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THE STATUS OF NEONATAL HEARING SCREENING IN SUB-SAHARAN
AFRICA: A SYSTEMATIC REVIEW

by

DAVID ENGELMAN

A capstone research project submitted to the Graduate Faculty in Audiology in partial
fulfillment of the requirements for the degree of Doctor of Audiology, The City

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This manuscript has been read and accepted for the Graduate Faculty in Audiology in satisfaction of the capstone research requirement for the degree of Au.D.

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ABSTRACT

The Status of Neonatal Hearing Screening in Sub-Saharan Africa: A Systematic Review

by

David Engelman

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Late identification of hearing loss can lead to speech and language delays, as well as social and academic difficulties. Neonatal hearing screening has reduced the age of identification of hearing loss in many developed countries. However, the practice is limited or non-existent in much of the developing world, such as sub-Saharan Africa. This systematic review used currently available literature to determine the status of neonatal hearing screening programs in sub-Saharan Africa and to develop a model program for the region. Results suggested a lack of neonatal hearing screening services in sub-Saharan Africa, stressing the need for more pilot hearing screening studies to be conducted. Such studies would establish the feasibility and efficacy of neonatal hearing screening programs in specific countries and pave the way for programs targeting children at risk for hearing loss. These programs would facilitate timely interventions in those diagnosed with congenital or early-onset hearing loss.

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INTRODUCTION

Hearing impairment is the most common sensory deficit in humans, affecting more than 360 million people worldwide (WHO, 2013). In low- and middle-income countries, hearing disability ranks third on the list of non-fatal disabling conditions (Mathers, Lopez, & Murray, 2006 as cited in Fagan & Jacobs, 2009). A common theme amongst all discussions related to the global health agenda is that of unequal development in the world. Nowhere else in the world are such inequities and disparities in health and economics seen then in sub-Saharan Africa (Fagan & Jacobs, 2009).

Sub-Saharan Africa has a population of 910.4 million spread over a land mass of 24.3 million square meters and 46 countries with wide ethnic, linguistic, religious, economic, and cultural diversity. Although the population represents 13% of the world's population, an estimated 48.5% or more of its people live below the poverty line, on less than US\$1.25 a day (World Bank, 2010). The region's unique geography, climate, and substandard healthcare predispose its population to a heavy and wide-ranging burden of disease. Most of the fatal diseases and chronic conditions which are associated with substantial morbidity and disability are either preventable or treatable (Grantham-McGregor et al, 2007, as cited in Olusanya, 2008).

In sub-Saharan Africa, an estimated 180,000 infants are born annually with acquired hearing loss in the first few weeks of life as compared to 22,000-44,000 babies in all the industrialized countries combined (Olusanya & Newton, 2007; UNICEF, 2007; Smith et al, 2005 as cited in Olusanya, 2008). In Nigeria, for example, a prevalence of up to 28 per 1000 live births for all degrees of sensorineural hearing loss has been suggested, which is one of the

highest rates reported worldwide (Olusanya, 2011). Congenital and early-onset hearing loss is an etiologically heterogeneous chronic condition attributable to genetic and environmental causes (Olusanya, 2008). Hearing impairment in infancy is not readily detectable by behavioral observations, although as the infant matures it can be suspected by parents through a baby's inattention or erratic response to sound. Depending on the severity, hearing impairment in an infant may not be detected until the infant is well over 18 months of age by parental suspicion (Olusanya, 2008). The consequences of these delays in identification are dramatic in that undetected hearing loss can result in delayed speech and language acquisition, social-emotional or behavioral problems, and lags in academic achievement (Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998; Bess, 1985; Bess, Dodd-Murphy, Parker, Oyler & Matkin, 1988).

In contrast, neonatal hearing screening, a mandated health promotion activity in 36 states in the United States, as well as Guam, Puerto Rico and the District of Columbia, has successfully reduced the age of identification of hearing loss which in turn has had implications for early intervention and the successes achieved through cochlear implantation and hearing aids. Since the initiation of newborn hearing screening in the United States, the average age of identification of hearing loss decreased from 2.5 - 3 years of age in the early 1990's to 2 - 3 months of age according to most recently available research (White, 2008; Hoffman & Beauchine, 2007; Harrison, Roush, & Wallace, 2003).

Similar successes in early identification have been seen as well in many developed nations worldwide. For example, Israel's ministry of health issued a directive establishing universal newborn hearing screening in all hospitals in the country beginning on January 1, 2010. Gilbey, Kraus, Ghanayim, Sharabi-Nov, & Bretler (2013) evaluated the performance of the newly established Israeli screening program from March 15, 2010 until the end of 2011 at Ziv

Medical Center in Zefat. Using the United States' Joint Committee on Infant Hearing (2007)'s recommended quality benchmarks, Gilbey, et al. (2013) found a screening coverage of 94.8%, closely approaching the recommended benchmark of 95%. In England, from November 2003 to February 2004, during the first phase of a newborn hearing screening program there, screening coverage was found to be 97.5%, surpassing the recommended 95% benchmark (Bamford, Uus, & Davis, 2005).

Such early identification results in early intervention via hearing aids, cochlear implants, and various assistive listening devices. This intervention allows for speech and language development and academic achievement to remain on target. In research conducted in Colorado, children whose hearing losses were identified by 6 months of age demonstrated significantly better receptive and expressive language skills than did children whose hearing losses were identified after the age of 6 months (Yoshinaga-Itano, et al., 1998). Yoshinaga-Itano & Apuzzo (1998) found that children identified with hearing loss after 18 months of age were almost twice as delayed in their expressive language and language understanding abilities when compared with children identified before 6 months of age. Furthermore, children receiving cochlear implants between 12 and 24 months of age show similar language skills as typical peers (Svirsky, Teoh, & Neuburger, 2004).

Fulcher, Purcell, Baker, & Munro (2012) performed a comparative study of a cohort of 45 early identified (≤ 12 months) and 49 late identified (> 12 months to < 5 years) children with hearing loss, with all severities of hearing loss and no other concomitant diagnoses. The children all attended the same oral auditory-verbal early intervention program in New South Wales, Australia. Speech and language assessments standardized on typically developing hearing children were conducted at 3, 4 and 5 years of age.

Those who were diagnosed early had received amplification by 3 months, enrolled into auditory-verbal intervention by 6 months and received a cochlear implant by 18 months if required. These children were able to “keep up with” rather than “catch up to” their typically hearing peers by 3 years of age on measures of speech and language, including children with profound hearing loss (Fulcher, et al, 2012). Fulcher, et al. (2012) found that the early identified children significantly outperformed the late identified at all ages and for all severities of hearing loss. By 3 years of age, 93% of all early identified participants scored within normal limits for speech; 90% were within normal limits for understanding vocabulary; and 95% were within normal limits for receptive and expressive language. Progress was maintained and improved so that by 5 years of age, 96% were within normal limits for speech, with 100% within normal limits for language.

In the absence of a systematic effort to screen infants with hearing loss the average age of detection is well over two years, and detection may be as late as six years in sub-Saharan Africa (Olusanya, 2008). In Kenya, for example, many children with hearing loss are not identified until five to seven years because of stigma, while some are hidden and are never diagnosed (Wilson, 2006 as cited in Olusanya, 2008).

Olusanya, Luxon & Wirz (2005) developed a questionnaire based study of 363 parents of children attending the only public schools for the deaf in Lagos, Nigeria, with a total enrollment of 429. Olusanya, et al. (2005) found that parents were predominantly (81%) the first to suspect or detect hearing difficulty in their children. Parental suspicion occurred mostly at 12-24 months, compared with 8-14 months in developed countries. Only 12% suspected hearing difficulty by age 6 months. The most common mode of detection was a child's failure to respond to sound (49%). Speech/language defects or unintelligible speech were least associated with

hearing difficulty. As in developed countries, doctors were most commonly consulted for help (77%). However, most children (80%) were not provided with hearing aids even where appropriate, granted that cochlear implantation was improbable. Parents were often told that their children were “slow starters” and would outgrow the speech delays, only to be enrolled in schools for the deaf when this optimism failed. Ironically, only 6% were so enrolled by age 6 years, with a mean age of enrollment of 10.3 years, only further protracting developmental delay (Olusanya, et al., 2005).

Even in South Africa, a sub-Saharan African nation with a relatively well developed infrastructure, the median age of diagnosis at Universitas Hospital in Bloemfontein was found to far exceed international benchmarks (Butler, Basson, Britz, de Wet, Korsten, & Joubert, 2013). Butler, et al. (2013) performed a retrospective, descriptive study, analyzing data from 2001 to 2010. A total of 260 cases of congenital hearing loss were analyzed. The median age of diagnosis of hearing loss was 44.5 months. The median age of first visit was 40.9 months, and the median delay between first visit and diagnosis was 49 days.

Current pilot infant hearing screening programs in developing countries are either hospital- or community based (Olusanya, 2006). Screening babies in hospitals before discharge is desirable for at least two primary reasons. First, screening eliminates the need to ask mothers to return specifically to have their babies tested. Parents are likely to be less enthusiastic to seek detection of an invisible and non-life threatening handicap in their apparently normal babies. Taking an apparently well child to the hospital is viewed as socially and culturally inappropriate in many communities because of the notion that hospitals are established only to serve the sick (Olusanya, 2006). Second, screening helps health care professionals satisfy an important ethical

obligation of ensuring that babies have been examined and tested for hidden, detectable abnormalities before discharge (Olusanya, 2006).

The protocol commonly used in hospital-based screenings is the two-stage screening protocol with Transient Evoked Otoacoustic Emissions (TEOAE) testing followed by Automated Auditory Brainstem Response (AABR) testing for children referred from the first-stage screen (Olusanya, 2006). There are, however, some challenges. Finding a suitable section within the hospital ward is necessary to minimize false referral rates where the ambient noise levels are excessive, which could be a major challenge for hospitals with space constraints (Olusanya, 2006). When the number of babies awaiting screening is large, some mothers may be too impatient to wait and may never return (Olusanya, 2006).

Hospital-based neonatal hearing screening pilot projects have been implemented in Nigeria, South Africa, India, Pakistan, Saudi Arabia, Iran, Qatar, Jordan, Oman, China, Hong Kong, Taiwan, Malaysia, Philippines, Singapore, Brazil and Mexico (Olusanya, 2006). Reports from these countries confirm that hospital-based neonatal hearing screening is feasible. The attitudes of parents and health care professionals generally were favorable, although some challenges remained unresolved. High default rates for follow-up services are common and require effective data management and tracking systems. Most programs were initiated and entirely managed by health professionals in hospitals with little or no government funding. Most of these countries have no free national health care service or publicly administered health insurance schemes; thus, only parents who can afford to pay seek these services (Olusanya, 2006).

For example, in Nigeria, Olusanya, Wirz, & Luxon (2008b) demonstrated the feasibility of hospital-based neonatal hearing screening by non-specialists at Lagos Island Maternity Hospital, a state-owned hospital in an inner city area of metropolitan Lagos, the largest city in Nigeria. The major finding of the Olusanya, et al (2008b) study was that hospital based universal neonatal hearing screening using non-specialists was indeed feasible in Lagos, Nigeria. Screening coverage exceeded the target of 95%, reflecting significant maternal willingness to participate.

Staff at Lagos Island Maternity Hospital received two weeks of training prior to commencement of the screening program. An informational booklet was produced for both hospital staff and parents. The screening protocol followed two stages, with an initial screening with TEOAE, followed by an AABR for those referred from the TEOAE. Emissions were recorded from 1.5 to 3.5 kHz. AABR screening was performed with at least 1,000 soft click stimuli at 35 dB HL.

All babies born in the hospital between May 2005 and February 2006 were eligible. Parents were informed of the benefits of early hearing detection and intervention with an informational leaflet. Infants were distinguished between those in the Well Baby Nursery (WBN) from those in the Special Care Baby Unit (SCBU). Well babies were screened with TEOAE between 24 and 48 hours after delivery, and the test was repeated if a refer result was obtained. Infants who failed the second TEOAE screening were scheduled for an AABR screening prior to discharge. Infants who were then referred for either one or both ears following the AABR screening were scheduled for a diagnostic evaluation at a nearby audiological center. Infants from the SCBU were screened with both TEOAE and AABR regardless of test outcomes, due to the high rate of auditory neuropathy in such infants. Infants who missed the screening

before discharge were screened as outpatients when visiting the hospital for their six-week post natal clinic appointment. Follow-up counseling appointments were scheduled for parents of babies with any degree of confirmed bilateral or unilateral sensorineural hearing impairment. Infants with severe to profound hearing impairment were referred for a hearing aid evaluation and fitting and early intervention services at the audiological center (Olusanya, et al., 2008b).

However, despite the demonstrated feasibility of hospital based hearing screening, in many developing countries, home births and deliveries at private maternity homes by birth attendants account for the majority of babies born outside of hospitals (WHO, 2006b as cited in Olusanya, 2006). The rest are delivered in health facilities within church premises or before arrival at hospitals (Olusanya, 2006). Therefore, a complementary program would be required in communities with low rates of hospital-based deliveries.

Routine childhood immunization is perhaps the most well-established public health program globally due to the substantial technical and financial support received yearly from UNICEF, the WHO and several donor agencies and partners. Consequently, immunization clinics have been used as platforms for delivering new child health intervention packages, especially in the developing world (WHO, 2002 as cited in Olusanya, 2006). They provide a ready framework for introducing infant hearing screening (Olusanya, Luxon, & Wirz, 2004).

Preliminary results from two pilot programs in South Africa and Nigeria confirm that infant hearing screening at immunization clinics is feasible and worthwhile in developing countries, although more reports are needed from other regions because screening protocols may differ from country to country (Olusanya, 2006). Difficulties may arise when routine immunization programs are interrupted because of vaccine shortages or other extraneous reasons. Another major challenge arises when screening cannot be completed and babies require follow-

up visits in addition to scheduled immunization clinics. Despite the possibility of program disruptions, the experiences in many countries strongly suggest that routine immunization programs are still the most effective platform for integrated child health intervention globally (Olusanya, 2006).

The status of hearing screening efforts is variable throughout sub-Saharan Africa and given the successes in developing nations, a first step in initiating screening activities is to learn the status, availability, and specific nature of neonatal hearing screening programs in sub-Saharan Africa. The goal of the present review will be to determine and assess the status, availability, and specific nature of neonatal hearing screening programs in sub-Saharan Africa based on currently available literature. Based on this information, we can then furnish recommendations and direction for current and future neonatal hearing screening programs in sub-Saharan Africa and the developing world at large, and develop a model program for neonatal hearing screening in sub-Saharan Africa.

The present review will address the following research questions:

1. What proportion of sub-Saharan African nations currently perform neonatal hearing screening?
2. What is the rank ordering of settings in which neonatal hearing screening is conducted in sub-Saharan Africa?
3. What is the rank ordering of procedures used for neonatal hearing screening?
4. What professionals are most and least likely to oversee the neonatal hearing screening?
5. What are the feasibility and efficacy data supporting neonatal hearing screening in sub-Saharan Africa?

6. What obstacles are predominantly encountered in establishing successful neonatal hearing screening programs in sub-Saharan Africa?
7. What are the rates of referral and follow-up and what are the typical settings to which neonates are referred in sub-Saharan Africa?

METHODS

Participants

Only studies that met the following criteria were included: (1) screening of hearing in neonates in either hospital or community based programs located in sub-Saharan Africa; (2) any article that was not a descriptive study or review involving currently implemented neonatal hearing screening programs in sub-Saharan Africa was discarded; (3) any article that did not address one of the aforementioned research questions was discarded.

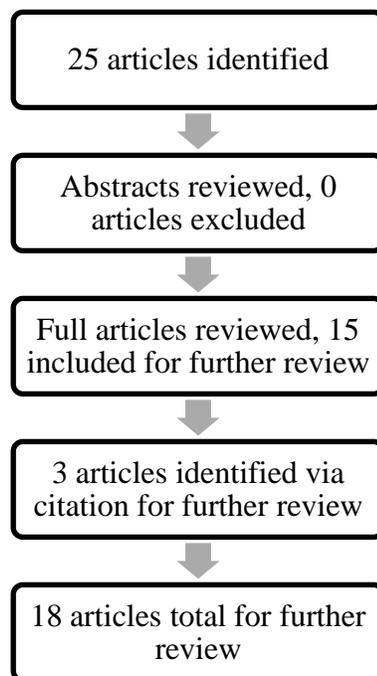
Procedure

Initial literature searches were conducted utilizing Medline, CINAHL, Scopus, ScienceDirect, and Google Scholar. Keywords included “neonatal hearing screening,” “newborn hearing screening,” “early hearing detection,” “hearing loss,” “congenital,” “syndrome,” “Africa,” “sub-Saharan Africa,” “settings,” “procedures,” “professionals,” “feasibility,” “efficacy,” “obstacles,” “referral,” and “follow-up”. The keywords of “Africa” and either “neonatal hearing screening,” “newborn hearing screening,” or “early hearing detection” were always present throughout the literature search. Articles not written in English were discarded. Articles were selected based on relevant titles and abstracts, and reviewed if it was determined that the article would address one of the aforementioned research questions. The selected articles were then read in full, with a secondary list consisting of relevant papers cited by these articles. The articles were then further sorted by their ability to address the specific research questions that have been presented above.

RESULTS

A total of 25 relevant articles were identified, using the key words and databases described earlier. After the abstracts were reviewed, no articles were excluded from further review as all articles initially met the inclusion criteria. The review of the full article was performed on the remaining articles to determine if they met the inclusion criteria, yielding a total of 15 articles for further review. Three relevant papers that were cited by these articles were included as well, yielding a total of 18 articles for further review. See Figure 1 for a flowchart representing this review process.

Figure 1. Flowchart representing article retrieval and review process.



The selected articles were reviewed to address the research questions described earlier. Presented below are the aforementioned research questions along with a detailed answer provided by the reviewed literature.

1. What proportion of sub-Saharan African nations currently performs neonatal hearing screening?

Out of a total of 46 sub-Saharan African nations, only 2 countries currently perform neonatal hearing screening, or roughly 4% of sub-Saharan African nations. Hospital based neonatal hearing screening programs have been implemented in South Africa and Nigeria as pilot projects. Pilot infant hearing screening programs have been implemented at immunization clinics in those countries as well (Olusanya, 2006).

2. What is the rank ordering of settings in which neonatal hearing screening is conducted in sub-Saharan Africa?

Current pilot infant hearing screening programs in developing countries are either hospital or community based. Available data to produce an accurate rank ordering of settings was scanty. However, based on currently available research, fair assumptions have been made to create such a rank ordering. See Table 1 for a summary of this hierarchy.

In South Africa, private sector hospitals are likely to lead the rank for settings offering newborn hearing screening. Newborn hearing screening services in the private health care sector are mostly dependant on individual initiatives from private practice audiologists in hospitals but is not mandated by hospital management (Swanepoel, Störbeck, & Friedland, 2009).

Meyer, Swanepoel, le Roux, & van der Linde (2012) conducted a telephone survey of all private hospitals ($n = 166$) in South Africa with obstetric units. Data on the existence of and type of newborn hearing screening programs were collected. Questionnaires were then subsequently distributed nationally to audiologists providing hearing screening at the respective private sector institutions who reported hearing screening services ($n = 87$). A return rate of 89% was obtained

for the questionnaires across all nine provinces of South Africa. Newborn hearing screening was available in 53% of private health care obstetric units in South Africa of which 14% provided universal screening.

Public sector hospitals in South Africa are likely to be the second highest ranking setting for neonatal hearing screening. Theunissen & Swanepoel (2008) performed a questionnaire based study, sampling all public sector hospitals ($n = 86$) with audiology and/or speech/language therapy services in eight out of the nine provinces of the country. Based on a 51% return rate, only 27% of the hospitals sampled offered any form of infant hearing screening, and only one hospital offered universal screening. Based on these findings, Theunissen & Swanepoel (2008) estimated that 7.5% of public sector hospitals nationally provide some form of infant hearing screening and less than 1% provide universal screening.

Immunization clinic-based screening programs have been proposed as an alternative to hospital-based programs, and are likely to rank as the third most common setting for neonatal hearing screening in South Africa. Swanepoel, Hugo, & Louw (2006) described a hearing screening program implemented at two immunization clinics in a representative South African community. Community-based universal infant hearing screening has since grown in South Africa, and a program has been initiated at eight primary health care clinics in the Cape Metropolitan area, as described by Friderichs, Swanepoel, & Hall (2012). Exact numbers on the availability of community-based infant hearing screening in South Africa was not available at time of research.

However, the rank order of settings is reversed in Nigeria. Community-based programs are likely to be more common than hospital-based programs, which is a reflection of Nigerian birthing patterns. A total of five universal neonatal hearing screening programs have been

piloted in Nigeria (Olusanya, et al, 2007). All of the piloted screening programs have been funded through a combination of public and private sources including donation or loan of equipment by manufacturers, and are offered at no charge to parents (Olusanya, et al, 2007). Details regarding exactly which of these five programs were hospital-based and which of these were community based were not available.

At least one of these programs was hospital based, as Olusanya et al. (2008b) has described the feasibility and efficacy of a universal neonatal hearing screening program at Lagos Island Maternity Hospital. Olusanya, Wirz, & Luxon (2008a) described a community-based infant hearing screening program in Lagos in which all infants aged 3 months or under attending four bacille Calmette–Guérin (BCG) immunization clinics accounting for over 75% of the BCG coverage in the study location were screened. Okhakhu, Ibekwe, Sadoh, & Ogisi (2010) aimed to detect the crude prevalence of congenital hearing loss among newborns in Benin City, Nigeria. Neonates at designated immunization centers in Benin City were screened for hearing loss via the detection and analysis of DPOAE from both ears. Information regarding the settings of the remaining neonatal hearing screening programs in Nigeria was not available at time of research.

Table 1. Hierarchy of Settings Where Neonatal Hearing Screening Occurs in Sub-Saharan Africa.

South Africa	Nigeria
1. Private sector hospitals	1. Community-based
2. Public sector hospitals	2. Hospitals
3. Community-based	

3. What is the rank ordering of procedures used for neonatal hearing screening?

Table 2 provides a ranking of commonly used procedures in neonatal hearing screening in sub-Saharan Africa. In hospital-based neonatal hearing screening programs, the most common protocol of procedures used for neonatal hearing screening in sub-Saharan Africa consists of a two-stage screening. TEOAEs are performed first followed by AABR for children referred from the TEOAE screen (Olusanya, 2006). However, Meyer, et al. (2012) found that in the private health sector of South Africa, most (81%) of the healthy baby screening programs used only OAE screening. Auditory brainstem response screening was employed by 24% of neonatal intensive care unit screening programs with only 16% repeating ABR screening during the follow-up screen. Theunissen, et al. (2008) found that in public sector hospitals in South Africa, a screening OAE was used as the initial screen in seven out of 12 hospitals with a screening program, while AABR was used by only two hospitals. Diagnostic distortion product evoked otoacoustic emissions (DPOAE) tests were used by three hospitals to screen infants. Three hospitals reported using behavioral observation as part of their screening procedure although all of these reported using it in combination with objective methods.

The rank ordering of community-based hearing screening procedures differs between which sub-Saharan African nation the screening is conducted in. In South Africa, a hearing screening program was implemented at two maternal and child healthcare clinics over a 5-month period, 3 days per week, in the Hammanskraal district. The screening protocol consisted of a first-stage screen with DPOAE and a high frequency probe tone (1 kHz) tympanometry for infants ages birth to 12 months. A planned second-stage screen with AABR was initially included but discontinued as it did not prove to be an effective screening tool for a majority of

infants attending the clinics due to wakefulness and restlessness (Swanepoel, Hugo, & Louw, 2006).

Friderichs, et al. (2012) evaluated a community-based universal infant hearing screening program in the Cape Metropolitan area of South Africa over a 19-month research period. The study employed a two-stage DPOAE screening protocol. A two-stage screening protocol was employed to reduce the burden of false positive referrals to tertiary hospital level. The DPOAE screening parameters included evaluation of four frequencies (5, 4, 3, and 2 kHz) using a 65/55 stimulus level (L1/L2). Three of the four frequencies were required to pass (with a 6 dB signal to noise ratio) for an overall pass result. This screening technology was chosen instead of AABR testing based on recommendations from Swanepoel, et al., (2006)'s pilot research project in the Hammanskraal district.

In contrast, in Nigeria, a two-stage protocol with an initial TEOAE screen at BCG immunization clinics was used. Olusanya, et al. (2008a) performed a cross-sectional study in an inner city area of Lagos that was home to seven primary health care centers that offered routine BCG immunizations. Four centers that accounted for over 75% of BCG immunizations in the study location were selected. A two-stage screening protocol was implemented. It consisted of first-stage screening using TEOAE and second-stage screening using AABR for all first-stage referrals. However, Okhakhu et al. (2010)'s work on screening newborns in Benin City was performed only with DPOAE. An AABR was used for the second-stage screening, but this was not available at the immunization clinic where the first-stage screening took place.

Table 2. Ranking of Most Commonly Used Procedures for Neonatal Hearing Screening in Sub-Saharan Africa.

South Africa: Hospital-Based	South Africa: Community-Based	Nigeria: Hospital-Based	Nigeria: Community-Based
1. DPOAE	1. DPOAE	1. TEOAE	1. TEOAE
2. AABR		2. AABR	2. AABR

4. *What professionals are most and least likely to oversee the neonatal hearing screening?*

Table 3 provides a comparison of which professionals are most likely to oversee infant hearing screening in sub-Saharan Africa. Not all of the cited literature specified the professionals involved in overseeing infant hearing screening. Olusanya, et al. (2007) performed a cross-sectional, descriptive and questionnaire-based study to examine the progress achieved in developing counties in regards to early hearing detection. Olusanya, et al. (2007) found that existing healthcare personnel in hospital-based projects are more commonly entrusted with screening except in a few countries like Nigeria where non-specialists are recruited and specially trained to conduct screening. Olusanya, et al. (2008b)'s study in Lagos, Nigeria confirmed the feasibility and effectiveness of using non-specialists for neonatal hearing screening in a hospital setting.

The use of non-specialists as screeners at primary healthcare level has been found to be cost-effective while regular health workers are effective in educating parents on the program. Restricting screening to highly skilled personnel like audiologists or other ear care specialists may not serve the course of rapid spread of infant hearing screening as an important public health program due to the general dearth of such manpower (Olusanya, et al., 2007).

In Olusanya, et al. (2008a)'s cross-sectional study of community-based infant hearing screening in Lagos, Nigeria, screening personnel consisted of two full-time and two part-time staff members with no prior experience in audiological testing. They were given two weeks of focused training by the principal investigator of the study. This included an overview of the peripheral auditory pathway and basic screening techniques. One fulltime staff member with experience as a community health worker was the designated screener, and the other full-time staff member provided the screener with administrative support. One part-time staff member provided extra administrative support on days when clinics were exceptionally busy while the second part-time staff member provided clerical assistance for data entry.

In Friedrich et al. (2012)'s evaluation of community-based infant hearing screening in South Africa, clinic nurses served as screening personnel. Similar to the training of non-specialists in Nigeria, the nurses were trained and mentored in infant hearing screening before the service commenced. Nurses were trained by the program manager and colleague at their relevant clinics and received ongoing support and training from the program manager throughout the course of the screening program. Alternatively, though, in Gauteng, South Africa, Swanepoel, Ebrahim, Joseph, & Friedland (2007) examined a universal neonatal hearing screening program in a private health care hospital where the screening was conducted by two qualified audiologists. Data on screening personnel in public sector hospitals in South Africa was not available at time of research.

Table 3. Comparisons of Which Professionals are most likely to Oversee Neonatal Hearing Screening in Sub-Saharan Africa.

Nigeria: Hospital-Based	Nigeria: Community-Based	South Africa: Community-Based	South Africa: Hospital-Based (Private)
Non-specialists	Non-specialists	Clinic nurses	Audiologists

5. *What are the feasibility and efficacy data supporting neonatal hearing screening in sub-Saharan Africa?*

Olusanya, et al. (2007) demonstrated that infant hearing screening is a feasible and viable early hearing detection strategy in developing countries, including sub-Saharan Africa. Their cross-sectional, descriptive study was based on responses to a structured questionnaire eliciting information on the nature and scope of early hearing detection services. Notwithstanding the myriad constraints in developing countries, the report demonstrated that early hearing detection programs are feasible as a public health initiative. Pilot projects using objective screening tests are on-going in a growing number of countries. Evidence from a rapidly expanding number of countries such as Brazil, Oman, and Chile that have progressed from rudimentary pilot projects to multi-city programs lend credence to infant hearing screening as an important and achievable public health initiative in the developing world, including sub-Saharan Africa (Olusanya, et al., 2007).

Olusanya, et al. (2008b) determined the feasibility and effectiveness of hospital-based universal newborn hearing screening in Lagos, Nigeria. They performed a cross-sectional pilot study based on a two-stage universal newborn hearing screening. The main outcome measures were the practicality of screening by non-specialist staff with minimal training, functionality of

screening instruments in an inner-city environment, screening coverage, referral rate, return rate for diagnosis, yield of congenital or early-onset hearing loss, and average age of hearing loss confirmation.

Olusanya, et al. (2008b) found that universal newborn hearing screening by a non-specialist staff was feasible in an inner-city environment in Lagos. Notwithstanding excessive ambient noise within and outside the wards, it was possible to identify a test site for TEOAE screening within the hospital. The screening coverage was 98.7% (1330/1347) of all eligible newborns and the mean age of screening was 2.6 days.

Olusanya, et al. (2008a) determined the feasibility and effectiveness of a community-based universal infant hearing screening program in Lagos. They performed a cross-sectional study in which all infants aged 3 months and under attending four BCG immunization clinics (that accounted for over 75% of BCG coverage in the study location) were screened by community health workers between July 2005 and April 2006. The main outcome measures were screening coverage, referral rates, return rates for second-stage screening and evaluation, yield and age at hearing loss diagnosis.

Olusanya, et al. (2008a) found that in total, 2003 (88%) of 2277 eligible infants attending the four BCG clinics were successfully screened between July 2005 and April 2006 at a mean age of 17.7 days, with no parent declining screening. The majority (55.2%) were born outside a hospital and, of such infants, 77% were born in traditional herbal maternity homes. The mean age at diagnosis was 51 days. The sensitivity, specificity and positive predictive value of the first screening stage were 80.4%, 99.7%, and 90.0%, respectively. Routine hearing screening of

infants attending BCG immunization clinics by community health workers was confirmed to be feasible and effective for the early detection of hearing loss in Lagos, Nigeria.

Swanepoel, et al. (2006) described a hearing screening program implemented at two immunization clinics in a South African community over a 5-month period, 3 days per week, in the Hammanskraal district. During the 5-month research period, 510 infants between the ages of 0 and 12 months were enrolled in the study, with coverage of 95% for all ears and 93% coverage for bilateral screening of subjects in the sample were obtained with OAE screening.

Friderichs, et al., (2012) evaluated the first systematic community-based infant hearing screening program in a developing South African community-based universal infant hearing screening program initiated at eight primary health care clinics in the in the Western Cape. The program was evaluated over a 19-month research period. During this time 6227 infants that were candidates for screening attended their 6, 10 or 14-week immunization visit at the relevant clinic. The study evaluated the efficacy of the program based on screening coverage, referral and follow-up rates and diagnostic outcomes according to guidelines specified by the Health Professions Council of South Africa 2007 Position Statement. Overall coverage rate across the eight clinics was 32.4% with 2018 infants (aged 0–14 weeks) screened. The mean age of the sample at first stage screen was 3.9 weeks of age and 13.5 weeks of age for first hospital visit. Friedrichs, et al., (2012) found that the community-based screening program was partially effective, although screening coverage was not sufficient.

Screening coverage in the current study was similarly not considered adequately high in the private health care sector in South Africa. Swanepoel, et al. (2007) performed a retrospective study of a universal neonatal hearing screening program at a private hospital in urban Gauteng,

South Africa over a 4 year period of time. Six thousand two-hundred and forty-one newborns were screened from 13,799 hospital births during the first 4 years. Ninety-four percent of these infants were from the well-baby nurseries. During the initial 22 months, while the service was subsidized as part of the hospital birthing package, coverage of 75% was attained compared to 20% during the subsequent 26 months. Feasibility and efficacy data on public sector hospitals in South Africa was not available.

Table 4 provides a summary of the above efficacy data.

Table 4. Summary of Efficacy Data Regarding Neonatal Hearing Screening in Sub-Saharan Africa.

	Olusanya, et al. (2008b)	Olusanya, et al. (2008a)	Swanepoel, et al. (2006)	Friderichs, et al., (2012)	Swanepoel, et al. (2007)
Country	Nigeria	Nigeria	South Africa	South Africa	South Africa
Setting	Hospital-based	Community-based	Community-based	Community-based	Hospital-based
Coverage	98.7%	88%	93%	32.4%	75%

6. *What obstacles are predominantly encountered in establishing successful neonatal hearing screening programs in sub-Saharan Africa?*

Commonly encountered obstacles are summarized in Table 5. Olusanya, et al. (2007)'s report on the progress towards early hearing detection in developing countries discusses a number of potential obstacles in the success of such programs. Finding suitable test environments especially in busy hospitals or community health centers may present challenges that must be managed creatively across potential locations.

Olusanya, et al. (2008b) reported that high ambient noise levels in the Lagos Island Maternity Hospital in Nigeria were indicative of the challenges of conducting neonatal hearing screening in inner-city environments in many developing countries. Olusanya (2010) presented an observational report on the potential impact of ambient noise levels with OAE screening instruments in hospital and community-based universal neonatal hearing screening programs in Lagos, Nigeria. The noise levels in both the hospital and the community centers were higher than the levels typically reported in developed countries. For example, the average noise levels in the special care baby unit (SCBU) of 65.4 to 76.9 dBA SPL significantly exceeded the 45 dBA recommended in the USA (American Academy of Pediatrics, 1997 as cited in Olusanya, 2010). Noise levels in the entire hospital also exceeded the 35 dBA daytime limit in patient treatment rooms recommended by the World Health Organization (WHO, 1999 as cited in Olusanya, 2010). However, the recorded levels are comparable to those reported in other developing countries. The high noise levels in the community centers were consistent with previous reports from Nigeria where daytime noise levels of up to 74.9 dBA have been reported (Ologe et al, 2006 as cited in Olusanya, 2010).

According to Olusanya (2010)'s report, finding a suitable section within the hospital ward was a major challenge due to the excessive ambient noise, primarily from the hospital's location on a very busy street with substantial human and vehicular traffic as well as trading activities of street hawkers, compounded by the fact that the windows in the hospitals were often kept open for ventilation. Within the hospital premises itself, incessant noise from crying/restless babies, activities of nursing staff in the open wards, and the human traffic on the adjacent stairway also contributed to the recorded noise levels. A separate room with air conditioning was made available by the hospital administrators for testing babies from the well baby nursery. The

air conditioner was powered on and off depending on the room condition at the time of screening. Even when the air-conditioner was powered during testing the average ambient noise was 60.3 dBA SPL, the lowest recorded in the entire hospital. The higher average noise level in the SCBU was attributable primarily to the incubators. It is most unlikely that this inner-city hospital like many others in poorly-resourced countries would be able to afford the substantial investment required to improve the acoustic environment to standards typically found in developed countries.

The cost of acquiring and maintaining screening equipment at community levels could also be a major concern for the rapid expansion of programs (Olusanya, et al., 2007). Typical OAE screeners cost about US\$3,000 and AABR about US\$8,000 excluding consumables and replacement parts. To ensure that screening is uninterrupted at least two of such screeners would be required even at the least busy birthing centers and many more would be required if the screening protocol combines both OAE and AABR (Olusanya, et al., 2007).

Government contributions to national health expenditure in developed countries range from an average of 80% in Europe to 45% in the United States (WHO 2006 as cited in Olusanya, et al., 2007). This contrasts sharply with the levels in developing countries, where government contributions can be as low as 26% in Nigeria (Olusanya, et al. 2007). In Nigeria, out-of-pocket spending accounts for at least 90% of private health expenditure. This data would suggest that it may be unethical to predicate the decision for introducing new health interventions in most developing countries solely on government financial capacity if the consumers ultimately will pay for the services (Olusanya, et al., 2007).

Olusanya, et al. (2007) discussed further that the attitude of parents also has a significant impact in optimizing the uptake of screening services. Initial enthusiasm for screening before hospital discharge may be short-lived due to poor parental education on the value of screening resulting in poor follow-up compliance. Sustaining physician support in environments overwhelmingly oriented towards treating fatal diseases is a major challenge.

Swanepoel, et al. (2007) reported that in a private healthcare setting in South Africa, a number of parents did not provide consent for screening. Swanepoel & Almec (2008) investigated maternal knowledge and attitudes amongst 100 South African mothers accessing the public healthcare system using a face-to-face survey. The questionnaire (16 items) probed knowledge of infant hearing loss, superstitious cultural beliefs, and attitude towards early detection and intervention for hearing loss. Results indicate that more than 50% of the sample correctly identified three common etiological factors (ear discharge, medication, congenital) for infant hearing loss. At least one superstitious cultural belief regarding a possible cause of hearing loss was held by 57%. Maternal attitude was overwhelmingly positive with 99% indicating the desire to have their baby's hearing screened after birth and a high acceptance of hearing aids (87%). The study demonstrated a need for increased maternal awareness regarding infant hearing loss but also a readiness for early hearing detection programs.

Olusanya, et al. (2007) reported that ensuring parents of babies who failed the screening test prior to discharge return for follow-up is a major problem in most developing countries. During the initial screening maternal consent may be occasioned by mothers simply not wanting to feel left-out since the majority of mothers are likely to consent. Return for follow-up may therefore serve as a more accurate index of parental commitment than the coverage achieved for the initial screening. Factors such as transportation costs, parental convenience and anxiety may

contribute to a high default rate for follow-up. Parents of children with severe-to-profound hearing loss are sometimes more cooperative when requested to attend follow-up appointments if their babies were tested later than 3 months of age as they may have already begun to suspect the child's hearing difficulty (Olusanya, et al., 2007).

On some occasions poor return rates are associated with the lack of an effective tracking system or poor communication between health professionals and the parents. Swanepoel, et al. (2007) reported that a small minority of cases parents were discharged from hospital before the screening could be conducted. This emphasizes the need for quality monitoring and control of screening programs against the benchmarked indicators. Meyer, et al. (2012) similarly discussed in their report on hearing screening in private sector hospitals in South Africa that hearing screening is not yet mandated or regulated in South Africa. Thus, there is a lack of program quality control and no systematic protocol for tracking parents and their babies to attend follow-up appointments which may contribute to poor follow-up compliance. In addition, insufficient support from other key health professionals such as family physicians and pediatricians may discourage parents from prioritizing hearing screening follow-up. The importance of tracking systems and parental education were stressed as well by Swanepoel, et al. (2006) regarding immunization clinics in South Africa and by Olusanya, et al. (2008a) regarding immunization clinics in Nigeria. These difficulties have also been reported in developed countries and programs have demonstrated increasing efficiency after implementing improved tracking systems and increasing awareness of hearing loss amongst healthcare professionals and families (Mehl & Thomson, 2002 as cited in Olusanya, et al., 2007).

Olusanya & Okolo (2006) discussed similar challenges faced specifically at immunization clinics in Nigeria. Difficulty was found in achieving an optimal follow-up rate

when the second-stage screening has to be conducted outside of the scheduled immunization clinic. For example, about 43% of babies scheduled for the second-stage were not returned to complete the protocol compared to 60% in South Africa. The logistics of conducting a second-stage screening are burdensome for both the often impatient parents and the overworked screening staff.

Olusanya, et al. (2004) discussed the issue of manpower shortages as unique to developing countries and one that must be addressed when initiating neonatal hearing screening programs. Friedrichs, et al. (2012) found in their evaluation of community-based infant hearing screening programs in South Africa that consistent short staffing at one of the clinics led to a low coverage rate. Friedrichs, et al. (2012) suggested that dedicated screening personnel may be necessary to ensure sufficient coverage rates, rather than using existing clinic personnel.

Theunissen, et al. (2008) reported that in public sector hospitals in South Africa, lack of equipment was the primary reason neonatal hearing screening was not performed, as reported by 38% of 86 speech therapy and audiology departments surveyed. Thirty-one percent of hospitals reported a lack of screening due to both equipment and staff shortages, while only 9% and 3% of hospitals reported a lack of screening solely due to either staff or equipment shortages, respectively.

Table 5. Summary of Commonly Encountered Obstacles to Neonatal Hearing Screening in Sub-Saharan Africa, as Reported by Currently Available Studies.

High ambient noise levels	Parental attitude	Return for follow-up	Manpower shortages	Lack of equipment
Olusanya, et al. (2008b); Olusanya (2010)	Swanepoel, et al. (2007)	Swanepoel, et al. (2007); Meyer, et al. (2012); Swanepoel, et al. (2006); Olusanya, et al. (2008a); Olusanya & Okolo (2006)	Friedrichs, et al. (2012)	Theunissen, et al. (2008)

7. *What are the rates of referral and follow-up and what are the typical settings to which neonates are referred in sub-Saharan Africa?*

Olusanya, et al. (2007) reported on the progress towards early hearing detection services in developing countries, including sub-Saharan Africa. Data was based on responses to a questionnaire eliciting information on the nature and scope of early hearing detection services from both hospital- and community-based screening programs. Olusanya, et al. (2007) found an 18% referral rate at discharge in Nigerian screening programs, and a 56.9% rate for return for follow-up. In South Africa, a 13.9% referral rate at discharge was found, and a 39.7% rate for return for follow-up.

Swanepoel, et al. (2006)'s investigation of infant hearing screening programs in two immunization clinics in South Africa found a 14% rate for referral for follow-up screening. Only 40% (27/68) of those subjects returned for a follow-up screening. Of the 27 subjects who did return for follow-up screening, three could not be screened due to restlessness and irritability and

were therefore referred for a second follow-up screening. Zero percent of the subjects referred for a second follow-up screening returned. Nine of the subjects (33%) who had returned for an initial follow-up screening were referred for a diagnostic assessment. Only one subject (11%) returned for the diagnostic assessment, and was evaluated with a diagnostic ABR. The study did not indicate the setting of the diagnostic assessment.

Friedrichs, et al. (2012)'s evaluation of community-based screening programs in the Western Cape, South Africa, found the overall first stage screen referral rate at clinic level was 9.5% (n = 191). The overall second stage screen referral rate for these subjects who were sent to tertiary hospital level dropped to 3% (n = 62). Referral rates varied greatly amongst the clinics from 2.6 to 23.9% at first stage screen and 0 to 18.8% at second stage screen. However in all cases, except for 1 clinic, the second stage screen referral rate dropped below 6%. The overall follow-up rate at clinic level was 85.1% and the follow-up rate of those subjects attending their initial appointment at tertiary hospital level was 91.8%. Follow-up rates varied amongst the clinics from between 50 to 100% at clinic level and 60 to 100% at tertiary hospital level. However in the majority of cases the follow-up rates at clinic and tertiary hospital level were above 80%.

Swanepoel, et al. (2007) reported on a universal neonatal hearing screening program in the private sector of South Africa. The retrospective study that took place over a four year period found an overall referral rate of 11.1%. The NICU referral rate was 6.7% compared to 11.4% in the WBN. There was a steady decrease in referral rate from the first to the fourth year of the study, decreasing by about 2 to 4% per year. Rescreens were completed at the hospital in 32% (219/694) of cases referring the discharge screen. Data was not available for the remaining infants because parents were provided a choice of follow up centers. A small percentage (9%) of

infants who received a rescreen was recommended for diagnostic testing. All 19 infants referred for diagnostic testing returned and 32% of these cases presented with sensorineural hearing loss.

Meyer, et al. (2012)'s national survey of early hearing detection services in the private health sector of South Africa found 80% of universal programs reported a referral rate of less than 5% for diagnostic assessments. Twenty-eight per cent of programs indicated that less than 20% of babies scheduled for follow-up testing returned. Follow-up return rates of between 21% and 69% were reported by almost half (44%) of programs. Follow-up return rates were reported to exceed 70% by only 28% of programs. The referral setting was not indicated by the survey.

Olusanya, et al. (2008b) found in their study of a hospital-based screening program in Lagos, Nigeria, that 44 babies out of the 1274 who completed the two-stage screening were referred, yielding a referral rate of 3.5%. Only 16% (7/44) of babies scheduled for a diagnostic evaluation returned, and all were confirmed with hearing loss. In Olusanya, et al. (2008a)'s study on community-based infant hearing screening in Lagos, the overall referral rate for diagnostic evaluation was 4.1%. Only 61% (50/82) of those referred returned for evaluation, and 45 of them were confirmed with hearing loss. In both the hospital- and community-based programs, diagnostic services were provided by a designated audiological center located within 5 kilometers of the hospital. Free transportation was provided from the hospital to the diagnostic center for all affected parents.

Olusanya, et al. (2008a) found in their report on community-based infant hearing screening in Lagos, Nigeria, that of the 2003 babies screened using TEOAE, 287 (14.3%) were referred for AABR. Of these 287, 57 (19.8%) passed, 82 (28.6%) were referred for diagnostic evaluation and 148 (51.6%) defaulted. Therefore, of the total population of 2003 babies

screened, 82 (4.1%) failed the two-stage screening process and were referred for diagnostic evaluation. Correspondingly, 82 of the 1855 (4.4%) who completed two-stage screening were referred for diagnostic evaluation. Thereafter, 50 of these 82 babies (61.0%) returned for diagnostic evaluation, and 45 (90%) of them were confirmed as having hearing loss. AABR screening was performed at one of the four community centers in the study, while diagnostic evaluations were performed at an established audiological center.

Okhakhu, et al. (2010) reported that at immunization centers in Benin City, Nigeria, a total of 400 neonates (218 males and 182 females) were screened for the presence of otoacoustic emissions in both ears. Ninety neonates (22.5%) were referred. When a neonate has a test result reading “Refer”, the test in this case was repeated a few minutes later and a persistent “Refer” was taken as an indication for further testing. Any neonate failing the test was requested to be made available by the mother within a week for repeat screening. Those failing the repeat screening were then referred for full audiological evaluation. Follow-up rates and the setting of the evaluation was not indicated by the study.

Table 6 provides a summary of the above referral and follow-up data.

Table 6. Rates of Referral and Follow-up in Sub-Saharan African Reports of Specific Neonatal Hearing Screening Programs.

	Swanepoel, et al. (2006)	Swanepoel, et al. (2007)	Olusanya, et al. (2008b)	Olusanya, et al. (2008a)
Country	South Africa	South Africa	Nigeria	Nigeria
Setting	Community-based	Hospital-based (private)	Hospital-based	Community-based
Referral rate	14%	11.1%	3.5%	4.1%
Follow-up rate	40%	100%	16%	61%

DISCUSSION

The consequences of congenital and early onset hearing loss can only be best avoided at individual and society levels if the incidence and prevalence is significantly reduced. However, the prevailing socioeconomic conditions and weak health-care systems in many countries in sub-Saharan Africa make a significant reduction in the disease burden via primary prevention unattainable within the foreseeable future. For instance, vaccinations against notable causes of congenital and early onset hearing loss such as meningitis, mumps, and rubella are rare in many countries (Olusanya, 2008). Primary prevention also has limited impact on genetic or hereditary etiologies such as Connexin 26, Pendred's, and Usher's syndromes, and perhaps is most challenging with idiopathic hearing loss (Dunmade et al, 2007; Sellars & Brighton, 1983, as cited in Olusanya, 2008).

In view of the limitations of primary prevention and current possibilities for early detection, secondary prevention is a complementary strategy. Secondary prevention involves actions to prevent congenital and early onset hearing loss from becoming a disability through early detection by infant hearing screening, and provision of appropriate and timely intervention (Olusanya, 2008). This early detection has been attained via neonatal hearing screening programs in many countries including the United States, as well as in a number of developing countries, such as Brazil and Oman (Olusanya, et al., 2007).

A first step in initiating neonatal hearing screening programs in any part of the world is to learn the status, availability, and specific nature of neonatal hearing screening programs in the region of interest. This systematic review was designed to determine those parameters in sub-

Saharan Africa by addressing the aforementioned research questions using information from currently available research.

1. What proportion of sub-Saharan African nations currently perform neonatal hearing screening?

Nigeria and South Africa, making up roughly 4% of sub-Saharan African nations, were the only countries currently performing neonatal hearing screening, greatly underscoring the lack of early hearing detection services in the region. The dearth of reports of neonatal hearing screening from the sub-Saharan region of Africa reflects a total lack of early hearing detection and intervention services and can be attributed to several factors including a high burden of infectious diseases, restricted resources and the lack of tertiary education for audiologists or other hearing health care specialists (Swanepoel, et al., 2009). Fagan & Jacobs (2009) presented an exhaustive study on the availability of ENT services in sub-Saharan Africa and found that the only sub-Saharan African nations that provide formal training programs in audiology are South Africa and Kenya, along with limited ENT services across the region, and with some countries having no ENT services at all.

2. What is the rank ordering of settings in which neonatal hearing screening is conducted in sub-Saharan Africa?

Swanepoel, et al. (2007) presented the first report on a hospital-based universal neonatal hearing screening program in the South African private healthcare sector. Private sector hospitals have since become the most likely setting for neonatal hearing screening in South Africa. Meyer, et al. (2012) found that in South Africa, 53% of private health care sector birthing units offer some form of hearing screening service, and only 14% provide true universal

newborn hearing screening. The private health care sector provides services to only 15% of the South African population (Meyer & Swanepoel, 2011 as cited in Meyer, et al., 2012).

Theunissen & Swanepoel (2008) estimated that 7.5% of public sector hospitals in South Africa provide some form of infant hearing screening and less than 1% provided universal screening. The public healthcare sector reportedly serves approximately 85% of South Africa's population. Theunissen & Swanepoel (2008) found that the most frequently reported reasons for the absence of a screening program were a lack of appropriate equipment and a shortage of staff.

Routine immunizations shortly after birth against tuberculosis with Bacille de Calmette-Guerin (BCG) vaccine, and against diphtheria-pertussis-tetanus (DPT) are widely promoted in sub-Saharan Africa. The DPT vaccinations are given in three doses at 6, 10, and 14 weeks, while the BCG vaccine is usually given at birth. The immunization routine for the BCG and DPT vaccinations is unique in that the coverage extends significantly beyond the population of babies born in health facilities in the majority of countries in sub-Saharan Africa. Olusanya (2008) argued that for infant screening purposes, evidence weighs heavily in favor of BCG clinics. Since the vaccine is given at birth, it allows screening to be conducted at the earliest possible time after birth when compared to other routine childhood immunizations. It also allows for a variety of screening protocols which includes AABR (Olusanya, et al., 2007). The clinic facilitates the early detection and confirmation of hearing loss even for a significant number of infants who present late for BCG immunization. DPT clinics may serve as a more effective follow-up platform for infants referred from the BCG clinics (Olusanya, 2008).

Immunization clinics have been proposed as an alternative to hospital-based settings for neonatal hearing screening, and are likely to rank as the third most common setting for neonatal

hearing screening in South Africa. However, in Nigeria, community-based programs such as in immunization clinics are likely to be more common than hospital-based programs, which is a reflection of Nigerian birthing patterns. As reported by Olusanya (2008), Nigeria's annual live births account for one-fifth of the total live births for sub-Saharan Africa. Roughly a third of Nigeria's births take place in institutions such as hospitals, reflecting an average of 36% of births being institutionalized for all of sub-Saharan Africa. South Africa has the highest proportion of institutionalized births in the region, with 92% of births being institutionalized. South Africa is also the most urbanized (60%) nation in the region, followed by Nigeria (49%) (Olusanya, 2008).

Olusanya, Emokpae, Renner & Wirz (2009) evaluated the cost effectiveness of hospital and community-based infant hearing screening programs in Lagos, Nigeria. The main outcome measure was cost per baby screened. Screening cost per child was lowest (US\$7.62) under community-based universal screening and highest (US\$73.24) under hospital-based targeted screening. Similarly, cost per child detected with hearing loss was lowest (US\$602.49) for community-based universal screening and highest (US\$4631.33) for hospital-based targeted screening. Based on these outcomes, Olusanya, et al. (2009) determined that community-based universal screening of infants during routine immunization clinics appears to be the most cost-effective model for early detection of hearing loss in low-income countries.

3. What is the rank ordering of procedures used for neonatal hearing screening?

The screening options for the early detection of hearing loss that best match the criteria for an ideal test, and have been used in various infant hearing screening programs in sub-Saharan Africa, consist of three objective technologies: transient-evoked otoacoustic emissions

(TEOAE), distortion product otoacoustic emissions (DPOAE), and automated auditory brainstem response (AABR) (Olusanya, 2008). Screening with TEOAE was the most commonly used procedure in both hospital- and community-based screening programs in Nigeria, while DPOAE was the most common procedure in South Africa.

In practice, an ideal hearing screening test would be simple to apply and safe, reliable, and valid. Health gains are likely to be maximized when primary prevention is complemented with effective screening programs (Olusanya, 2008). Kennedy, et al. (2006) studied the effects of universal newborn hearing screening on a large birth cohort in southern England. The authors found that the combination of TEOAE and AABR tests in two-stage screening, requiring AABR to be offered to those who failed in initial screen with TEOAE, has the most favorable combination of specificity, sensitivity, acceptability, and high coverage in settings with a wide range of birth rates.

However, although the above protocol has gained widespread application, it is important to note that a two-stage screening with TEOAE and AABR has been reported to miss about 23% of those with permanent mild hearing loss when a follow-up visit for repeat diagnostic evaluation occurred at about 9 months of age (Johnson, et al., 2005). Even children with more minimal hearing losses, such as those being related to otitis media, a unilateral sensorineural loss, or a mild bilateral sensorineural loss, are at risk for language delays and academic difficulties (Bess, 1985; Bess, et al., 1988).

Bansal, Gupta, & Nagarkar (2008) formulated a protocol for infant hearing screening in developing countries. Their study in India included both normal and high risk infants. TEOAE screening was performed on sleeping children in ideal test conditions. The pass criteria for the

TEOAE test were a signal to noise ratio of more than 3 dB at any three of the consecutive frequencies (1.0, 1.5, 2.0, 3.0 or 4.0 kHz); and reproducibility of the test by 50%. All the infants who passed the screening test were discharged from the study, whereas those who failed the test were called again after 1 month for a second TEOAE screening. The children who failed the second TEOAE test were taken up for an AABR which delivered 2048 broadband clicks. The presence of wave V at 70 dB nHL was used as pass criteria.

Bansal, et al. (2008) reported that that the pass rate for the TEOAE improved significantly as the infant's age increased at the first screening from 77.5% at 0—1 month of age to 92.8% at 2—3 months of age. On the second screening though the pass percentage increased from 84.7% at 1—2 months to 95.1% (2—3 months), there was no significant improvement beyond 3 months of age. Thus, community-based infant hearing screening that utilizes TEOAE can achieve greater specificity than screening a baby at birth. This is consistent with reports from Bess & Paradise (1994) that the specificity of TEOAE screening is lowest when the infant is between 24 and 48 hours. Bansal, et al. (2008) concluded that screening for hearing impairment at 3 months of age with TEOAE would significantly reduce the number of false positive cases, and thus maximize the benefits of a hearing screening program.

4. What professionals are most and least likely to oversee the neonatal hearing screening?

In both hospital- and community-based neonatal hearing screening programs in Nigeria, health workers without audiological experience are engaged and trained to undertake infant hearing screening services, while diagnostic and rehabilitative services are provided by audiologists, speech pathologists, and otolaryngologists. Olusanya (2008) discussed that a major

challenge confronting the vast majority of countries in sub-Saharan Africa is the acute shortage of health workers in all specialties against the backdrop of the fact that most of the population live in rural areas which are least attractive to health professionals. The only country that was not reported to have a critical shortage of health service providers was South Africa (WHO, 2006 as cited in Olusanya, 2008). South Africa was also the only country in the region to report health specialists such as audiologists and clinic nurses as the professionals who oversee neonatal hearing screening.

According to Fagan & Jacobs (2009), South Africa is home to 490 audiologists (1.021 per 100,000), while Nigeria is home to 5 (0.004 per 100,000). For purposes of comparison, the United Kingdom is home to 2,500 audiologists (4.1 per 100,000). This acute shortage of ear-care professionals portends the need for culturally-appropriate adjustments to ensure that the patients' interests are optimized within a multidisciplinary setting where the required specialists do not exist (Olusanya, 2008).

Moodley & Storbeck (2012) reported on the role that the neonatal nurse can play in newborn hearing screening programs in South Africa. The nurse can be trained to use equipment, namely OAE and AABR, to conduct the test appropriately. Having screeners on site and that have access on a daily basis to infants will ensure that fewer infants are lost to follow-up. If a refer result is obtained, for which no interpretation of the results is required, the nurse is able to counsel the parents immediately and inform them of the importance of returning for follow-up and diagnostic appointments.

5. What are the feasibility and efficacy data supporting neonatal hearing screening in sub-Saharan Africa?

Feasibility and efficacy data for neonatal hearing screening programs are best analyzed in view of the USA's Joint Committee on Infant Hearing (JCIH) recommended quality benchmarks that were established in 2007. These benchmarks have typically become the standard in evaluating the quality of newly implemented hearing screening programs. The recommended percentage of eligible newborns to be screened before hospital discharge within the first month of life was greater than 95%. The recommended percentage of newborns that failed the screening test and referred for diagnostic evaluation was less than 4% and the percentage for those who completed the evaluation was to be greater than 70%. The mean age of confirmation of hearing loss was to be less than 3 months (JCIH, 2007).

The hospital-based screening program in Lagos, Nigeria that was reported by Olusanya (2008b) was the only screening program in sub-Saharan Africa that exceeded the JCIH recommended quality benchmark, obtaining a screening coverage of 98.7%. Furthermore, Olusanya (2008b) reported that about 3.3% (44/1330) of the total population of babies screened were referred for diagnostic evaluation, meeting the JCIH's criteria of less than 4%. Olusanya, et al. (2008b) attributed much of the success of the program as being due to enthusiastic support and effective parental education by the nursing staff. The fact that the screening tests were non-invasive was highly welcome by doctors and nurses. Olusanya, et al. (2008b) cited that other contributory factors to the program's success included the absence of user fees for screening and follow-up service; the positive influence of the clear responses recorded for the majority of babies on prospective mothers; parent-to-parent communication; and testimonials of successful screening outcomes which increased the assurance for prospective mothers that the screening tests would not cause harm to their babies.

Other sub-Saharan African neonatal screening programs were not quite as successful when examined through the lens of the JCIH recommendations. For example, Swanepoel, et al. (2007)'s report on a screening program in the private health sector of South Africa attributed their reduced coverage rate of 75% to two main factors. Firstly, a number of parents did not provide consent to the screening, which can be avoided by providing parents with information about the necessity of early identification of hearing loss. Secondly, in a small minority of cases parents were discharged from hospital before the screening could be conducted. This emphasizes the need for quality monitoring and control of screening programs.

Swanepoel, et al. (2006)'s report on a neonatal screening program at immunization clinics in South Africa closely approached the JCIH recommendations by obtaining a screening coverage rate of 93%. However, in Friderichs, et al. (2012) investigation on community-based hearing screening in the Western Cape of South Africa, coverage significantly fell below the JCIH benchmarks, with a total coverage of only 32.4% across eight clinics. The authors attributed this partially to the fact that the nurses were heavily burdened with a variety of tasks and struggled to effectively combine screening with other regular duties often regarded as more important. High turnover of clinic staff as well as missed screening opportunities due to incorrect or inconvenient immunization times and shortage of immunization stock were also found to contribute to a lower coverage rate. Those clinics with higher coverage rates in the study had a dedicated day set aside in the week for screening and/or dedicated screening personnel with focused training who took ownership of the program.

All feasibility and efficacy studies on neonatal hearing screening programs essentially stem, however, from a landmark study conducted by Vohr, Carty, Moore, & Letourneau (1998). The authors evaluated the outcome of a hearing screen/rescreen program for all births in all 8

maternity hospitals located in the state of Rhode Island from 1993-1996. With legislation becoming effective on July 1, 1993, Rhode Island was the first state to mandate universal hearing screening for all infants born in any of the 8 maternity hospitals located in the state. The Vohr, et al (1998) study examined hearing screening data obtained with TEOAE to demonstrate the feasibility of a universal hearing screening program.

Infants in NICUs were screened the week before discharge, and infants in normal nurseries were screened between 6 and 52 hours of age with TEOAE. 99% of all infants were screened. Infants who passed the TEOAE were discharged from the program, while infants who passed but had a medical risk factor were referred for an audiologic assessment at 6 months of age. NICU infants who did not pass the TEOAE had an immediate ABR screen, followed by appropriate referrals. Normal nursery infants who did not pass the TEOAE were rescheduled for a second TEOAE in 2 to 6 weeks. If the infant did not pass the second TEOAE, an auditory brainstem response screen was performed, followed by appropriate referrals. The protocol for passing the TEOAE was defined as an emission between 1 and 4 kHz with 75% reproducibility. The protocol was modified in March 1995 to limit the criterion to between 2 and 4 kHz. This modification was due to the fact that the 1 kHz emission was difficult to collect because of noise interference as well as the fact that a lack of response at 1 kHz was not likely to be associated with a permanent sensorineural hearing loss (Vohr, et al, 1998).

The auditory brainstem response screen was performed with standard skin preparation and 3 electrode montage kept at <5000 ohms. The initial stimulus intensity was 60 dB HL. A response was scored if wave V was present within 1 SD of the expected norm for gestational age. The procedure was repeated at 30 dB HL for 60 dB HL passes and at 85 dB HL for 60 dB HL fails. Parents were informed about all procedures and the need to return for rescreening after

discharge, and they were provided with brochures and a video about the screening program (Vohr, et al, 1998).

During the four year time period that this retrospective analysis took place in, 111 infants were identified with permanent hearing loss, resulting in an impairment rate of 2 per 1000. The mean age of hearing loss confirmation decreased from 8.7 months to 3.5 months. The mean age of amplification decreased from 13.3 months to 5.7 months. Vohr, et al (1998) indicated the effectiveness of a two-stage universal hearing screen protocol to screen, track, identify, and habilitate infants in both NICUs and normal nurseries with significant permanent hearing loss.

6. What obstacles are predominantly encountered in establishing successful neonatal hearing screening programs in sub-Saharan Africa?

Olusanya, et al. (2007) reported that government contributions to national health expenditure can be as low as 26% in Nigeria. The authors argued that governments in low and middle income countries are unlikely to ever have adequate resources to cater for all of the healthcare needs of its citizens. The government can instead serve as facilitator of public-private partnerships and create public awareness and set standards for best practices. The government can also ensure that the training curricula of health professionals provide up-to-date skills for the broad spectrum of healthcare needs (Olusanya, et al., 2007).

Olusanya, et al. (2007) and Swanepoel, et al. (2007) both reported that parental attitude can have a significant impact on screening uptake as well as resulting in poor follow-up compliance. Olusanya, et al. (2007) discussed further that sustaining physician support in environments overwhelmingly oriented towards treating fatal diseases is a major challenge but

can be mitigated by government support through on-going public awareness campaigns on the value and efficacy of early detection and intervention of hearing loss.

Olusanya, et al. (2008b) and Olusanya (2010) reported that high ambient noise levels were a major challenge to successful neonatal hearing screening in both hospital and community-based programs. Ramesh, et al., (2009) examined the effectiveness and cost of implementing a noise reduction protocol in a neonatal intensive care unit in India. Low-cost noise reduction measures for neonatal intensive units consisting of behavioral initiatives such as moral persuasion among nurses to speak in low tones and to avoid shouting across a distance, as well as environmental modification such as the fitting of furniture legs with rubber shoes and replacement of metallic files with plastic files have been demonstrated to have the potential of keeping noise levels within 60 dBA, however this still exceeds the 45 dBA recommended in the USA.

Return for a follow-up diagnostic evaluation following referral from screening was the most common and significant obstacle experienced in neonatal hearing screening programs in the region. This will be discussed in the following section.

7. *What are the rates of referral and follow-up and what are the typical settings to which neonates are referred in sub-Saharan Africa?*

Olusanya, et al. (2008b) reported that approximately 32.3% of babies in the well-baby nursery in Nigeria initially failed TEOAE screening. The number was reduced to 3.1% following AABR screen. Similarly, 31.7% of the NICU babies failed TEOAE but this rate was reduced to 4.4%. Following the two-stage screening, 3.5% of the total population of infants that were screened were referred for a diagnostic evaluation. Olusanya, et al. (2008b) discussed that

the overall referral rate of 32.2% was higher than most TEOAE based screening programs in other developing countries. The authors speculated that the test environment was the probable cause of the high referral rate. The significant reduction in referral rates after AABR screening within the recommended target of 4% is consistent with the experiences from such two-stage screening programs worldwide and thus makes it as a protocol of choice for this population.

Swanepoel, et al. (2007) reported an overall referral rate of 11% in a private hospital setting in South Africa. The authors noted that employing a combined OAE and AABR or only AABR screening protocol would have resulted in lower discharge referral rates but the screening costs would be higher. Swanepoel, et al. (2006) similarly notes regarding their referral rate of 14% in South African immunization clinics, that a single OAE screen requires a second step screen to obtain acceptably low refer rates.

The JCIH recommended percentage of newborns that failed the screening test and referred for diagnostic evaluation was less than 4% and the percentage for those who completed the evaluation was to be greater than 70%. Both hospital- and community-based screening programs in Nigeria, evaluated by Olusanya, et al. (2008b) and Olusanya, et al. (2008a) respectively, met the JCIH criteria in terms of referral rate. Olusanya, et al. (2008b)'s evaluation of a hospital-based screening program in Lagos, Nigeria obtained a referral rate of 3.5%. Olusanya, et al. (2008a)'s evaluation of a screening program at immunization clinics in Nigeria obtained a referral rate of 4.1%, just barely meeting the JCIH quality benchmark.

Regarding follow-up rate, in Swanepoel, et al. (2007)'s report, all infants who were rescreened at the hospital and referred for a diagnostic evaluation, returned for the evaluation. However, only 32% of infants who were referred for the second-stage screen returned to the

hospital, while other infants rescreened at an audiologist of the parents' choice. No data was available for these remaining infants, emphasizing the importance of tracking infants scheduled for referrals who are at an increased risk of hearing loss. Programs must implement data management and tracking systems that will ensure infants are followed-up and the data documented. This is essential for accountable service provision to families and also to supply comprehensive data on program efficiency and effectiveness for quality control (Swanepoel, et al. 2007).

Olusanya, et al. (2007) discussed that the rate of return for follow-up after discharge from the hospital or the screening center is an index of how effective the tracking system is as well as the voluntary disposition towards the completion of the screening by parents or the logistics of returning to the screening centers. Low follow-up rates that did not meet the JCIH benchmarks were reported by all other studies, and were also reported as the most commonly encountered obstacle to neonatal hearing screening in sub-Saharan Africa. Olusanya, et al. (2008b)'s report achieved the lowest follow-up rate of all, with a rate of 16%. The authors speculated that this high default rate was possibly due to the fact that a significant number of mothers lived far from the hospital and were referred for specialist care during delivery from private hospitals. Olusanya, et al. (2008a) noted that factors such as inaccurate contact details, change of address, lack of family support, work constraints, the "inconvenience" of travelling and superstitious beliefs about childhood deafness accounted for their default rate of 61% falling short of the JCIH target of 70%.

Future Directions: A Model Demonstration Program

Olusanya (2011a) summarized the outcome of an informal consultation of the WHO in 2009, urging member states to promote programs for early hearing detection. The consensus was that where universal neonatal hearing screening was not immediately practicable, interim approaches consisting of targeted neonatal hearing screening based on physiological tests and/or questionnaire and behavioral techniques should be explored guided by evidence from local pilot studies. Regardless of the approach adopted, early hearing detection and intervention programs must necessarily be linked to existing health, social and educational systems in each country (Olusanya, 2011a).

Olusanya (2006) reported on current major global initiatives from United Nations agencies such as UNICEF and UNESCO that provide platforms and impetus for the promotion of neonatal hearing screening in the developing world. One of the five cardinal priorities of UNICEF is the promotion of optimal early childhood development because the organization believes that giving a child the best start in life lays the foundation for learning and school achievement. The organization expresses interest for the needs of children of who have been “excluded and made invisible” by hearing loss and other disabilities (UNICEF, 2005 as cited in Olusanya, 2006). UNESCO provides for an Early Childhood Care and Education (ECEE) initiative that was launched and adopted by 164 countries in 2000 (UNESCO, 2006 as cited in Olusanya, 2006), which is aimed at supporting children’s growth, development, and learning from birth. Initiatives such as these recognize the importance of early childhood development and can aid in extending neonatal hearing screening in sub-Saharan Africa and the developing world at large. The pilot studies that have been reported in this review represent an important way forward in addressing the lack of early hearing detection in sub-Saharan Africa.

The JCIH's 2007 position statement recommended heightened surveillance of all infants with risk indicators for either congenital or delayed onset hearing loss. The position statement recommends use of risk indicators for hearing loss for three purposes. The first use of risk indicators is for the identification of infants who should receive audiological evaluation but who live in geographic locations where universal hearing screening is not yet available. The second purpose of risk-indicator identification is to help identify infants who pass the neonatal screening but are at risk of developing delayed-onset hearing loss and, therefore, should receive ongoing medical, speech and language, and audiological surveillance. Third, the risk indicators are used to identify infants who may have passed neonatal screening but have mild forms of permanent hearing loss.

The presence of all risk indicators for acquired hearing loss should be determined in the medical home during early well-infant visits. Early and more frequent assessment may be indicated for children with CMV infection, syndromes associated with progressive hearing loss, neurodegenerative disorders, trauma, or culture positive postnatal infections associated with sensorineural hearing loss; for children who have received ECMO or chemotherapy; and when there is caregiver concern or a family history of hearing loss (JCIH, 2007).

However, Olusanya (2011) presented a review of relevant literature on the effectiveness of targeted neonatal hearing screening based on these JCIH risk factors. Evidence from the review suggested that countries in regions such as sub-Saharan Africa may be constrained in applying all of the risk factors. Notwithstanding their limited number and scope, universal neonatal hearing screening studies in this region have unveiled other risk factors not listed by JCIH consistent with the epidemiological profile in many developing countries. Universal neonatal hearing screening studies are therefore warranted in individual countries to establish

context-specific risk factors, their performance for screening purposes as well as operational issues related to effective implementation before embarking on targeted neonatal hearing screening where universal screening is not immediately practicable (Olusanya, 2011).

Kanji & Khoza-Shangase (2012) similarly reported in a retrospective study to determine the type and frequency of risk factors for a group of very low birth weight neonates at a hospital in South Africa that the list of risk indicators for hearing loss still requires constant modification. More detailed categorization in terms of severity is necessary as risk factors may be influenced by the resources, community and diseases present in different contexts during different time periods. The less frequently occurring risk factors need to be investigated further by audiologists as this may lead to growing evidence regarding the inclusion of additional risk factors on the high-risk register (Kanji & Khoza-Shangase, 2012).

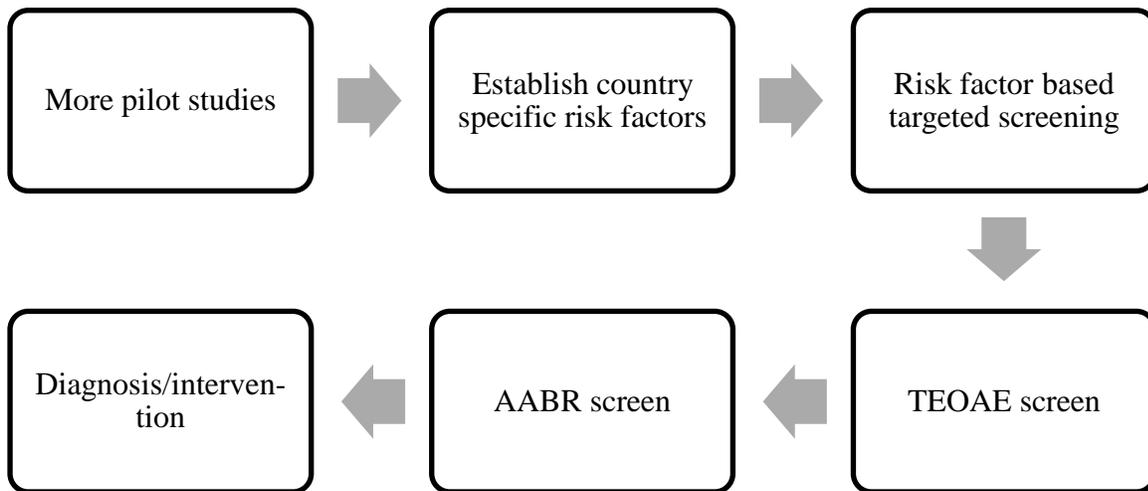
Based on the findings from this systematic review, recommendations for future neonatal hearing screening programs in sub-Saharan Africa and the developing world at large can be furnished. See Figure 2 for a diagram of the development of future screening programs. In developing a model program, it is essential that more pilot studies such as the ones cited in this review be conducted in more sub-Saharan African nations. As explained by Olusanya (2008), this can provide empirical evidence for engaging relevant government ministries for appropriate provisions for early detection and intervention of hearing loss. Furthermore, country specific risk factors for congenital and early onset hearing loss can be determined, along country specific practicalities of operating a neonatal hearing screening program. Once country specific risk factors are established, targeted neonatal hearing screening can be explored as a more practical option in areas where universal screening is less feasible.

A community-based setting, such as an immunization clinic, has been shown to be the most cost effective setting for newborn hearing screening in the developing world (Olusanya, et al., 2009). Pilot studies would, again, be necessary to confirm this in individual countries, as well taking into account the percentage of institutionalized births in each country, in order to determine the best setting for screening. A two-stage screening process utilizing an initial TEOAE screening followed by an AABR screening for referred babies has typically been cited as the most favorable combination in terms of specificity and sensitivity. Personnel without prior audiological experience can be trained on screening procedures, while diagnostic evaluations and interventions will be conducted by the appropriately trained professionals. Low cost noise reduction measures would be important as well in order to create an optimal testing environment and can assist in reducing referral rates. Having a specific day set aside for screening can be useful to help maximize coverage rates for screening. Programs must implement data management and tracking systems that will ensure infants are followed-up. Proper data management is crucial to a program's success.

The attitude of parents, along with professional staff involved with hearing screening, can greatly affect the success of a neonatal hearing screening program. These individuals must be presented with the necessary information and communication to ensure enthusiastic support. Olusanya (2008) made a number of pertinent suggestions that can address such issues. Promoting public awareness of the consequences of late detection of congenital and early onset hearing loss is essential and can be done through various forms of media. The author recommended the formation of an association of parents of children with deafness as a rallying force for advocacy and lobbying. Local governments can serve as facilitators of public-private partnerships to enable funding of screening programs. Lobbying for free import duties on

audiological equipment and related materials can also be helpful, as well as tax incentives for organizations that provide support for children with hearing loss. Fees associated with hearing screening can then either be eliminated or greatly reduced, further aiding in parental support of screening their babies for hearing loss.

Figure 2. Diagram of the Development of Future Neonatal Hearing Screening Programs in Sub-Saharan Africa.



CONCLUSIONS

This systematic review sought to determine and assess the status, availability, and specific nature of neonatal hearing screening programs in sub-Saharan Africa based on currently available literature. Several implications can be made for research and clinical purposes based on the findings of this systematic review.

Research Implications

The dearth of reports from this poverty stricken region of the world underlines the need for more universal neonatal hearing screening pilot studies across sub-Saharan Africa. These pilot studies can lead to the identification of country specific risk factors that can be utilized in targeted screening programs. The studies can also determine if a hospital-based or community-based setting is more appropriate. Most importantly, these pilot studies can establish the feasibility and efficacy of universal neonatal hearing screening in individual countries in the region, paving the way for a widespread application of newborn hearing screening services across sub-Saharan Africa.

Clinical Implications

Implementing both universal and targeted neonatal hearing screening programs in sub-Saharan Africa can potentially reach a wide range of infants across the region. This can allow for timely diagnosis of congenital and early-onset hearing loss, allowing for timely intervention of the hearing loss to commence. Such intervention can help avoid the consequences of untreated hearing loss in children and allow for more children to develop normal speech and language abilities along with healthy social and academic achievement.

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