

PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

ARTICLE DETAILS

TITLE (PROVISIONAL)	Universal Newborn Hearing Screening in South Africa - a single centre study
AUTHORS	Gina, Ayanda Bednarczuk, Nadja F Jayawardena, Asitha Rea, Peter Arshad, Qadeer Saman, Yougan

VERSION 1 – REVIEW

REVIEWER	Reviewer name: Dr. selvarani moodley Institution and Country: not applicable Competing interests: none
REVIEW RETURNED	24-Dec-2020

GENERAL COMMENTS	There are grammar errors and run-on sentences in the manuscript. IT needs to be carefully edited. There is no paediatric audiologist specialisation in South Africa. this needs to be changed to reflect that this is an audiologist with experience in paediatric audiology. The costs in terms of AABR equipment as well as consumables, in addition to possible repair costs should be acknowledged as a possible negative.
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REVIEWER	Reviewer name: Dr. Lucretia Petersen Institution and Country: University of Cape Town, Health and Rehabilitation Sciences, F45 Old Main Building, Groote Schuur Hospital, Observatory, Western Cape, 7130, South Africa Competing interests: none
REVIEW RETURNED	22-Jan-2021

GENERAL COMMENTS	The authors used AABR as the gold standard to determine the sensitivity and specificity values for transient evoked otoacoustic emission screening. This gold standard also has limitations, as pointed out by Levit et al. (2016), for example. In their study 25% of children with positive OAE results and negative AABR would have been missed if only AABR was used as a screening tool. Thus, to determine the true sensitivity and specificity, diagnostic audiological test results would be more useful. The high false positive rate with TEOAEs is expected, seeing that the babies were screened within less than 6 hours after birth. I suggest the authors motivate why it is crucial to screen babies within these 6 hours after birth to help justify their choice. Minor comments: (1) Line 12 "...utilisation and implementation ..." (2) Line 41 "...Amajuba district, KwaZulu-Natal, South Africa..."
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VERSION 1 – AUTHOR RESPONSE

Our response is pasted below and can also be found in the attached document:

We thank the Editor and the 2 reviewers for their insightful comments and overall positive assessment of our manuscript. Please find below, a point by point response to each of the criticisms raised.

Reviewer: 1

We thank reviewer No.1 for their comments. Please find our detailed response to each of the raised comments below:

Q1. There are grammar errors and run-on sentences in the manuscript. It needs to be carefully edited.

R1. We have now shortened some of the sentences to avoid running on. Please see the revised manuscript for these changes,

Q2. There is no pediatric audiologist specialization in South Africa. this needs to be changed to reflect that this is an audiologist with experience in pediatric audiology.

R2. We agree with the reviewer and have made the requested change. Please see below for this edit, or alternatively please refer to page 5 paragraph 1 of the revised manuscript.

"All testing was performed by an audiologist, with expertise in pediatric testing (AG), assisted by two research nurses. "

Q3. The costs in terms of AABR equipment as well as consumables, in addition to possible repair costs should be acknowledged as a possible negative.

R3. We have now included this as an association limitation of AABR. Please find this change on page 5 paragraph 4 of the revised manuscript, or alternatively see below.

"Although, the cost-base of ABR testing is higher, when factoring in not only the equipment costs but also the costs associated with consumables and maintenance, this can effectively be mitigated against by the volume of avoidable referrals."

Reviewer: 2

We thank reviewer No.2 for their comments. Please find our detailed response to each of the raised comments below:

Q1. The authors used AABR as the gold standard to determine the sensitivity and specificity values for transient evoked otoacoustic emission screening. This gold standard also has limitations, as pointed out by Levit et al. (2016), for example. In their study 25% of children with positive OAE results and negative AABR would have been missed if only AABR was used as a screening tool. Thus, to determine the true sensitivity and specificity, diagnostic audiological test results would be more useful.

R1. The reviewer is correct that a more accurate sensitivity and specificity measures would have been obtained if we implemented the diagnostic as opposed to the screening ABR. However, such an approach would have been difficult to achieve in this setting. We now acknowledge this limitation by deleting the calculated values of sensitivity and specificity and replaced it by simply stating that OAE testing would have resulted in most of babies needing referral – in accordance with the Editors comment below. This change can be found on page 5 paragraph 3 of the revised manuscript, or alternatively please see below for this change.

"We observed that in neonates screened with ABR, 2120 babies passed the test, and 149 of the cohort failed the screening. Contrastingly, the OAE test passed only 655 babies in the cohort and failed 1614

babies. Accordingly, OAE testing would have resulted in most babies needing referral."

Q2. The high false positive rate with TEOAEs is expected, seeing that the babies were screened within less than 6 hours after birth. I suggest the authors motivate why it is crucial to screen babies within these 6 hours after birth to help justify their choice.

R2. We thank the reviewer for raising this point which is a crucial reflection within the paper. In high-income settings where children tend to be discharged later, this is clearly not as significant an issue, as is underlined as a principle in the 2019 position paper by the Joint committee on Infant Hearing that encourages all infants to be screened before discharge.

Healthy neonates in South Africa on the other hand, are typically discharged 6 hours after birth according to South African Government "Guidelines for Maternity Care in South Africa" (Chapter 17 Page 136):

<https://www.knowledgehub.org.za/elibrary/guidelines-maternity-care-south-africa-2016>

Where the child failed the initial screen, we had significant difficulty getting families to return for rescreening/diagnostic tests, therefore a newborn hearing screening service is most likely to be successful with screening prior to hospital discharge, particularly as South Africa seems to centralize Maternity services with purpose built hospitals.

This paper, and this problem highlights the need for contextually relevant screening as a prerequisite to engage all stakeholders including the families, Government services and clinicians, in order for such programs to be deemed as viable. A lack of pragmatism at this level has perhaps led to a significant delay in the initiation of screening program.

We now discuss this on page 5 paragraph 4 of the revised manuscript. Alternatively please see below for these changes:

We thus highlight the need for contextually relevant screening as a prerequisite to effectively engage all stakeholders including the families, government services and clinicians in order for such programs to be deemed viable. Ideally children need to be screened prior to discharge as the birth hospital is the ideal setting to ensure compliance. For a service to be successful in the South African context where early hospital discharge is the norm, screening programs have to adjust to ensure uptake.

Q3, Minor comments: (1) Line 12 "...utilisation and implementation ..."

(2) Line 41 "...Amajuba district, KwaZulu-Natal, South Africa..."

R3. We thank the reviewer for highlighting these typographical errors – which we have now amended in the revised manuscript.

Editor in Chief

We thank the Editor for their positive assessment of our manuscript.

Comments to the Author:

Q1. Title - amend to "Universal Newborn Hearing Screening in South Africa - a single centre study"

R1. We have made the requested change. Please refer to the title of the revised manuscript for this change.

Q2. You need to be more cautious in your conclusions as you did not include a diagnostic audiological test - see comments of reviewer 2. Better to omit the sentence "Accordingly, for transient OAE, we calculated the following values of sensitivity (87.9%), specificity (30%), positive predictive value (8.11%) and negative predictive value (97.3%)" and simply state that OAE testing would have

resulted in the majority of babies needing referring.

R2. We appreciate the Editors concern that we need to be more cautious. We have made the requested change and this can be found on page 5 paragraph 3 of the revised manuscript, or alternatively please see below.

"We observed that in neonates screened with ABR, 2120 babies passed the test, and 149 of the cohort failed the screening. Contrastingly, the OAE test passed only 655 babies in the cohort and failed 1614 babies. Accordingly, OAE testing would have resulted in most babies needing referral."

Q3. Abstract 1st sentence replace "developed world" with "high income countries"

R3. This change has been made. Please refer to the abstract section of the revised manuscript.

VERSION 2 – REVIEW

REVIEWER	Reviewer name: Dr. selvarani moodley Institution and Country: not applicable Competing interests: none
REVIEW RETURNED	16-Feb-2021
GENERAL COMMENTS	Thank you for addressing all comments as raised in in the initial review.