Validation of DPOAE screening conducted by village health workers in a rural community with real-time click evoked tele-auditory brainstem response

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List of abbreviations and acronyms:

DPOAE: Distortion Product Otoacoustic Emissions
TEOAE: Transient Evoked Otoacoustic Emission
AABR: Automated Auditory Brainstem Response
ABR: Auditory Brainstem Response
VHW: Village Health Workers

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Key words
Community-based
Grass roots worker
DPOAE screening,
Telemedicine
Tele-audiology
Tele-auditory brainstem response
Abstract

Objective: This study assessed the validity of DPOAE screening conducted by Village Health Workers (VHWs) in a rural community. Real-time click evoked tele-auditory brainstem response (tele-ABR) was used as the gold standard to establish validity. Design: A cross-sectional design was utilized to compare the results of screening by VHWs to those obtained via tele-ABR. Study samples: 119 subjects (0 to 5 years) were selected randomly from a sample of 2880 infants and young children who received DPOAE screening by VHWs. Method: Real time tele-ABR was conducted by using satellite or broadband internet connectivity at the village. An audiologist located at the tertiary care hospital conducted tele-ABR testing through a remote computing paradigm. Tele-ABR was recorded using standard recording parameters recommended for infants and young children. Wave morphology, repeatability and peak latency data were used for ABR analysis. Results: Tele-ABR and DPOAE findings were compared for 197 ears. The sensitivity of DPOAE screening conducted by the VHW was 75%, and specificity was 91%. The negative and positive predictive values were 98.8% and 27.2% respectively. Conclusion: The validity of DPOAE screening conducted by trained VHW was acceptable. This study supports the engagement of grass-root workers in community-based hearing health care provision.

Background

A number of alternative models have been implemented in developing countries to meet the needs of individuals unable to access traditional health care services (Olusanya, 2010). Likewise alternative models may be required to provide infant hearing screening for rural
locations where births occur at home or in local primary health clinics instead of traditional hospital settings (Swanepoel, Louw & Hugo, 2007). In order to address this concern, several countries including Bangladesh, Nigeria, Taiwan, Netherlands, U.K and India, have explored innovative community-based infant hearing screening programs (Owen et al, 2001; Lin et al, 2004; Neumann et al, 2006 as cited in WHO-SEARO, 2009).

In hospital-based programs often audiologists or support personnel such as trained nurses conduct hearing screening (White, 2008). In contrast, community-based programs have engaged health visitors or community health workers to conduct screening at babies’ homes or in community clinics and primary health centres (Owen et al, 2001; Danhauer et al, 2006; Olusanya et al, 2008). This task shifting is considered an acceptable practice (Hansen & Tobler, 2008). Availability of handheld objective hearing screening tools such as distortion product otoacoustic emissions (DPOAE), transient evoked otoacoustic emissions (TEOAE) and automated auditory brainstem response (AABR) systems has increased the possibility of such task shifting. These procedures are considered viable infant hearing screening methods by the Joint Committee of Infant Hearing (JCIH, 2007). Although these procedures are thoroughly validated, there is little doubt that the screener’s skill in obtaining appropriate responses is imperative for valid infant screening outcomes. Indeed, researchers have noted that the learning curve is high in respect to hearing screening skills and that the quality of referral rates change based on screener skills (Vohr et al, 1998; De Ceulaer et al, 1999; Lemons et al, 2002; Benito-Orejas et al, 2008). To date community-based program’s effectiveness has focused on feasibility or qualitative outcomes such as coverage, refer rate and follow-up rate (Bantock & Croxson, 1998; McPherson et al, 1998; Owen et al, 2001; Olusanya et al, 2007; Friderichs, Swanepoel & Hall, 2012; Kock, Swanepoel & Hall, 2016).
Both hospital-based (Freitas et al, 2009; Lim, Kim & Chung, 2012), and community-based programs (Olusanya et al, 2008) have validated screening program effectiveness using data derived from diagnostic testing of only those babies who refer screening. However, some research exists where initial hearing screening outcomes were validated by using the AABR as the “gold standard” measure. For example, in Korea, Lim et al (2012), screened 10,879 neonates using AABR results and further reported positive predictive values alone based on diagnostic click ABR results of those who failed the screening. In Brazil, Freitas et al, 2009, estimated the specificity and the false-positive rates of TEOAE and AABR on 200 neonates screened by nurses, by evaluating one screening test with the other. In a community-based infant hearing screening for early detection of permanent hearing loss in Lagos, Nigeria, trained community health workers conducted TEOAE and AABR screening. The efficiency of first-stage screening was assessed by subjecting every tenth baby who passed to a second-stage screening with AABR (Olusanya et al, 2008).

The purpose of this study was to validate the screening conducted by VHWs to evaluate the quality of such a community-based approach, as this program is the first known attempt in rural villages in India. In the state of Tamil Nadu, in South of India, the authors conducted community-based hearing screening program for infants and young children up to 5 years of age. This geographical area was chosen as it is located in rural southeast India and consequently warrants an innovative hearing screening model to provide consumer services.

In this program, trained VHWs conducted DPOAE screening at the child’s home. A two-stage screening protocol was adopted in which a “did not pass” screening outcome in first screen was followed by re-screen in two weeks. Real-time click evoked tele-Auditory Brainstem Response (tele-ABR) evaluation was administered to confirm threshold (Baldwin & Watkin, 2014; Steinhoff et al, 1988).
The practical challenge in determining sensitivity and specificity is in completing follow-up evaluations to confirm hearing loss on all newborns after the hearing screening (Davis, Bamford & Stevens, 2001; Hall, Smith & Popelka, 2004). While traditional ABR was difficult to provide to all infants in the rural location, ABR could be provided via tele-technology by audiologists located in Chennai city, India. Use of tele-ABR in this study reduced the need for long, costly and unreliable travel by the patient. Consequently, tele-ABR was incorporated to provide cross-check of screening outcomes in this study. Hence, tele-ABR was essential to demonstrate the VHWs effectiveness for screening infants and young children. Tele-ABR was validated on infants prior to the commencement of the program (Ramkumar, Hall, Nagarajan, Shankarnarayanan & Kumaravelu, 2013).

Method
This study was approved by the Institutional Ethics Committee of Sri Ramachandra University

Sample
One hundred and nineteen children were selected using random sampling from 2880 infants and young children under the age of 5 years who had received DPOAE screening by VHWs. Informed consent was obtained from the parents.

Procedure

DPOAE screening instrument settings
Two portable GSI Audioscreener+ hand-held devices, which display automatic screening results as “pass” or “refer”, were used for DPOAE screening. The DPOAE screening in this study was piloted in the community on 14 ears of children to check responses in frequencies
between 2 to 6 kHz in a home environment. A digital sound level meter (Lutron SL-4001) was used in ‘A’ frequency weighting and ‘fast’ time weighting network to measure ambient noise levels. The sound level meter had the capability of measuring between the frequency range of 31 Hz to 8000 Hz. Consequently, the DPOAE screening was conducted using the “noisy” environment setting. A “pass” response could be obtained at 2, 3 and 4 kHz when the maximum ambient noise levels were within 50 dBA.

Based on the pilot study, the DPOAE test frequencies were set as 2, 3, and 4 kHz. The DPOAE stimulus intensity level was 55 dB SPL for L2 and 65 dB SPL for L1. The automated algorithm for ‘pass’ criteria was 6 dB SNR at two out of the three frequencies. The test was terminated automatically once the results were obtained. The instrument also displayed warnings if ambient noise levels were high as ‘noisy’ and the DPOAE probe fit error was automatically displayed as ‘the probe is not completely sealed, please adjust probe’. VHWs were trained to read and respond to these messages appropriately.

**Real-time click evoked tele-ABR**

The children who were selected by random sampling were assessed by tele-ABR as the gold standard test. Tele-testing was conducted using satellite as well as broad band internet connectivity, depending on the availability of network. The investigator was blind to the results of screening at the time of tele-ABR testing.

Tele-ABR was conducted in a mobile van equipped with a bed, air-conditioner and power supply, or in a space provided in a nongovernmental organisation’s rural women’s social empowerment centre. Testing was conducted in real time by an audiologist at the tertiary care hospital in Chennai, India, using satellite connectivity in the mobile van or using broadband
internet connectivity. Trained VHWs prepared the child for testing by placing electrodes, positioning the child and ensuring that they were asleep through the testing. A tele-technician set up equipment and established connectivity. The hospital audiologist took remote control of the ABR software. Dedicated video-conferencing was used in the mobile van and Team viewer software (version 7) was used for remote computing and video-conferencing in the centre. Though satellite connectivity was preferred due to its stable connectivity in remote locations, the satellite dish and modem in the mobile van required repairs and service. As an alternative, broadband internet connectivity was used.

The GSI Audera was used for acquiring tele-ABR waveforms. Tele-ABR results were recorded using click stimuli (0.1ms) with monaural stimulation at a rate of 33.1/sec in rarefaction polarity. If waveform morphology was poor during the hearing evaluation, the click stimulus rate was reduced to 11.1/sec. The click stimulus was presented through E.A.R. tone 3A insert transducers. When required, polarity was changed to alternating to eliminate artefacts and confirm presence of peak V. Disposable electrodes were used with a non-inverting electrode placed at high forehead level (Fz). The inverting electrode was located on the mastoid (M1, M2) of the stimulus ear and a ground electrode was located on the lower forehead (FPz). Electrode impedance was ensured to be less than 5 kohms. Testing was started at 70 dB nHL and intensity was changed in 20 dB nHL steps, 10 dB step was used to arrive at threshold. Two thousand sweeps were recorded at each intensity. Band pass filter of 30-3000 Hz was used. Filter settings were altered to eliminate frequencies up to 100 Hz, if electrical interference was noted.

Wave morphology, repeatability and peak latency, were used for ABR analysis. Presence of peak V up to 30 dB nHL was assessed. At times, the presence of power-backup generator in
Figure 1. (a) Tele-ABR of a 1.5-year-old child with bilateral normal hearing sensitivity. (b) Tele-ABR of a 2-year-old child with bilateral mild hearing loss.
the mobile van, increased ambient noise levels, resulting in poor waveforms at 30 dB nHL. In such circumstances, presence of peak V up to 40 dB nHL was considered normal for tele-ABR testing. Examples of tele-ABRs recorded is shown in Figure 1.

Analysis
The aim of the screening was to identify children with unilateral and bilateral hearing loss which is \( \geq 30 \) dB nHL (40 dB nHL, in case of high ambient noise level). The screening sensitivity, specificity, positive and negative predictive values were analyzed for the 2\(^{nd}\) stage (re-screen) screening conducted by VHWs using DPOAE.

Results
One hundred and nineteen infants and young children (76 infants, 43 young children) were screened by VHWs and assessed with tele-ABR. ABRs could not be recorded till threshold in 25 ears and ABRs could be recorded only in one ear in 16 children, as these children woke up during testing or were restless and not co-operative. Hence these ABRs were not included for analysis. In all 197 ears of infants and young children were included in the analysis. The age wise distribution of validity measures is given in Table 1.

Table 1 shows that there were no false responses up to the age of 6 months. The false positive responses increased with age. The following two-way matrix (Table 2), was obtained to assess the overall validity of DPOAE screening conducted by VHWs. All sensitivity and specificity calculations including associated confidence intervals are given in Table 3.
Table 1. Age wise data of validity of DPOAE screening

<table>
<thead>
<tr>
<th>Age range</th>
<th>Ears</th>
<th>TP</th>
<th>TN</th>
<th>FP</th>
<th>FN</th>
</tr>
</thead>
<tbody>
<tr>
<td>0-3 months</td>
<td>61</td>
<td>1</td>
<td>60</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>&gt;3-6 months</td>
<td>26</td>
<td>0</td>
<td>26</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>&gt;6 months-1 year</td>
<td>27</td>
<td>2</td>
<td>21</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>&gt;1-2 years</td>
<td>24</td>
<td>1</td>
<td>21</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>&gt;2-3 years</td>
<td>22</td>
<td>0</td>
<td>18</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>&gt;3-5 years</td>
<td>37</td>
<td>2</td>
<td>27</td>
<td>8</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>197</td>
<td>6</td>
<td>173</td>
<td>16</td>
<td>2</td>
</tr>
</tbody>
</table>

(TP= True Postive; TN= True Negative; FP= False Positive; False Negative= FN)

Table 2. Sensitivity, specificity, positive predictive value and negative predictive value of DPOAE screening

<table>
<thead>
<tr>
<th>Tele-ABR result</th>
<th>Hearing loss</th>
<th>Normal hearing</th>
</tr>
</thead>
<tbody>
<tr>
<td>DPOAE screening</td>
<td></td>
<td></td>
</tr>
<tr>
<td>result</td>
<td>False Negative</td>
<td>True Negative</td>
</tr>
<tr>
<td>Pass</td>
<td>2</td>
<td>173</td>
</tr>
<tr>
<td>Refer</td>
<td>6</td>
<td>16</td>
</tr>
<tr>
<td>Refer</td>
<td>True Positive</td>
<td>False positive</td>
</tr>
<tr>
<td>Refer</td>
<td>6</td>
<td>16</td>
</tr>
</tbody>
</table>
Table 3. Validity measures with confidence interval

<table>
<thead>
<tr>
<th>Measure</th>
<th>Value</th>
<th>Confidence interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sensitivity</td>
<td>75%</td>
<td>±6 (69% - 81%)</td>
</tr>
<tr>
<td>Specificity</td>
<td>91%</td>
<td>±4 (87% - 95%)</td>
</tr>
<tr>
<td>Positive Predictive Value</td>
<td>99%</td>
<td>±1 (98% - 100%)</td>
</tr>
<tr>
<td>Negative Predictive Value</td>
<td>27%</td>
<td>±6 (21% - 33%)</td>
</tr>
</tbody>
</table>

Table 4. Description of significant history for children with true positive

<table>
<thead>
<tr>
<th>Age of child</th>
<th>Significant history or complaint</th>
<th>DPOAE 2nd screening results</th>
<th>Follow-up ABR</th>
</tr>
</thead>
<tbody>
<tr>
<td>13 days</td>
<td>NICU admission for 7 days, low birth weight 1.900gms, pre term baby</td>
<td>Unilateral Refer</td>
<td>Unilateral Mild hearing loss</td>
</tr>
<tr>
<td>1 year</td>
<td>Family history (mother) of hearing loss, jaundice, low birth weight</td>
<td>Bilateral Refer</td>
<td>Bilateral Mild hearing loss</td>
</tr>
<tr>
<td>2 year</td>
<td>Child not speaking clearly</td>
<td>Unilateral Refer</td>
<td>Unilateral Mild to Moderate hearing loss</td>
</tr>
<tr>
<td>3 years 11 months</td>
<td>Child not speaking age appropriately</td>
<td>Unilateral Refer</td>
<td>Unilateral Mild to Moderate hearing loss</td>
</tr>
<tr>
<td>5 years</td>
<td>Family history (father) of hearing loss, NICU admission for 3 days</td>
<td>Unilateral Refer</td>
<td>Unilateral Mild to moderate hearing loss</td>
</tr>
</tbody>
</table>

1. Mild hearing loss indicate ABR thresholds up to 50 dB nHL
2. Mild to moderate hearing loss indicates ABR peak V threshold up to 60 dB nHL
Results of this study indicated that, the DPOAE screening by VHWs correctly identified 75% of all children with hearing loss (the sensitivity) including unilateral hearing loss up to mild degree. A ‘refer’ in screening by VHWs is in itself limited at confirming hearing loss (positive predictive value) and further testing must be undertaken. However, the negative predictive value (99%) suggests that as a screening test, a pass result in DPOAE screening conducted by VHW accurately identifies when a patient does not have hearing loss and the DPOAE screening correctly identifies 91% of those who do not have hearing loss (the specificity). These results support the engagement of VHW as the primary provider of infant hearing screenings.

The two ears with false negatives had mild hearing loss on tele-ABR. The 16 ears with false positives were of older children between 2 to 5 years of age. Table 4 describes the age, significant history and hearing loss of children with true positive results. It was found that all children with hearing loss had one or more significant risk factors for hearing loss. The 13 day old neonate was recommended to follow-up to rule out neuromaturational delay.

**Discussion**

The merit of a screening program lies in its validity (JCIH, 2007). Over referral, can result in unnecessary diagnostic testing which, in turn, increases costs and makes the program less viable. On the other hand, under referral can result in missing children who may potentially have hearing loss, thereby defeating the purpose of the screening program.

Even though AABRs have higher positive rate compared to DPOAE screening (de Kock et al, 2016), AABR is not feasible in the absence of minimal infrastructure such as power
supply and a computer. While some AABR instruments are battery operated, elaborate pre-
testing process was a deterrent for use by VHWs. OAE screening assesses the integrity of the outer hair cells in the cochlea and requires only a few minutes of testing. Their accuracy in the detection of hearing loss has made them a popular objective tool for screening hearing among neonates (Norton et al, 2000; Abdala & Visser-Dumont, 2001). Referral rates are lower for DPOAE than TEOAE as the influence of noise can be reduced by selecting appropriate test frequencies in DPOAE (Dhar & Hall, 2012). Hence, DPOAE screening was conducted in this program.

As VHW’s were key for hearing screening success, the selected VHWs were provided an intensive training program. VHW’s involved with this study underwent a five day training program. They were selected to conduct screening only after their competence was assessed by skill set testing administered once training was completed. Regular reviews were conducted to ensure the retention of VHW skills and knowledge. Such measures are likely to have ultimately resulted in high specificity for infant hearing screening outcomes. Specifically, the results show that the community-based hearing screening program achieved a high negative predictive value of nearly 99%, but a low positive predictive value of 27%. These results are consistent with others (Keren, Helfand, Homer, Mcphillips & Lieu, 2002; Burke, Shenton & Taylor, 2012) who reported that, the high negative predictive value and low positive predictive value of hearing screening programs, is owing to the low prevalence of congenital hearing loss (1 per 1000 births) (Davis et al, 2001; Hall et al, 2004). Though there is lack of a systematic population based study in India, data from programs conducted in South India suggests a similar trend in prevalence (Nagapoornima et al, 2007; Paul, 2016; Ramkumar, 2017). Therefore, these measures do not directly address the effectiveness of screening by VHWs.
The lower sensitivity is due to the two false negatives, found to have mild hearing loss through tele-ABR. False negative results are explained by deterioration of hearing threshold from a conductive overlay that may develop after the screening (Clemens et al, 2000). However, the latency measures found in the ABR recordings of these two ears and lack of any overt ear related symptoms did not suggest a conductive hearing loss. False negative results are explained by deterioration of hearing threshold from a conductive overlay that may develop after the screening (Clemens et al, 2000). However, the latency measures found in the ABR recordings of these two ears and lack of any overt ear related symptoms did not clearly indicate a conductive hearing loss. Delayed-onset hearing loss was not suspected since the time lapse between screening and tele-ABR was no later than 4 months. The false negative result may be attributed to the reduced sensitivity of DPOAEs to detect mild hearing loss when used as a standalone tool for infant hearing screening. This outcome has been noted previously by others who suggest that with powerful excitation with a continuous stimulus, DPOAEs are recorded even up to moderate losses (R., 2006; Harris & Probst, 2002). TEOAEs are more sensitive to milder hearing losses, however, they are less reliable in the presence of noise due to lack of frequency specificity. With DPOAEs, the lower frequency responses, which are more susceptible to noise contamination, could be excluded (Hall, 2015) and was preferred for this community-based program.

Hospital-based hearing screening programs are usually restricted to neonates, but this community-based program included children up to 5 years of age. Older children were included as they had no prior access to screening, and were still likely to benefit with rehabilitation. However, the false positive rates were higher in the older age group, possibly due to the difficulty in controlling movements in these children. The validity measures
represent the outcomes based on a wider age group. Unlike other community-based programs that are conducted in immunization clinics or primary health centres (Friderichs et al, 2012; Olusanya & Akinyemi, 2009; Olusanya et al, 2008), in this program door-to-door screening was conducted in the rural community, therefore the numbers screened in each age group is small. In this study, the 2nd screening refer rates were low, and fewer parents of children who passed screening consented for follow-up tele-ABR testing, resulting in smaller numbers in each age group. The false positive rates may vary with inclusion of more children in the younger age group.

Norton et al. (2000) used behavioural audiometric thresholds as gold standard to validate DPOAE screening at L1/L2 of 65/50 dB SPL, and the sensitivity was 88% and specificity was 83%. Several hospital-based studies using TEOAE have reported specificity of 80 to 85% and sensitivity of 90 to 99% (Keren et al, 2002; Johnson et al, 2005; Lin et al, 2004; Hall et al, 2004). Though the sensitivity of the present community-based program is lower, the specificity is higher than the hospital-based programs using TEOAE and DPOAE screening. Lower sensitivity (80%) and higher specificity (99%) have been reported in another community-based infant hearing screening program using TEOAE, where babies who passed the screening underwent AABR for sensitivity evaluation, while those with ‘refer’ underwent diagnostic evaluation (Olusanya et al, 2008). Higher sensitivity is desirable in a hearing screening program, as children with hearing loss should not be missed, this can result in economic burden (Burke et al, 2012). Therefore, an optimum trade-off between sensitivity and specificity should be achieved by selecting a suitable criterion for screening outcome.
Conclusion

Studies conducted on community-based models have often not been able to report validity using a gold standard such as ABR. This may be largely due to the unwillingness of parents to subject their children to diagnostic testing, owing to extra travel and unnecessary testing. This study presented the opportunity of using tele-ABR testing as gold standard, which was provided in close proximity to children’s homes. As a result, compliance to participate was better. Therefore, this is one of the very few studies that has included a sub-section of children who passed the screening in addition to those who were referred. The sensitivity, specificity, positive and negative predictive values obtained for the DPOAE screening conducted by VHWs in the community is acceptable and comparable to findings reported in other hospital-based studies. This study adds further to the body of literature that supports the increasing role of grass-root workers in the provision of community-based health care services (Olusanya et al, 2008).

Acknowledgements

This study would not have been possible without the support of GSI Inc. in providing the required equipment, accessories and support for repairs. Many thanks to Dr. P. Balasubramanian, Executive Director, RUWSEC for his support in the community throughout this study. We also thank Dr. Mark Krumm, Associate Professor, Music and Speech, Kent State University, Ohio, USA, for his suggestions on this manuscript.

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