

**NEWBORN HEARING SCREENING AT A COMMUNITY-BASED OBSTETRIC UNIT:
SCREENING AND DIAGNOSTIC OUTCOMES**

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ABSTRACT

Objective: Postnatal visits at community-based midwife obstetric units (MOUs) have been proposed as an alternative primary healthcare screening platform in South Africa. This study evaluated the outcomes of distortion product otoacoustic emissions (DPOAEs) and automated auditory brainstem response (AABR) screening conducted by a dedicated non-professional screener at a community-based MOU in the Western Cape, South Africa.

Methods: Universal newborn hearing screening (UNHS) at a community-based MOU was evaluated over a 16-month period. A dedicated non-professional screener was trained to follow a two-stage screening protocol targeting bilateral hearing loss. A two group comparative design was used alternating AABR (Maico MB11 BERAphone™) and DPOAE (Bio-logic AuDX I) technology on a daily basis. Infants referring the initial screen received a follow-up appointment in two days' time and were rescreened with the same technology used at their first screen. Those referring the second stage were booked for diagnostic assessments.

Results: 7452 infants were screened including 47.9% (n=3573) with DPOAE and 52.1% (n=3879) with AABR technology. Mean age at first stage screen was 6.1 days. The initial bilateral referral rate was significantly lower for AABR (4.6%) compared to DPOAE (7.0%) and dropped to 0.3% and 0.7% respectively following the second stage screenings. First rescreen and initial diagnostic follow-up rates of 90% and 92.3% were obtained for the DPOAE group and 86.6% and 90% for the AABR group. Follow-up rates showed no significant difference between technology groups. Diagnostic assessment revealed a higher prevalence rate for bilateral sensorineural hearing loss among the AABR group (1/1000) compared to the DPOAE group (0.3/1000). Screening technology had no significant influence on daily screening capacity (23 AABR/day; 24 DPOAE/day).

Conclusions: Postnatal visits at community-based MOUs create a useful platform for hearing screening and follow-up. AABR technology with negligible disposable costs provides opportunity for AABR screening to be utilised in community-based programmes. AABR screening offers lower initial referral rates and a higher true positive rate compared to DPOAE.

Keywords

Developing countries

Community-based newborn hearing screening

Midwife Obstetric Units

Distortion Product Otoacoustic Emissions

Automated Auditory Brainstem Response

Dedicated screener

Abbreviations

UNHS: Universal Newborn Hearing Screening

EHDI: Early Hearing Detection and Intervention

HPCSA: Health Professions Council of South Africa

DPOAE: Distortion Product Otoacoustic Emissions

AABR: Automated Auditory Brainstem Response

PCEHL: Permanent Congenital or Early Onset Hearing Loss

MOU: Midwife Obstetric Unit

INTRODUCTION

Infant hearing loss is the most common congenital sensory birth defect with an estimated prevalence of four to six in every 1000 live births in developing countries [1]. The necessity of early hearing detection and intervention (EHDI) to contest the detrimental consequences, both individual and societal, of permanent congenital or early-onset hearing loss (PCEHL) is widely documented [2-4]. With at least 90% of infants with PCEHL residing in the developing world [5], focus has shifted from validation of EHDI to the development of contextually feasible models of service delivery [6-7].

Although awareness of the need for EHDI in South Africa has grown, legislation requiring infant hearing screening is still lacking. National surveys in the private and public healthcare sectors of South Africa reveal that approximately 90% of newborns have no prospect for hearing screening [8-9]. In the public health care sector, which services approximately 85% of the population, only 7.5% of hospitals offer some form of infant hearing screening whilst less than 1% offer universal screening [9]. Subsequently, the reported average age at time of diagnosis range from 23 to 44.5 months of age [10-13]. Most infants with hearing loss in South Africa do not receive early auditory stimulation which is the foundation for optimal speech and language development [6,14].

Due to the significant number of births taking place outside of hospitals, immunisation clinics have been recommended as platform for community-based infant hearing screening programmes to supplement hospital-based programmes in developing countries [5,15]. Despite initial reports verifying immunisation clinics as a useful platform for infant hearing screening [5,15], Friderichs, Swanepoel & Hall [16] reported low coverage rates mainly attributed to the use of already burdened nursing staff as screeners. To date, only one systematic government

supported community-based infant hearing screening programme has been reported at immunisation clinics in the Western Cape [16]. Friderichs et al. [16] emphasised the need for dedicated screening personnel and proposed an alternative community-based platform such as midwife obstetric units (MOUs). MOUs are birthing units run by midwives in the community for primary healthcare patients. Although discharge at these units usually happens six hours after birth if both mother and baby are in good health, they return to the MOU for postnatal follow-ups focussing on umbilical cord stump care and feeding advice [17]. A small scale study in Gauteng South Africa verified that MOU postnatal visits (also called three-day assessments) offered a practical and efficient option for hearing screening [7].

A significant challenge in implementing widespread hearing screening programmes in developing countries is the general lack of personnel [5]. The Health Professions Council of South Africa (HPCSA) position statement on EHDI programmes in South Africa (2007) states that nursing staff, community health care workers or lay volunteers can be utilised as screening personnel as long as they have received adequate training. The use of these persons as screeners is cost-effective and releases the audiologist to resume the role of programme coordinator or diagnostic specialist. However, despite these recommendations, infant hearing screening conducted by audiologists is still common practice in South Africa [7]. A few studies have investigated the use of nursing staff as screening personnel [15,16], but there are no published reports on the use of non-professional screeners. A dearth of research also exists describing the capacity of screeners, that is, the number of tests that can be performed per day or per month, for different screening technology. Information on screening capacity is essential for programming planning.

Currently, the only techniques endorsed for infant hearing screening are otoacoustic emissions (OAEs) and automated auditory brainstem responses (AABRs) [4,18]. OAEs measure outer hair

cell functioning in the cochlea and are recommended for screening in well-baby nurseries and community-based programmes [18]. OAE measurement utilising rather simple probe placement and automated 'pass/refer' criteria is feasible by non-professional screeners [15,19]. The AABR is a measure of neural synchrony in the eighth cranial nerve and lower brainstem. AABR is the technology of choice in neonatal intensive care units (NICUs). There is a higher prevalence of infants with auditory neuropathy spectrum disorder (ANSD) in the NICU population. ANSD is only detectable with a neural-based screening test such as the AABR [18].

AABR screening can also be conducted by non-professionals [5], but it is an ineffective screening tool for immunisation visits because six week old infants rarely remain in a sleeping state required for successful recordings [15]. Furthermore, AABR screening with most devices is more expensive than OAE screening especially due to increased disposable costs [20]. New generation AABR technology, such as the Maico MB11 BERAphone™, offers several advantages for more widespread application including reduced test-time, ease of use, and negligible disposable costs [21-24].

In designing the current study we posed the following research question: *How do the outcomes of infant hearing screening with DPOAE and AABR using the MB11 BERAphone™ compare when performed by a dedicated screener within a community-based MOU?*

MATERIAL AND METHODS

Study design

A two group comparative design was employed to investigate infant hearing screening outcomes at a community-based MOU. A dedicated non-professional screener alternatively performed either AABR or DPOAE hearing screenings on a daily basis. Referral rates, follow-up rates and

diagnostic outcomes were investigated for both technologies. The study was approved by the institutional review board of the University of Pretoria and the Western Cape Government: Health (WCGH) prior to the commencement of data collection.

Research context

A community-based universal newborn hearing screening (UNHS) programme was initiated at three MOUs in the metropolitan area of Cape Town (Western Cape, South Africa) as part of a government supported pilot project. MOUs are birthing units linked to Community Health Centres (CHCs). In addition, the MOUs offer antenatal and postnatal care encompassing all aspects of mother and baby health and well-being [17].

This study was conducted at the largest of the three units based on the number of births (approximately 3000 annual live births) and postnatal follow-up visits. The unit is the only MOU within the Mitchell's Plain health district that covers an area of approximately 5 000 ha with a population of 507 237. The socio-economic profile of the health district is characterised by an unemployment rate of 32% and 61% of households having a monthly income of R3 200 (\pm 229 USD) or less [25].

Study population

Infants that were born either at the MOU, at home or at surrounding hospitals, together with their mothers/caregivers, attend postnatal follow-up visits at their local community-based MOU. They often return every second day until the infant's umbilical cord has fallen off. There were no exclusion criteria in this study as all infants attending the postnatal follow-up visits were offered routine screening as part of the universal screening programme. Informed consent was obtained from each parent/caregiver prior to enrolling the infant into the study. Data collection stretched over 16 months (24 September 2012 – 31 January 2014).

Material and apparatus

The Bio-logic AuDX-I (Natus Medical Inc., CA, USA) was used for DPOAE screening. It represented the technology typically used in existing community-based screening. The pre-set DPOAE screening parameters were used including a 65/55 dB SPL stimulus intensity level for the lower f1 frequency (L1) and the higher f2 frequency (L2). Overall pass criterion was a DPOAE to noise floor difference of ≥ 6 dB for three out of four f2 test frequencies (f2 of 5, 4, 3, and 2 kHz). AABR screening was done with the MB11 BERAphone™ (MAICO Diagnostic GmbH, Germany) operated through a netbook computer and software that stored test results in a database. The MB11 BERAphone™ is a handheld headphone unit with integrated spring-mounted electrodes that only require application of electrode gel prior to placement. Screening settings included the use of the CE-Chirp stimulus™ at a rate of 93 stimuli per second and a stimulus level of 35dB nHL. The MB11 BERAphone™ was selected because of reduced disposable supply costs and preparation time.

Screening personnel

A dedicated non-specialist screener was appointed to perform hearing screening at the MOU, Monday to Friday from 7am to 3pm. The managing audiologist provided training which included a 2.5 hour theoretical session and a practical component where the screener first observed the audiologist doing screening, followed by ten supervised screens per technology. Weekly quality control and support visits continued throughout the research period. Three dedicated screeners participated in the study. Screener 1 collected the first month's data (24 September 2012 - 16 October 2012). A second screener collected data for the remainder of the period. Screener 3, the resident health promoter at the MOU, was trained to function as an auxiliary for screener 2 on sick and leave days. Both Screeners 1 and 2 were non-professionals with no formal healthcare training. They were recruited on the basis of (1) having completed high school, (2) demonstrating basic computer literacy, (3) cultural sensitivity and fluency in at least two of the

three main languages of the area, (4) comfortable with handling newborns, and most importantly, (5) possessing character traits such as patience, empathy and meticulousness yet being able to function well under pressure. The job description included providing antenatal talks, hearing screening, basic counselling, data-capturing and assistance with follow-up management.

Protocol and methods

The MOU screening programme implemented a two-stage screening protocol at primary healthcare (PHC) level to reduce the burden of false positive referrals to tertiary hospital. A bilateral refer criteria was used as criterion for an overall refer in both stages of the screening protocol. The decision does not disregard the impact of unilateral hearing loss but it was made on the basis of cost-effectiveness and practicability. This follows recommendations by the HPCSA Year 2007 Position Statement as well as former pilot research and community-based screening programmes for resource constrained areas [15,16,18].

The screener completed a test form including an informed consent form signed by the parent or caregiver, demographic information, a brief medical case history, a high-risk checklist, and screening outcome for each screening session. A bilateral screening with either DPOAE or AABR was performed. The screening technology used, DPOAE or AABR, alternated from day to day with the exception of periods of equipment breakdown. Infants with a bilateral refer outcome were referred for a second screening to coincide with their next postnatal follow-up visit or in two days' time. The follow-up screening was performed with the same screening technology as the first screening. If a second bilateral refer result was obtained the infant was referred directly to the tertiary hospital for diagnostic audiological and ear, nose and throat (ENT) services.

In instances when an infant became restless or irritable, the parent/caregiver was asked to make an attempt to feed and/or calm the baby. The screener was allowed to repeat the measurement once if the screening was terminated due to the baby's state or in an attempt to improve the probe fit for OAE measurement or impedance levels for AABR. If the screener was unable to test an infant due to restlessness, irritability or a technical fault, the outcome was treated as a 'refer' and a follow-up appointment was scheduled. Counselling with language-appropriate pamphlets regarding normal speech, language and hearing development within the first two years of a child's life was given to all parents/caregivers of participants – regardless of screening outcome.

It was not possible to have access to a room in the MOU dedicated to the screening programme during the research period. As a result, screening was conducted in five different rooms in the facility with the screener repeatedly selecting the most appropriate space. Majority of the screenings took place either in the phototherapy room (51.7%) or a student doctor bedroom (39.4%), both of which were in close proximity to the postnatal visit consulting room. As a consequence ambient noise present during screening was variable, but levels were adequate for testing according to internal equipment parameters.

Waiting times for diagnostic follow-up at the tertiary hospital often were as long as six months. In an attempt to speed up the diagnostic process, a private audiologist provided follow-up services to the first six infants who yielded a refer outcome. However, the arrangement could not be sustained throughout the study as research funding was depleted.

Coverage rates for hearing screening programmes are typically reported. We do not have accurate information on the coverage rate for the MOU screening programme during the study as none of the MOU's routinely kept indicators could be used to measure the number of hearing screenings against. The conventionally used birth statistics would not suffice as babies who

were born elsewhere could also access the postnatal follow-ups offered at the MOU. Similarly, the recorded data indicator for postnatal follow-up visits could not be used as it reflects the total amount of visits which includes multiple visits by the same mother and baby.

Data analysis

All data were captured in MS Excel 2010. Statistical package SPSS version 21.0 was used for the analysis. Descriptive statistics were applied to show basic trends in investigated variables like gender, age, referral- and follow-up rates for DPOAE and AABR screening. Inferential statistics, specifically parametric tests such as the Chi-square and t-test, were utilised to determine significance of differences between the various aspects of the two screening technologies. A significance level of 1% was applied.

RESULTS

Study sample

A total of 7452 infants (51.7% male, 48.3% female) underwent hearing screening by a dedicated screener. Of the sample, 47.9% (n=3573) were screened with DPOAE and 52.1% (n=3879) were screened with AABR. Figure 1 summarises the screening outcomes of both groups.

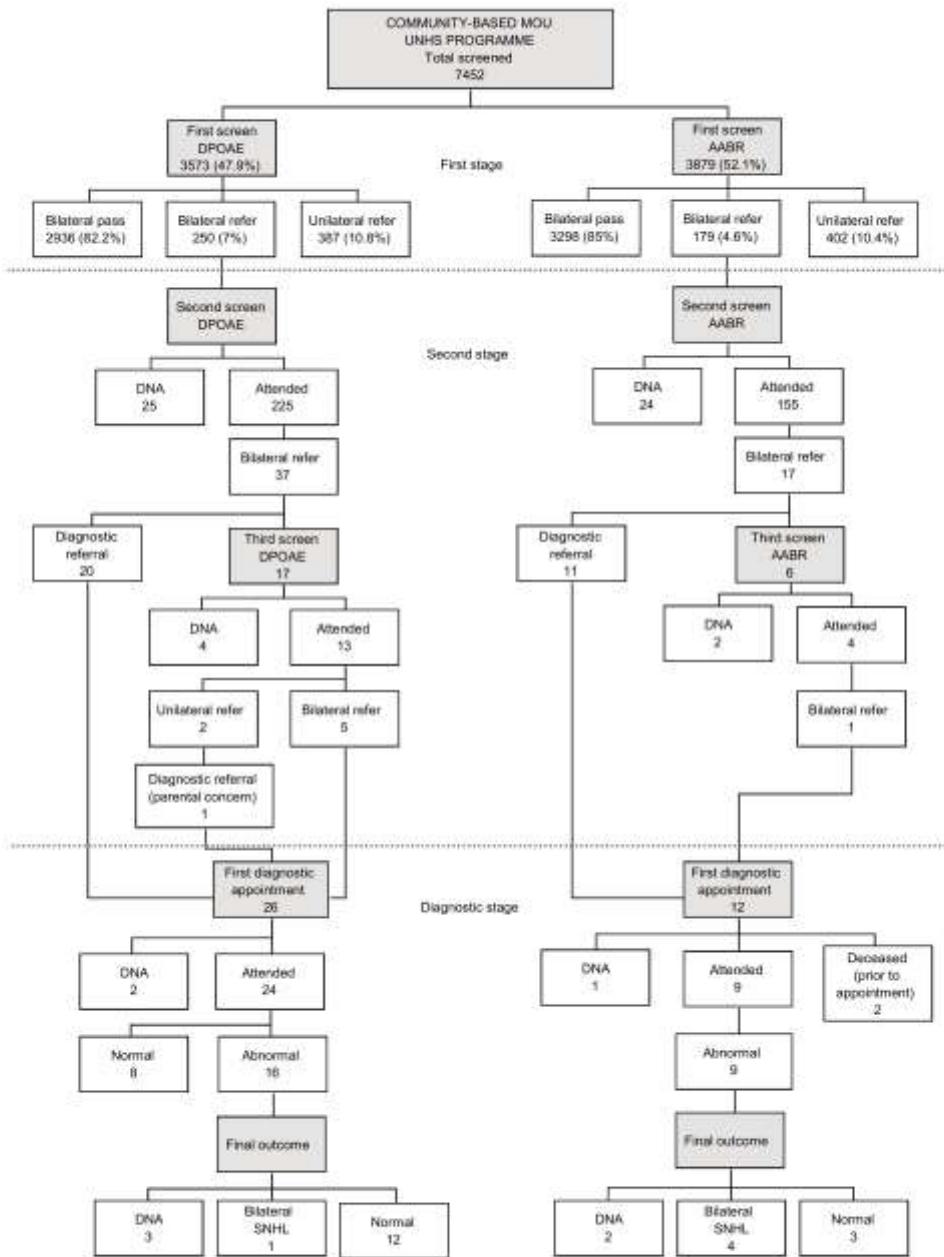


Figure 1. Overall outcome of stages within the screening and diagnostic process (*DNA = Did not attend; SNHL = Sensorineural hearing loss)

No parent or caregiver refused to consent to the screening service. Table 1 provides a demographical overview of the sample. The mean age at first stage screen was 6.1 days (SD 8.1). However, the age of hearing screening ranged from 0 to 189 days because other hospitals referred some preterm infants to the MOU for follow-up visits post discharge.

Table 1. Demographics of study sample

Description	Mean value/Percentage/Number
Birth characteristics	
<i>Mean gestational age</i>	39 weeks (SD 2.1, n=7444)
<i>Mean birth weight</i>	3076.0 gram (SD 552.1, n=7433)
<i>Normal vaginal delivery</i>	72.7% (5414/7451)
Risk factors	
<i>Preterm births^a</i>	11.3% (838/7444)
<i>Low birth weight^b</i>	13% (969/7433)
<i>> 5 days in NICU</i>	0.7% (49/7451)
Mean age of mothers/caregivers at time of first screen	26.3 years (SD 6.0, n=7414)
Recorded places of birth	26
<i>Mowbray Maternity Hospital</i>	50.9% (3825/7512)
<i>Mitchell's Plain MOU</i>	38.8% (2918/7512)
<i>Home births</i>	1.5% (113/7512)
<i>Born in transportation</i>	0.4% (29/7512)

^a Born before the 37th week of gestation [26]. ^b <2500gram [27]. The total number of infants (*n*) for each category differs due to information not being available for all infants at time of recording.

Referral rate

The bilateral first screen referral rate was 7% (250/3573) for DPOAE compared to 4.6% (179/3879) for AABR (Figure 1). These rates are significantly different for the techniques (Chi-square test; $p < 0.01$). Table 2 provides a breakdown of the bilateral refer outcomes as well as the unilateral refer rates for DPOAE and AABR.

Table 2. Distribution of first and second stage screening results (*CNT = Could not test)

Refer Category	DPOAE (n=3573)	AABR (n=3879)
First stage referral rate		
Bilateral refer outcome	7.0%	4.6%
<i>Refer bilaterally for OAE or AABR</i>	6.2%	2.4%
<i>CNT bilateral (restless)</i>	0.5%	1.4%
<i>CNT bilateral (technical error)</i>	0.1%	0.1%
<i>CNT/Refer unilaterally for OAE or AABR</i>	0.1%	0.7%
Unilateral refer outcome	10.8%	10.4%
Second stage/diagnostic referral rate	0.7%	0.3%

During the second stage, a sub-group of 23 infants, including 17 who referred for DPOAE and 6 who referred for AABR, received appointments for a third screen at the MOU (Figure 1). The main reason for a third screen was a CNT/CNT result on previous screening. The overall second stage refer rate dropped to 0.7% (26/3573) for DPOAE and 0.3% (12/3879) for AABR (Table 2). One infant from the DPOAE group who obtained a second stage unilateral refer result was given a diagnostic appointment because of parental concern (Figure 1).

Gender had no significant effect on first screen results (Chi-square test; $p > 0.01$). Age and screening technology had a significant effect on screen results (Chi-square test; $p < 0.01$). Figure 2 shows hearing screening outcome for DPOAE and AABR technologies as a function of age at screening. Initial screen referral rates of newborns younger than ≤ 6 days ($n=5437$) were significantly lower (Chi-square test; $p < 0.01$) for AABR compared to DPOAE but there was no significant difference in screening outcome for the two technologies for infants older than 6 days ($n=2011$; Chi-square test; $p > 0.01$).

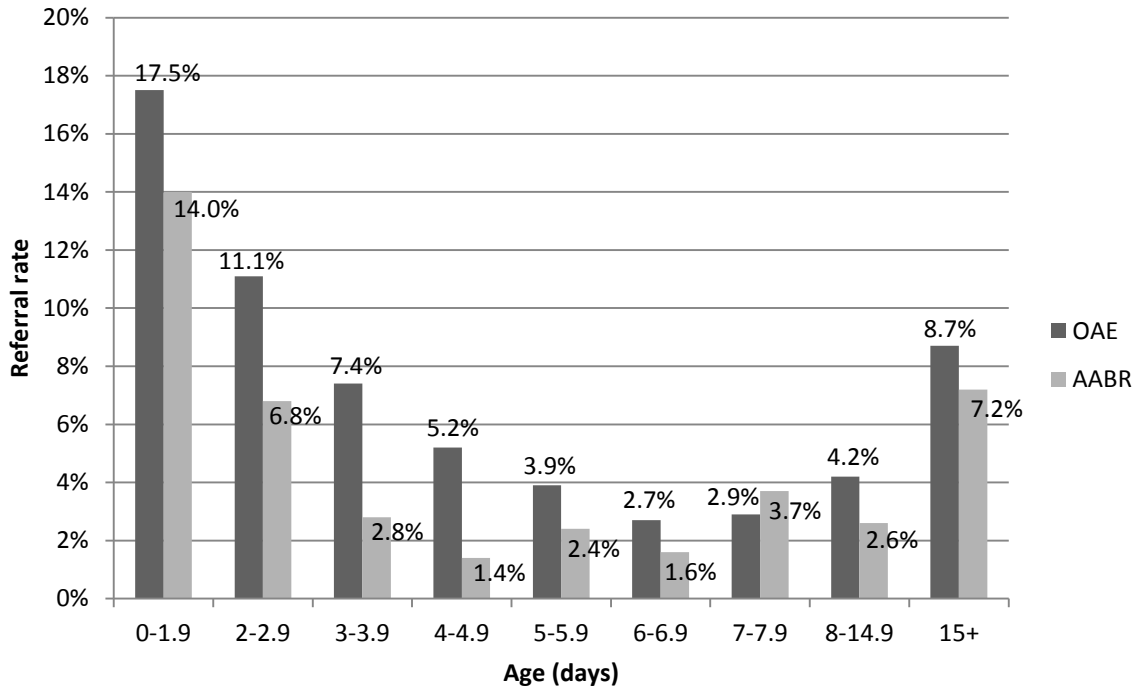


Figure 2. Distribution of first screen referral rate for DPOAE and AABR across age categories

Follow-up rate

There was no significant difference (Chi-square test; $p > 0.01$) between rescreen follow-up return rate for DPOAE (90%) and AABR (86.6), as displayed in Table 3. Majority of rescreens (67.4%, 256/380) coincided with the infants' second postnatal visit whilst 28.7% (109/380) returned only for hearing screening. Two infants, both from the AABR group, passed away prior to their diagnostic appointments (Figure 1) and were thus excluded from the diagnostic follow-up analysis (Table 3).

Table 3: Distribution of first and second stage follow-up rates

Follow-up Category	DPOAE group	AABR group	Total sample
First stage (MOU)			
<i>First follow-up</i>	90% (225/250)	86.6% (155/179)	88.6% (380/429)
<i>Second follow-up</i>	76.5% (13/17)	66.7% (4/6)	73.9% (17/23)
Second stage (Hospital)			
<i>Diagnostic follow-up</i>	92.3% (24/26)	90% (9/10)	91.7% (33/36)

Diagnostic outcomes

A total of 33 infants returned for a first diagnostic assessment. Among these infants, 24 had a refer outcome for DPOAE screening and 9 had a refer outcome for AABR screening (Figure 1). The 33 infants were grouped into a 'normal' (24.2%, 8/33) and 'abnormal' (75.8%, 25/33) category based on initial visit results. These findings are summarized in Table 4. All those in the 'normal' category (n=8) were within the DPOAE refer group and were discharged based on normal results elicited from repeat OAE screening (62.5%) or diagnostic ABR (37.5%).

The 'abnormal' group (n=25) received various ENT and audiological follow-ups with an average of three and a maximum of six follow-up visits. Subsequently, 15 infants were diagnosed with middle ear effusion (MEE) of which four went on to receive pressure equalising tubes. Not all these infants received complete diagnostic assessments. Final diagnostic outcomes are also summarised in Table 4 and Figure 1. Outcomes classified as normal hearing (n=15) were based on a combination of ENT confirming that the middle ears were clear and pass results on either repeat OAE screening (33.3%), freefield behaviour observation audiometry (46.7%), diagnostic ABR (13.3%), or the absence of parental concern (6.7%).

The prevalence rates of bilateral sensorineural hearing loss (SNHL) among infants undergoing diagnostic assessment were 0.3/1000 (1/3573) for the DPOAE refer group, 1/1000 (4/3879) for the AABR refer group and 0.7/1000 (5/7452) for the combined research sample. DPOAE screening resulted in a 4.2% (1/24) true positive rate of those who attended the diagnostic follow-up compared to 44.4% (4/9) for AABR screening. The prevalence rates of MEE, including unilateral and bilateral cases, were 2.2/1000 (8/3573) for the DPOAE group, 1.8/1000 (7/3879) for the AABR group and 2/1000 (15/7452) for the total sample.

Table 4. Diagnostic outcomes for referred infants (*HL = Hearing loss)

Outcome Category	DPOAE n=3573	AABR n=3879	Total sample n=7452
Total referred for diagnostic assessment	26 (0.73%)	12 (0.31%)	38 (0.51%)
Deceased (prior to 1 st visit)	-	2 (0.05%)	2 (0.03%)
Lost to follow-up (1 st visit)	2 (0.06%)	1 (0.03%)	3 (0.04%)
Initial diagnostic visit			
<i>Normal</i>	8 (0.22%)	-	8 (0.11%)
<i>Abnormal</i>	16 (0.45%)	9 (0.23%)	25 (0.34%)
<i>Conductive HL - unilateral</i>	2 (0.06%)	1 (0.03%)	3 (0.04%)
<i>HL unspecified - unilateral</i>	1 (0.03%)	-	1 (0.01%)
<i>HL unspecified - bilateral</i>	3 (0.08%)	4 (0.11%)	7 (0.09%)
<i>MEE - unilateral</i>	2 (0.06%)	1 (0.03%)	3 (0.04%)
<i>MEE - bilateral</i>	6 (0.17%)	6 (0.15%)	12 (0.16%)
Final diagnostic outcomes			
<i>Bilateral SNHL</i>	1 (0.03%)	4 (0.11%)	5 (0.07%)
<i>Lost to follow-up (diagnostic process)</i>	3 (0.08%)	2 (0.05%)	5 (0.07%)
<i>Normal (post follow-up)</i>	12 (0.34%)	3 (0.08%)	15 (0.20%)

Mean age at time of referral to diagnostic services was 22 days (n=38) but long waiting lists at the tertiary facility resulted in a mean age of 106 days (15 weeks; n=33) at the time of the first diagnostic appointment (Table 5). The age at time of diagnosis of SNHL came to a mean of 269

Table 5. Distribution of infants' ages and waiting times at various stages of the follow-up process

	Age (days) at screen referral (n=38)	Age (days) at 1st diagnostic session (n=33)	Waiting time (days) screen referral to 1st diagnostic session (n=33)	Age (days) at diagnosis (n=5)
Mean (SD)	22 (28.2)	105.9 (43.4)	84.7 (44.5)	269 (279.9)
Minimum	3	29	11	29
Maximum	138	191	176	718

days (38.4 weeks; n=5). The cases of these five infants varied greatly ranging from being diagnosed under the age of one month to being diagnosed just before the age of two years.

Screening personnel

Only the data of screener 1 and 2 were considered in this section as screener 3 conducted limited screening and did not screen on consecutive days. Screener 1 completed 200 DPOAE and 174 AABR screens with bilateral refer rates of 14.5% for DPOAE and 5.2% for AABR screening. Screener 2 performed 3301 DPOAE and 3635 AABR screens and yielded bilateral refer rates of 6.5% for DPOAE and 4.6% for AABR screening. The difference in referral rates between the two screeners was statistically significant for DPOAE screening (Chi-square test; $p < 0.01$) but not for AABR screening.

No statistically significant difference was noted in the amount of screens per day between the two technologies (t-test; $p > 0.01$) as shown in Table 6.

Table 6. Screening capacity of dedicated screener with DPOAE and AABR technology

	<i>DPOAE</i>	<i>AABR</i>
Mean tests per day (SD)	24 (9.1)	23 (7.0)
Maximum tests per day	51	43
Number of test days	155	182

DISCUSSION

Referral rates

Two-stage screening protocols reportedly reduce referral rates [16,22,28]. The current study supports this conclusion. Initial referral rates of 7% for DPOAE and 4.6% for AABR decreased to

0.7% and 0.3% after the second stage. These values are well below the national benchmark of 5% [18] and the international benchmark of 4% [4]. Initial and second stage DPOAE referral rates were lower than the 9.5% and 3% respectively reported in a preceding immunisation-linked hearing screening programme that employed a similar protocol [16]. The reduction might be attributed to the younger point of entry on the MOU postnatal visit platform (mean age 6 days compared to 3.9 weeks at first screen).

Comparative studies reporting referral rates for OAE and AABR screening in hospital-based settings in South Africa (TEOAE 37.9%; AABR 16.7%; [22]), Spain (TEOAE 8.2%; AABR 0.35%; [29]) and Turkey (TEOAE 10.5%; AABR 2%; [30]), demonstrate lower AABR referral rates, similar to findings in this study.

To date, AABR has not been recommended for primary health care contexts due to high disposable costs, test time and difficulty in obtaining results in infants beyond the newborn period [15,31]. However, advances in technology, like the MB11 BERAphone™, combined with the younger point of entry achieved in this study, demonstrated that cost-effective infant hearing screening with AABR is feasible and well-suited for countries with limited resources. Reduced initial referral rates not only result in cost and time savings but also in fewer caregivers attending additional appointments and experiencing stress related to their infant's hearing status [29,32,33].

If a unilateral refer criteria was to be implemented, as is standard practice in many countries, the programme would be severely pressured with initial referral rates rising to 17.8% for DPOAE and 15% for AABR. This would result in approximately three times the amount of second stage rescreens with a causal sequence on the diagnostic referral rate and first screen coverage.

Consistent with previous reports [7,22], the least optimal time to screen was within the first 48 hours after birth for both technologies. Lowest initial referral rates were at the age of 4 days for AABR (1.4%) and 6 days for DPOAE (2.7%). Screening between 3 and 14 days after birth with AABR and 5-14 days of age with DPOAE resulted in referral rates meeting a $\leq 5\%$ benchmark [18]. Referral rates of both technologies increased post two weeks of age, as infants are typically more difficult to test and transient middle ear effusion might be more prevalent [16,34]. MOU postnatal follow-up visits present a very useful platform for screening that results in lower referral rates compared to earlier (<48h) or later (immunisation-linked) screening [7,16]. Additionally, this study highlights that AABR technology could significantly reduce initial referral rates and allow more efficient screening at an earlier age on this platform.

Follow-up rates

Internationally, loss to follow-up is one of the greatest challenges experienced in newborn hearing screening programmes with follow-up rates in the region of 50% often being reported [29,31,35]. In contrast, follow-up rates in the current study were encouragingly high for both technology groups. The first MOU follow-up was 90% for infants with a DPOAE refer and 86.6% for infants with an AABR refer. Diagnostic follow-up rates for DPOAE refer outcomes (92.3%) and AABR refer outcomes (90%) surpassed the target of 70% for community-based screening programmes [4,18]. These findings are in agreement with an earlier immunisation-linked screening programme that had follow-up rates of 85.1% for clinic level and 91.8% for diagnostic follow-up [16]. Friderichs et al. [16] ascribed high follow-up rates to the dedicated monitoring of the programme by a screening coordinator which included strategies such as telephone calls and reminders in folders. Contributors to higher follow-up rates in our study may have included the facilitative platform offered by the MOU postnatal visits, the use of a dedicated screener and shorter time between initial screen and follow-up. The dedicated screener was well trained in providing thorough information counselling at the point of first screen referral. 67% of repeat

screens happened in conjunction with the babies' second postnatal follow-up visit and was achieved without any reminders. Strategies only had to be implemented for the remainder; reducing time spent and related costs. The dedicated screener contacted parents/caregivers on the same day that they defaulted on their infant's follow-up appointment to establish the reason for not attending. Concerns or misconceptions could be addressed and a suitable follow-up date rescheduled. Lastly, as follow-ups took place within a few days of the initial screen, the majority of infants were still within the newborn period and mothers/caregivers were cooperative.

In the current study follow-up rates deteriorated with additional screening or diagnostic appointments. For example, five babies who received a second diagnostic appointment were lost to follow-up after having attended their first appointment. This highlights the importance of following protocols and obtaining complete and accurate results as early as possible.

Diagnostic outcomes

The prevalence rate for bilateral SNHL was 1/1000 for those screened with AABR and 0.3/1000 for those screened with DPOAE. Both rates were lower than the 1.5/1000 reported for an earlier immunisation linked programme that implemented a two stage DPOAE protocol [16]. However, the rate in the earlier study included unilateral and mixed losses of permanent nature [16]. A universal community-based programme in Nigeria, employing a two stage OAE/AABR protocol, resulted in a 22.5/1000 yield of PCEHL (uni-/bilateral). The high yield was attributed to the large percentage (55%) of screened infants who were born outside of hospital settings without skilled attendance, a factor that influences the incidence of PCEHL [31]. In contrast, only 1.9% of births in our sample took place without skilled attendance.

The low yield obtained in this study is partially attributed to loss to follow-up. If the ideal scenario of 100% attendance was achieved for all screening and diagnostic follow-ups, the prevalence

rates are estimated to increase to 0.6/1000 for the DPOAE group and 2.3/1000 for the AABR group. Secondly, variability in the applied diagnostic protocols could have added to the low yield. Diagnostic ABR measurements were only employed in eight of the 33 initial evaluations (5 DPOAE group; 3 AABR group). Repeat screening at diagnostic level, absence of bone conduction measures and use of freefield behavioural observation audiometry instead of ear specific visual reinforcement audiometry were noticed. All these factors have been described as potential pitfalls in the audiological assessment of young infants [36].

Standardised implementation of a diagnostic test battery, aligned with international best practice, as well as skill development in the areas of audiological diagnosis and management of young infants (1-6 months) are required to reach the goals of EHDI [4,37,38]. Implementation of clinical peer review and quality assurance systems have been facilitative in many countries [39]. Furthermore, strengthening district level audiology and ENT services could alleviate pressure on tertiary level and improve timeous access to diagnostic services. These factors should be addressed prior to pursuing more stringent screening criteria.

Dedicated screening personnel

Screeener experience reduced DPOAE referral rates as evident from the 14.5% achieved by screener 1 who worked a month compared to 6.5% by screener 2 who worked for 15 months. Interestingly, AABR referral rates were not influenced by screener experience. Although previous studies showed that AABR (MB11 BERAPhone™) test time is still longer than OAE screening [22,40], this study showed comparable daily capacity for both technologies with an average of 24 DPOAE screens per day and 23 AABR screens per day. This is higher than the reported average of 13 OAE screens per day in a community-based programme in Nigeria but corresponds to the stated capacity of 20 screens per day [31]. The only difference being that their screener was supported by an administration assistant whilst the screener in this study

worked alone. Daily number of screens is dependent on access to infants which is an important benefit of community-based programmes [31]. It was also an observed benefit of the MOU postnatal visit platform over the immunisation clinics within the Cape Town area.

The appointment of a dedicated non-professional screener positively influenced consistent screening services, follow-up rates, follow-up administration, electronic data-capturing, equipment maintenance, and provision of antenatal information regarding UNHS. These benefits contribute to an efficient and cost effective screening programme [16]. They cannot be expected in screening programmes relying on existing nursing personnel with high caseloads.

The current study showcased that the role of dedicated screeners could be fulfilled by non-professional individuals with character and experience being more important than qualification. Quality of training and regular supervision is vital.

CONCLUSION

Postnatal follow-up visits at community-based MOU facilities create a useful platform for access to UNHS and facilitate high follow-up rates. AABR technology with negligible disposable costs and improved test time provides opportunity for AABR protocols to be utilised in community-based screening programmes. Benefits hereof include significantly lower initial referral rates, higher true positive rates, more efficient screening at an earlier age and the ability to identify neural hearing losses. This however needs to be seen in the light of the MOU context evaluated in this study with infants mostly younger than two weeks of age. Well trained and managed non-professionals can successfully be utilised as dedicated screeners and positively impacted programme efficiency and administration. This may be valuable in settings where UNHS feasibility has been poor due to overburdened nursing personnel. Alongside the development of

contextual UNHS models, timeous access to diagnostic services as well as diagnostic protocol and skill development coherent with international best practice should be fostered.

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Conflict of interest statement

The authors have no conflict of interests to declare.

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