods section but nevertheless indicate there that the test is specific in this age group. A sentence has been added to the conclusions indicating that the recent availability of a screening multi-frequency tympanometer may provide a useful alternative to screening in young infants.
7. Figure legends are after the references as instructed by the journal.

Reviewer 2
We found it difficult to identify specific points this reviewer would like us to address. We attempt to respond by comments related to individual paragraphs.

“Overall strategic alignment with Arch Dis Ch and publication priority”
It would appear reviewer 2 is not aware that we have submitted to a different journal

“Some of the lessons here generalise to other aspects of health and other indigenous peoples. Whilst not to be overplayed, this can be mentioned and constitutes some claim on journal space.”
In the discussion section entitled OAE as predictor we have included a comment recommending further evaluation of OAE as a predictor in other Indigenous populations. The editors may choose to move this to the section entitled ‘Recommendations for research’.

“There is a well known high burden of ear disease in Aboriginal populations and a definite literature. Does this work add precision, generality, variety to profile, detail to profile, specific predictive risk factors, or pathogenetic explanation? For publication in Arch Dis Ch one would wish to see a clear claim on this as justification for submission, and this being largely borne out by findings. Etc etc.”

We agree that there is a good body of literature indicating high rates of OM in Aboriginal populations, though most data documenting very high rates of OM are from remote communities in the Northern Territory of Australia. However, in the 2nd, 3rd, 4th and 5th paragraphs of the introduction we have outlined data available to date, what data are missing i.e. namely no population-based Australian data for non-Aboriginal children in the past 30 years, sparse urban Aboriginal data, no information on prevalence of failed OAE or OAE as a predictor in Indigenous people other than a recent American study and no data on OAE as a predictor in early infancy after the neonatal period (this has now been clarified in paragraph 4 of the introduction) and in the final paragraph of the introduction we have
presented the rationale behind the present submitted paper and the study as a whole. We have commented under “strengths and limitations of the study” in the discussion on the novelty of our findings.

We have altered the title of the paper.

“It is well known that the otoacoustic emission technique (OAE) is sensitive to fluid in the middle ear as well as to malfunction of the inner-ear hair cells (PCHI). It is known but less widely appreciated that, given the difficulties over non-standard (high-frequency) tympanometry, if it is appropriate from a public health (PH) point of view that a screen for otitis media and/or inner-ear hearing loss be carried out in the very young (say below 1 year), then OAE becomes consequently a more suitable technique than tympanometry etc etc.”

These issues have been addressed in the paper, particularly following revision requested by Reviewer 1.

(a) In the introduction we have reported on available literature.
(b) With regard to the 2nd sentence in this paragraph by reviewer 2, we are considering OAE assessment (and not tympanometry as a public health measure) and have discussed the reservations about tympanometry in young children (and made some revisions to the text in response to comments by reviewer 1)

Again the reviewer states the incorrect journal to which the paper has been submitted.

“The problem then divides into two sub-problems. Is a screen so early merited overall on PH grounds ? There is not a large evidence base on this question specific to aboriginals , but I am prepared to accept an a priori epidemiological case that where a rural setting with scattered population does not permit universal neonatal screening in large concentrated maternity units, a combined screen for OM and PCHI a bit later in the first year is a sensible proposition. The idea of combination points directly to use of OAE, but of course a clinical pathway with a diagnostic test potentially dividing into 2 distinct paths. Again, this could be more succinctly and clearly stated than it is.”

(a) We have indicated in the discussion and in our conclusions the public health importance of screening in view of the early onset and silent nature of the disease.
(b) We have indicated that this is not a scattered but an urban population. Furthermore, we have pointed out the importance of access to audiology services for referral which would preclude very remote areas (item 3. under ‘Recommendations for research’ in Discussion section). We indicate optimal use of OAE in the second month of life rather than neonatal screening in a maternity unit as implied by reviewer 2.
(c) In the discussion under “Recommendations for surveillance” we point out the public health importance of the proposed screening schedule.

“The second sub-problem is the evaluation of a field model for a real-world screen, which is broad and pragmatic (perhaps including total service costings) and how far towards that the present work goes. The Conclusions recommend precisely such a thing, but also contain some statements about the capital costs (only) of different equipments. These should not be in the Conclusions but in the background justification for the work. The nature and magnitude of the step that
has been made by the present study towards the recommended wider evaluation is not the subject of a very clear claim. It does not seem to be the operational validation of a technology (for which there is not much need, anyway). Rather it seems to be a documentation of the yield, acknowledged to be less than in some other Aboriginal populations (or at least studies), but material nevertheless. etc etc"

(a) It does not seem appropriate to comment on costs of equipment in the background since we had not yet undertaken a study and did not know what outcomes we would find.  
(b) We agree that an economic evaluation of the proposed screening schedule would be important. The 3rd paragraph of the section of the discussion entitled “Recommendations for surveillance” has been amended: “Such an evaluation should consider detection and referral rates, ……. as well as an economic evaluation of the program."

“There is apparently an interesting cross-over whereby the hazard ratio (the legend to Table 4, should re-state for what) is higher in the very youngest of non-aboriginals but reverts later to having the expected higher value in aboriginals. The implication of this may be that for the aboriginal population a very early screen is unsuitable.”

In our recommendations for surveillance in the discussion section we have written: “An optimal screening program in infants should include newborn hearing screening, followed by otoacoustic emission testing at age 1-2 months to identify children at increased risk of subsequent OM, and then audiometry and tympanometry between the ages of 6 and 12 months.” If universal newborn hearing screening becomes available at a particular location then, irrespective of any other reason, Aboriginal children should also be screened. However, given the high pass rate in both Aboriginal and non-Aboriginal children, we have indicated that screening for OM is best done at age 1-2 months.

“However we are not given the appropriate test for an interaction with age (is the age-difference in HR itself significant – statistical advice needed here on whether to test for the interaction in the logistic with a continuous or dichotomised age – I would start by doing continuous for scientific power and only dichotomise for representation and optimal action). This crossover idea only makes it into the conclusions in a very partial and implicit way via an absolute statement about screening at later age.”

There were very few OAE failures amongst non-Aboriginal children and hence the confidence intervals for the hazard ratios are wide. This was also the case for Aboriginal children aged <1 month. We don’t think it is useful to examine discrepancies between point estimates which aren’t statistically significant. However the OAE hazard ratio for subsequent OM was significantly greater than 1 amongst Aboriginal children aged 1-2m and it is this result which forms the basis of the recommendations in the paper.

“Recommendation I would be very prepared to more detailed work on points in the text of a resubmission but this is premature and would not be a good use of time. The authors need to decide more clearly what are the few most important ideas their results most directly speak to and to re-submit a shortened and conceptually clarified version. They should in this be mindful throughout of what would go in the traditional “BMJ Boxes”. (What was known on this topic, what this study
The introduction should succinctly cover the first of these and address the PH desirability, potential health-technology promise etc of the second, and then clearly list the questions that the results (may) answer. There are too many cathedratic professional opinion statements about need. I don’t disagree that there are needs, but these need to be converted into statements of morbidity, prevalence, severity and consequences, and potential for cost-effective treatment or secondary prevention.”

The reviewer is apparently not aware that we have submitted to an online journal (for which we are paying!). We are reluctant to shorten the current paper unless the editors request this. We feel we have succinctly explained the rationale behind the study in the introduction, the methods and the results (with no duplication between text and tables or figures) and we have provided a comprehensive discussion. We note other articles in your journal of similar length and such articles have similar headings in the discussion. The statements with regard to ‘need’ are there intentionally since interventions to reduce the high burden of disease are urgently needed.

“Middle-level point
The recommendations for research are sensible but far too long and too broad for the main material of the article, unnecessarily adding to the impression of clutter. Failure of 7-valent pneumococcal vaccination to remedy the OM problems of Aborigines is an important point, but it should go only briefly in the introduction as a strategic justification of the continuing relevance of screening-treatment approach, as the substance of the article is not bacteriological.”

We are proposing areas of research addressing the high rates of disease documented in this study and such recommendations are inevitably broad. Details of efficacy of pneumococcal vaccines would not be appropriate in the background since it is not relevant to the rationale behind the study. We have therefore not altered the section on pneumococcal vaccine research.

Additional changes:
Reference number 18 has been updated as it is now published.