

2016; 1(1): 2-12

Effective And Efficient Pre-School Hearing Screening: Essential For Successful Early Hearing Detection And Intervention (EHDI)

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Abstract

An unacceptable number of infants failing newborn hearing screening do not receive necessary follow-up services in a timely fashion as a result of loss to follow-up problems. In addition, a high proportion of children who pass newborn hearing screening later acquire hearing loss during the preschool years. Systematic pre-school hearing screening offers a logical strategy for detection of hearing loss among these children. Pure tone hearing screening of older preschool children has questionable test performance and validity. And, there is consensus that a behavioral technique is not feasible for routine hearing screening of younger preschool children. Otoacoustic emissions (OAEs) offer the most promising option for systematic hearing screening of the preschool population. Multiple advantages of OAEs are cited in support of their role in preschool hearing screening. This paper summarizes a new evidence-based and clinically feasible strategy for effective and efficient preschool hearing screening that relies on objective auditory tests.

Acronyms: AAA = American Academy of Audiology; ABR = auditory brainstem response; AABR = automated auditory brainstem response; ASHA = American Speech-Language-Hearing Association; ANSD = auditory neuropathy spectrum disorder; BBN = broadband noise; CDC = Centers for Disease Control and Prevention; DHH = deaf or hard of hearing; DP = distortion product; DPOAE = distortion product optoacoustic emissions; EHDI = Early Hearing Loss Detection and Intervention; HL = hearing level; LTFU = loss to follow-up; NIH = National Institutes of Health; OAE = otoacoustic emissions; SPL = sound pressure level; UNHS = universal newborn hearing screening

Rationale For Pre-School Hearing Screening

In the United States, universal newborn hearing screening (UNHS) has been a reality for more than a decade (White, 2014). The emergence of UNHS can be traced back to a convergence in the 1990s of multiple distinct developments. First, advances in hearing screening technology led to clinical trials of automated auditory brainstem response (ABR) and otoacoustic emissions (OAE) devices (Hall, Kileny & Ruth, 1987; Stewart et al., 2000: Vohr. Carty. Moore. & Letourneau. 1998: Vohr et al.. 2001). Second, several multidisciplinary groups such as the National Institutes of Health (NIH; Consensus Conference on Early Identification of Hearing Impairment in Infants and Young Children, 1993) and the Joint Committee on Infant Hearing (1994) began to support UNHS. Third, systematic investigations provided unequivocal evidence of the benefits of early intervention for children who are deaf or hard of hearing (DHH; e.g., Moeller, 2000; White, 2006; Yoshinago-Itano, Sedley, Coulter, & Mehl, 1998). These developments in the late 1990s contributed to the American Academy of Pediatrics endorsing UNHS and establishing benchmarks for UNHS programs (American Academy of Pediatrics, 1999). During the same time period, EHDI (Early Hearing Loss Detection and Intervention) grants were first authorized in the Newborn and Infant Hearing Screening and Intervention Act of 1999 and reauthorized through the Children's Health Act of 2000.

Serious Loss to Follow-Up Problems

Unfortunately, the era of UNHS in the United States has not yet led to universal diagnosis of and early intervention for children who are DHH. In other words, early intervention does not occur for many young children who are DHH. When infants and young children who are DHH are not diagnosed or do not receive early intervention services it is often referred to as a loss to follow-up (LTFU) problem.

There are at least three general explanations for LTFU. First, a small proportion of infants (~3% nationwide) are not screened at birth. Prominent reasons for missed hearing screenings are listed in Table 1. Although the percentage of babies who miss the birth screening is small, the actual number of babies is substantial. In 2013 more than 134,000 babies began their preschool years with unknown hearing status (Centers for Disease Control and Prevention [CDC], 2013), just like infants did before the era of UNHS. Among these children there were likely 400 or more who were DHH.

A second and equally serious problem is the substantial number of newborns who have a refer outcome at the time they leave the hospital, but never complete the diagnostic assessent process. There are a variety of reasons for why infants are lost to follow-up after a refer outcome on newborn hearing screening. Some of the important factors are listed in Table 1.

Table 1. Two General categories of factors contributing to loss to follow-up rates for infants born in the United States

 Missed Newborn Hearing Screening Parent refusal of newborn hearing screening Hospital discharge before hearing screening can be completed Transfer to another hospital before hearing screening can be completed. Infant does not undergo scheduled re-screening following initial refer outcome
 Loss to Follow-Up: Undocumented Diagnosis or Intervention of Hearing Loss Inappropriately high newborn hearing screening failure rate Infants are screened in one state who live in another state Parent misunderstanding or lack of commitment about the need for follow-up testing following a refer hearing screening outcome Physician misunderstanding about the need for follow-up testing following a refer hearing screening results is not shared with proper persons, including medical home, audiologists, hospitals and/or state EHDI
 program Inadequate number and geographical distribution of audiologists skilled, experienced, and equipped for diagnosis of and intervention of infant hearing loss Parent problems with transportation to diagnostic assessment
 Infants with no primary care physician who are essentially medically homeless Infants whose families lack health insurance and who cannot afford diagnostic services
 Parent refusal to consent to the diagnostic evaluation The diagnostic assessment cannot be completed due to technical issues that are encountered during the assessment or due to infant non-compliance when ABR testing under sedation is not an option Diagnostic assessment is not documented A report of diagnostic test results is not distributed to medical home, audiologists, state EHDI program, and/or those responsible for intervention

Note. ABR = auditory brainstem response; EHDI = Early Hearing Detection and Intervention.

As a result of these varied factors, an unknown number of children with hearing loss do not receive timely intervention services for lack of diagnostic information on hearing status. The CDC (2013) estimated that the nationwide LTFU rate in 2013 was 32.1% for diagnostic assessment and 25.8% for early intervention. These percentages may not accurately reflect the true status of the problem given concerns about the methods used to calculate loss to follow-up statistics.

Despite the uncertainty about the precise extent of the loss to follow-up problem, there is no question that an unacceptable number of infants do not receive necessary follow-up services in a timely fashion. Systematic programs for preschool hearing screening can play an important part in promoting early intervention for childhood hearing loss and minimizing the negative consequences for children who are lost to follow-up at some stage in the EHDI process.

Late Onset Hearing Loss

Another reason for expanding hearing screening programs for preschool-aged children is the surprisingly high proportion of children who pass newborn hearing screening but acquire hearing loss during the preschool years. For example, Fortnum, Summerfield, Marshall, Davis, & Bamford (2001) described a significant increase in prevalence of hearing loss from birth to school age. Up to 50% of children with hearing loss at age 9 passed newborn hearing screening. Bamford and colleagues (2007) and White (2014) also noted greater prevalence of hearing loss in the range of 6 to 10 per 1000 for school-age children versus 2-3 per 1000 for infants. And, according to Grote (2000), UNHS programs do not detect 10 to 20% of children with permanent hearing loss. Clearly, a substantial proportion of children who are DHH would be missed even if EHDI programs did not have any problems with LTFU.

There are a number of risk indicators for late-onset permanent hearing loss in the preschool years as delineated in the 2007 Joint Committee on Infant Hearing statement (JCIH). The term *delayed or late onset hearing* *loss* implies normal auditory function at birth with the rather abrupt onset of auditory dysfunction and associated hearing loss sometime during infancy or early childhood. Depending on the etiology, hearing loss may begin in one ear or both ears and may affect any frequency. Hearing loss often gradually progresses from slight to more serious during early childhood, and sometimes even into school age years.

Screening Protocol and Equipment Considerations

A pass outcome for screening with OAEs or automated auditory brainstem response (AABR) technology depends mostly on hearing status for a high frequency region. Distortion product (DP) or transient OAE screening is usually limited to measurement of cochlear activity within the range of about 2000 to 4000 Hz. Screening outcome for click-evoked AABR also is most closely correlated with auditory status within a similar frequency range. It's likely that a proportion of children with the diagnosis of late-onset hearing loss actually had undetected auditory dysfunction as newborn infants.

Factors putting children at risk for late-onset hearing loss are summarized in Table 2. Documentation of these risk factors is essential for prompt identification of hearing loss in young children, even in the era of UNHS. To summarize, a substantial number of infants with apparently normal hearing at birth will acquire hearing loss before they enter school. It's also likely that some infants with certain patterns of hearing loss in the perinatal period will pass newborn hearing screening with existing techniques. In any event, a remarkably high proportion of children passing hearing screening as newborn infants have hearing loss at school age. Systematic pre-school hearing screening offers a logical strategy for detection of hearing loss among these children.

Historical Review of Pre-School Hearing Screening

Early Recommendations

Multi-disciplinary support and general recommendations for hearing screening of preschool children date back to the 1980s. In 1989 the United States Department of Health and Human Services suggested a protocol for screening and assessment of speech, language, and hearing in preschool children that included a risk register, parental questions about their child's response to sound, and formal middle ear screening and hearing screening with pure tone audiometry. A 1984 American Academy of Pediatrics Policy Statement included endorsement of screening for middle ear disease and language development. The American Public Health Association in 1989 also supported preschool hearing screening.

In 1985 the American Speech-Language-Hearing Association (ASHA) released guidelines for identification audiometry that contained detailed recommendations about screening technique, personnel, and environment. The guidelines, limited to identification of hearing loss in children 3 years and older, specified that an audiologist must conduct pure tone hearing screening under earphones at an intensity level of 20 dB HL for frequencies of 1000, 2000, and 4000 Hz in an environment with maximum ambient noise levels of < 49.5 dB sound pressure level (SPL) at 1000 Hz.

1997 ASHA Guidelines for Audiologic Screening

Updating and extending the 1985 guidelines, ASHA published a 64-page document in 1997 that is the most comprehensive and, until recently, the most widely used set of guidelines for childhood hearing screening. The guidelines begin with an in-depth description of screening for outer and middle ear disorders for children birth through 18 years of age. It then includes sections devoted to hearing screening of children within four age groups: (a) newborn babies and infants from birth to 6 months, (b) infants and toddlers age 7 months through 2 years, (c) children age 3 to 5 years, and (d) school age children age 5 through 18 years. This article focuses on recommendations for children within the preschool age range of 6 months to 5 years—specifically who should conduct the screening, the technique recommended for screening, and the test environment.

The 1997 ASHA guidelines unequivocally state that, "Screening infants and children for hearing disorder and hearing impairment requires considerable professional expertise and technological sophistication. The Panel recommends that the screening process be designed, implemented, and supervised by an audiologist with the Certificate of Clinical Competence (CCC-A) from ASHA, and state licensure where applicable" (ASHA, 1997, p. 9). The guidelines emphasize repeatedly that it is "appropriate and necessary" that only certified audiologists conduct preschool hearing screening, particularly for younger children. Three categories of personnel are allowed for hearing screening of children within the age range of 3 to 5 years, including certified audiologists, certified speech pathologists, or "support personnel under supervision of a certified audiologist."

Consistent with earlier ASHA recommendations, the 1997 guidelines call for pure tone hearing screening with conditioned play audiometry at 20 dB HL for test frequencies of 1000, 2000, and 4000 Hz. Detailed instructions are offered in the guidelines for performing conditioned play audiometry. Criteria for a refer outcome are the absence of a reliable response for at least 2 out of 3 signal presentations at 20 dB HL for any frequency in either ear or inability to condition the child to the task. The 1997 guidelines refer to insert earphones as well as conventional supra-aural earphones for presentation of pure tone signals, although children who can be conditioned for visual reinforcement audiometry should be screened at 30 dB HL. *Pass* criteria are "... clinically reliable responses" at each

Table 2. Factors Associated with Delayed Diagnosis of Hearing Loss and Contributing to Late Intervention for Infants who Pass Newborn Hearing Screening

Caregiver concern regarding

- Hearing
- Speech and language
- Developmental delay

Family history of permanent hearing loss

Intensive care nursery stay of > 5 days and/or

- Extra-corporeal membrane oxygenation (ECMO)
- Assistive ventilation
- Exposure to ototoxic medicines
- Hyperbilibrubinemia requiring exchange transfusion

In utero infections, e.g.,

- Cytomegalovirus (CMV)
- Herpes
- Rubella
- Syphilis
- Toxoplasmosis

Craniofacial anomalies involving

- Pinna
- Ear canals
- · Ear tags and pits
- Temporal bone

Neurodegenerative disorders, e.g.,

- Hunter syndrome
- · Sensory motor neuropathies such as Friedrich ataxia and Charcot-Marie-Tooth syndrome

Culture positive post-natal infections associated with sensorineural hearing loss, such as bacterial meningitis

Head trauma requiring hospitalization

Chemotherapy with potentially ototoxic drugs

Physical findings associated with syndrome

Syndromes associated with hearing loss, e.g.,

- Neurofibromatosis
- Osteopetrosis
- Usher
- Waardenburg
- Pendred
- Alport
- Jervell
- · Lange-Nielson

Note. Adapted from Joint Committee on Infant Hearing (2007).

frequency in each ear (ASHA, 1997, p. 39). The guidelines also recommend screening in a calibrated sound field for children who do not comply with earphone placement. The 1997 guidelines specify that hearing screening must be done with calibrated audiometers, in an environment with sufficiently low ambient noise (< 49.5 dB SPL), and minimal visual and auditory distractions.

2011 American Academy of Audiology Childhood Hearing Loss Guidelines

The most recent document with recommendations relevant to preschool hearing screening is the 2011 American Academy of Audiology (AAA) Clinical Practice Guidelines on Childhood Hearing Screening. The 62-page AAA guidelines include detailed discussions of methods and techniques for childhood hearing screening, among them pure tone hearing screening, aural immittance measures (tympanometry and acoustic reflexes), and both distortion product and transient evoked otoacoustic emissions.

The 2011 AAA guidelines provide a very detailed section on pure tone hearing screening that begins with the statement, "Historically, the most widely preferred hearing screening procedure and the one that has been considered the gold standard is the pure tone audiometric sweep test ..." Expectedly, the AAA guidelines concur with earlier ASHA recommendations that children "chronologically and developmentally" age 3 or older undergo pure tone screening at 20 dB HL for test frequencies of 1000, 2000, and 4000 Hz. Response criteria and requirements for the test environment are similar to those stated in the ASHA guidelines. Tympanometry is recommended as a secondstage screening method for children who do not pass pure tone hearing screening. The 2011 AAA guidelines do not specifically provide recommendations for personnel involved in preschool hearing screening but they do acknowledge that non-audiologists often manage hearing screening programs.

Otoacoustic emissions are discussed in considerable detail in the 2011 AAA document with the recommendation that they should be used " ... only for preschool and school age children for whom pure tone screening is not developmentally appropriate (ability levels < 3 years). That is, OAEs are offered as an alternative for pure tone screening for young children" (p. 28). Also, follow-up screening with tympanometry is recommended for children who do not pass OAE screening.

The 2011 AAA guidelines cite limitations of OAE screening including the insensitivity of OAEs in ears with mild-tomoderate hearing loss (hearing sensitivity within the range of 20 to 50 dB HL), the difficulty of recording OAEs for test frequencies below 2000 Hz due to excessive ambient noise, and the possibility that children with auditory neuropathy spectrum disorder (ANSD) are missed with an OAE screening program. These alleged limitations of OAEs as a preschool hearing screening technique are addressed below in a discussion of new screening strategies.

Clinical Experience with Existing Guidelines

Published studies of preschool hearing screening highlight challenges in the application of existing guidelines. There is general acknowledgment in the guidelines that hearing screening of children younger than 3 years is not feasible with behavioral techniques. Representative studies in older preschool children are cited briefly here. Krishnamurti, Hawks, & Gerling (1999) described findings for 100 preschool children within the age range of 3 to 5 years. In some respects, the study reflects a "best case scenario" for preschool hearing screening with a pure tone technique. An experienced audiologist performed the screening according to ASHA guidelines in day care centers. Still, screening was unsuccessful for 3 children. Initial pure tone hearing screening refer rate was 24% and average hearing screening test times were 45 seconds for instruction prior to pure tone screening and another 60 seconds for the actual screening.

Allen, Stuart, Everett, & Elangovan (2004) reported hearing screening data for 1,462 children age 3 and 4 years old. Audiology or speech pathology graduate students performed hearing screening under the supervision of an audiologist in public preschool, day care, or Head Start centers following 1997 ASHA guidelines. An audiology supervisor performed tympanometry following pure tone hearing screening of each child. The supervising audiologist also performed pure tone screening of "difficult-to-test" children. Refer rates for this older preschool sample were 10% for otoscopy, 29% for pure tone screening, and 29% for tympanometry.

In one of the largest studies of preschool hearing screening, Serpanos and Jarmel (2007) reported data for 34,979 children age 3 to 5 years screened "on site in private, non-profit, or public preschools, day care centers, or Head Start programs" (p. 5). Graduate level audiology or speech pathology students conducted the screening under the supervision of a state licensed and ASHA-certified audiologist. The overall refer rate for pure tone and/or tympanometry screening was 18%, whereas 7% of the children did not pass both tympanometry and pure tone screening. In this study 2% of the children did not pass the pure tone hearing screening and an additional 3% could not be tested.

Halloran, Wall, Evans, Hardin, & Woolley (2005) described perhaps the most real world experience with hearing screening of older preschool children. Indeed, the study design purposefully did not require "standardization of screening techniques" because "screening in primary care settings is highly dependent on operator techniques and practice characteristics" (Halloran et al., 2005, p. 954). Data were reported for 1,061 children age 3 to up to 19 years who underwent pure tone hearing screening in 8 pediatric practices in Alabama, including 5 non-academic private practices and 3 that were within an academic setting. A trained research assistant conducted the screening with a calibrated audiometer coupled to supra-aural earphones pure tone hearing screening at 20 dB hearing level (HL; 1000, 2000, and 4000 Hz) in an examination room. Most (95%) of the children were screened with conventional technique whereas conditioned play audiometry was required for 5%. Neither gender nor race (African American versus white) was a factor in the likelihood that hearing screening was completed, but older children were more likely to complete screening. The rates for successful completion of hearing screening as a function of age were: ≥ 6 years = 100%; 5 years = 97%; 4 years = 93%; 3 years = 55%. That 45% of the younger children did not complete the hearing screening is quite discouraging. Of the total population, 67 children (7%) could not complete the screening procedure.

Interestingly, pass versus refer rates among children with normal development who could be successfully screened were consistently \ge 90% and unaffected by gender, race, or chronological age. Halloran et al. (2005), however, report a pass rate of only 67% for 21 developmentally delayed children. The overall failure rate was 10%, but a total of 162 children or 15% of the population either failed hearing screening or could not be tested. One of the rather surprising findings was the reluctance of pediatricians to refer children for further evaluation. As Halloran et al. (2005) noted: "The findings from this study are worrisome because physicians took no further action in more than 50% of the children who failed the hearing screening and more than 70% of the children who could not be tested" (p. 954).

Halloran et al. (2005) offered several possible explanations for the low follow-up rates, explanations that are relevant in any discussion of preschool hearing screening. Financial constraints presumably did not play a role in the decision against further testing because only infants with Medicaid or private health care insurance were enrolled in the study. However, some pediatricians may have elected to retest later as part of their typical follow-up. Also, physicians in private practice who have long-standing relations with families are presumably comfortable with continued monitoring for signs and symptoms of hearing loss. Additionally, physicians may believe that infants in generally good health and with higher socioeconomic status are at lower risk for hearing loss. Halloran et al. (2005) stated: "Lastly, little is known of the accuracy of conventional audiometry in the primary care setting; therefore, pediatricians may distrust their screening results and rely primarily on the history and physical examination or may seek stronger evidence of hearing loss in the form of a second failed screening prior to referral" (p. 953). Primary care physician attitudes about screening programs in general are explored in more detail in the next section.

Four years after the 2005 paper, Dr. Halloran and two of the authors published a follow-up article entitled: "The validity of pure-tone hearing screening at well-child visits" (Halloran, Hardin, & Wall, 2009). The authors raised serious questions about the value of pure tone hearing screening during well-child visits because of poor sensitivity (50%) and only fair specificity (78%), plus a high no-show rate for children referred for complete hearing evaluation by their primary care physician. Based on their data, Halloran et al. (2009) concluded, "Given the poor validity of pure tone audiometry, other methods of hearing screening should be considered for the primary care setting. One such option that practices and schools are increasingly using is otoacoustic emissions" (p. 161).

A New Strategy For Preschool Hearing Screening

Rationale for a New Strategy for Preschool Hearing Screening

Several strategies often used for preschool "hearing screening" in physician offices are not evidence-based options for accurate identification of hearing loss in young children (Eiserman, Shisler, et al., 2008). They include parent questionnaire and behavioral observation of responses to hand clapping, bell ringing, and other noisemaking devices. Otoscopy is an important part of the physical examination of young children but it clearly is not a measure of auditory function. Likewise, tympanometry is a useful measure of middle ear function, but it provides no information on hearing status. There is a role for tympanometry in conjunction with other hearing screening techniques in follow-up testing of children who yield a refer outcome with the primary hearing screening technique.

The collective experience from published studies (e.g., Brooks, 1971; FitzZaland & Zink, 1984; Fonesca, Forsyth, & Neary, 2005; Halloran et al., 2009) highlight at least five oft-cited serious challenges associated with reliance on the existing guidelines that recommend pure tone hearing screening for the preschool population.

- Audiologists are required for preschool hearing screening. However, audiologists are rarely available at sites where preschool hearing screening is conducted, such as day care centers, Head Start centers, or physician's offices. This challenge is significant, especially given the increasing demand for audiology services coupled with a stable or even declining supply of practicing audiologists (Windmill & Freeman, 2013).
- Acceptable ambient sound levels for pure tone screening are not always achievable in typical preschool hearing screening settings.
- •When pure tone screening is done, the time for each child, including instructions and data collection, may be 4 to 5 minutes or longer.
- Pure tone hearing screening doesn't consistently identify middle ear disorders, a common problem in the preschool population (Roush & Tait, 1985).
- A child's age, cognitive level, and language skills are significant factors in pure tone hearing screening. Because of these factors, hearing screening cannot be successfully completed for at least 3 to 5% of older preschool populations and can-not-test rates for chronologically or developmentally younger children are unacceptably high, even when an audiologist performs the screening.

Preschool hearing screening must be quick and simple for children age 3 years and younger (Northern & Downs, 1991). According to a national survey of pediatricians, guidelines are most likely to be adhered to if they are simple, feasible, and lead to proven improved outcomes (Flores, Leo, Bauchner, & Kastner, 2000). Halloran et al. (2005) reported the discouraging finding that pediatricians did not refer 59% of the children who failed the screening and 73% of the children who could not be tested. These statistics may reflect primary care physician distrust with screening outcome. Unfortunately, behavioral pure tone screening does not consistently meet minimal screening criteria even for older preschool children. There is consensus that a behavioral technique is not feasible for routine hearing screening of children in the range age 6 months to 3 years. However, a simple and fast technique

for hearing screening of younger preschool children is essential for systematic early identification of hearing loss.

Rationale for OAEs

OAEs offer the most promising option for systematic hearing screening of the preschool population from age 6 months to 5 years. Multiple advantages of OAEs can be cited in support of their role in preschool hearing screening. As an objective technique, OAE findings are not influenced by the many listener variables that confound hearing screening with a behavioral technique such as pure tone measurement. Listener variables include chronological or developmental age, cognitive level, language skills, motor abilities, and the combination of visual and auditory distractions in the environment. Sensitivity to the types of auditory problems commonly encountered in preschool children is a major advantage of OAEs. Abnormal OAE findings are very likely in children with middle ear dysfunction and/or with cochlear hearing loss involving outer hair cell dysfunction (American Academy of Audiology, 2011; Dhar & Hall, 2012; Hall, 2014). Many studies confirm the sensitivity of OAEs to even subtle outer hair cell dysfunction or damage (see Dhar & Hall, 2012 for review). Most etiologies for childhood hearing loss affect outer hair cell function.

Recording OAEs in young children is feasible and technically simple as evidenced by widespread application of OAEs in newborn infants undergoing hearing screening. Many hundreds of peer-reviewed research publications confirm that assorted personnel including volunteers, technicians, and nurses can successfully complete newborn hearing hearings using OAEs (Dhar & Hall, 2012). An audiologist is not required for OAE-based hearing screening. OAE screening test time is quick, often less than 30 seconds per ear. The signal averaging process employed during OAE measurement, in combination with a properly fitted probe, permits screening in test environments with substantial levels of ambient noise (American Academy of Audiology, 2011). OAE devices are easily portable and often hand-held. Also, OAE test outcome is documented with a display that can be stored electronically, interfaced with data management systems, and printed immediately.

Dozens of articles describe the application of OAEs in preschool hearing screening. Transiently evoked OAEs were recorded in most of the earlier studies published in years up to about 2001. More recently distortion product otoacoustic emissions (DPOAEs) have emerged as the technique of choice for preschool hearing screening (e.g., Bhattia, Mintz, Hecht, Deavenport, & Kuo, 2013; Dille, Glattke, & Earl, 2007; Eiserman, Hartell, et al, 2008; Foust, Eiserman, Shisler, & Geroso, 2013; Hunter, Davey, Kohtz, & Daly, 2007; Janssen, 2013; Kreisman, Bevilacqua, Day, Kriesman, & Hall, 2013; Lyons, Kei, & Driscoll, 2004). Collectively these papers confirm the feasibility and usefulness of DPOAEs for hearing screening in the preschool population.

Two representative studies in different preschool populations are cited here. Kreisman and colleagues (2013) performed hearing screening of 198 children (mean age 4.5 years) in 8 different facilities using pure tones with a conditioned play technique and also with a DPOAE protocol. Several findings of this study highlight the advantages of DPOAEs compared to pure tone hearing screening. In addition to the subjects for whom data were reported, two children successfully screened with DPOAEs could not be tested with pure tones. A total of 57 children failed DPOAE screening whereas only 21 children failed pure tone hearing screening, but none of the children who failed pure tone screening passed DPOAE screening. Sensitivity to hearing loss appeared greater for DPOAEs than for pure tones. Also, average hearing screening time for both ears was less than 1 minute for DPOAEs but over 3 minutes for the pure tone technique.

Foust et al., (2013) reported findings for DPOAE hearing screening in primary care medical settings. Subjects included 848 children (842 in the target population of < 5 years of age and four older siblings) primarily from families whose incomes were at or below the federal poverty level. Audiologist-trained technical staff conducted DPOAE screenings at well-child visits, illness visits, or ear/hearing visits to the primary care physician. As expected, failure rates varied depending on the reason for the physician visit-10% for well-child visits, 13% for illness visits, and 85% for ear/hearing visits. Children who did not pass the initial screening received follow-up screening. Five percent of all children did not pass the final screening. Three children were identified with permanent hearing loss (one was < 5 years of age and two were 5 years old). The study provides further evidence that OAEs offer a feasible approach for hearing screening of young preschool children.

An OAE Protocol for Efficient and Effective Preschool Hearing Screening

Acknowledging the challenges of pure tone screening in young children and those with special needs, the 2011 AAA Clinical Practice Guidelines for Childhood Hearing Screening cited the need for an alternative technique such as OAEs. The AAA guidelines reviewed the literature about hearing screening of young children with OAEs, including measurement techniques, screening considerations, test environment, and time. Three limitations of OAEs as a screening technique are cited in the 2011 Guidelines.

One limitation is the difficulty of recording OAEs in the low frequency range (< 1000 Hz) due to contamination from physiological and ambient noise. The same limitation also applies to pure tone hearing screening in the preschool population. ASHA and AAA guidelines recommend the use of pure tone stimuli of 1000, 2000, and 4000 Hz, but not 500 Hz. Published research shows that DPOAE measurement for test frequencies of 2000 Hz and above is adequately sensitive to middle ear dysfunction and

cochlear hearing loss affecting lower frequencies (see Dhar & Hall, 2012 for review). Although DPOAE are plotted as a function of the higher of the two test frequencies (f2), the actual distortion product that is measured arises from a lower frequency region in the cochlea as predicted with the equation: 2f1 - f2. In other words, the DP frequency is always lower than either of the two stimulus frequencies (f1 or f2).

Another limitation cited in the 2011 AAA Guidelines is the possibility that children with ANSD will be missed with reliance on OAE screening. Although this possibility exists, it is remote due to the rather low prevalence of ANSD, particularly in the well-baby nursery population. It is not reasonable to insist that a hearing screening strategy designed for detection of relatively few children with ANSD be used for all children. Almost all babies with ANSD who are admitted to an intensive care nursery will be identified and diagnosed within the perinatal period. Consideration of JCIH (2007) recommendations offers valuable guidance in addressing this limitation. A preschool child at risk for ANSD who has not yet been diagnosed can presumably be identified based on a "yes" answer to one or more simple questions: 1) Did the child require admission to an intensive care nursery at birth? 2) Is there any evidence of a neurological problem? 3) Does the child have an older sibling with known hearing loss? Children who are at risk for ANSD should undergo pure tone hearing screening, if feasible. At risk children who cannot be tested with a behavioral technique like pure tone screening, or even those who can, should then be tested with acoustic reflexes. Absent acoustic reflexes and/or abnormal pure tone thresholds would prompt a referral for comprehensive audiologic and medical assessment.

The third limitation cited in the 2011 AAA Guidelines is the possibility of recording an apparent OAE in children with mild-to-moderate hearing loss. The Guidelines caution

that pass/fail criteria in OAE-based preschool hearing screening must be "chosen carefully to maximize sensitivity and specificity" (p. 32). Clearly, a preschool hearing screening technique must have the best possible test performance. The problem with false-negative screening errors (i.e., a pass outcome in children with some degree of sensory hearing loss) is associated with reliance on a pass/fail criterion that is based on the relative difference between OAE amplitude versus noise floor levels, and without regard to the absolute OAE amplitude value. Most published studies in neonatal and preschool hearing screening have employed a pass criterion limited to an OAE-to-noise floor difference of > 3 or > 6 dB SPL.

A simple strategy for increasing sensitivity to varying degrees of sensory hearing loss is the addition of a second criterion involving the absolute amplitude of OAEs. Sensitivity of OAE screening to even mild sensory or conductive hearing loss is achieved with criteria for a pass outcome of an OAE amplitude minus noise floor difference of 6 dB SPL plus the requirement for an absolute OAE amplitude of \geq 0 dB SPL. Building both of these requirements into the automated pass-fail algorithms of DPOAE screening equipment could be done easily by manufacturers if there were a demand for it. Long-standing research on the relation between OAE amplitude and hearing threshold levels supports the application of these two criteria in combination for identification of persons with any degree of sensory hearing loss involving the outer hair cells (Gorga et al., 1997).

The application of an absolute amplitude level of 0 dB SPL to differentiate children with no hearing loss versus some degree of sensory hearing loss is illustrated in Figure 1. The dashed vertical line depicts the decision criterion of 0 dB SPL. Most children with hearing thresholds of less than 20 dB HL within the region of the OAE test frequencies have OAE amplitudes \ge 0 dB SPL. As with any sensitive

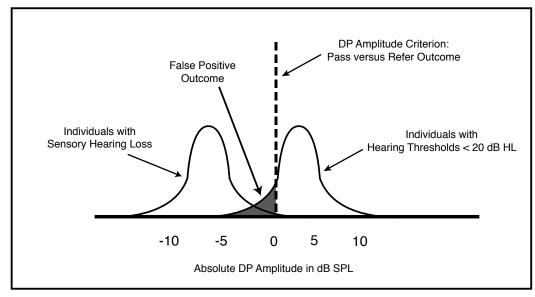


Figure 1. Pre-School Hearing Screening with OAEs

Note. DP = distortion product; HL = hearing level; SPL = sound pressure level.

screening measure, there is a possibility that a child with normal hearing will not meet this criterion. Among the common explanations accounting for a false-positive hearing-screening outcome is middle ear dysfunction. Insisting on a rather rigorous criterion of ≥ 0 dB SPL for absolute OAE amplitude in defining a pass outcome enhances screening detection of children with sensory hearing loss. Indeed, sensitivity of this OAE strategy for identifying middle ear or cochlear auditory dysfunction in preschool children may well exceed the sensitivity of pure tone hearing screening.

To summarize, the best use of OAE screening for young children would include the use of pass-fail algorithms that incorporate two criteria for pass. First is to document the presence of OAE activity with verification that OAE amplitude for the test frequencies is at least 6 dB greater than noise floor at the same frequencies. The second criterion, taken only for children who meet the first criterion, is to document that absolute OAE amplitude for the test frequencies is at least 0 dB SPL.

Closing Comments

The EHDI process is not flawless. Some children do not undergo hearing screening within the first month after birth even in the current era of UNHS. Two more serious problems compromise the goals of EHDI programs. One double-pronged problem is the rather sizeable proportion of children failing newborn hearing screening who are lost to follow-up before diagnostic hearing testing is completed or before intervention for hearing loss is implemented. Another problem is that a substantial number of children who had normal hearing at birth acquire a lateonset hearing loss. Thus, there is a strong rationale for widespread and systematic preschool hearing screening. Preschool hearing screening offers a viable strategy for early detection of childhood hearing loss beyond the newborn period.

A new evidence-based and clinically feasible strategy for effective and efficient preschool hearing screening is summarized in Table 3. The strategy relies on OAEs as the primary tool for hearing screening of all preschool children from age 6 months through 5 years. Pass/fail criteria used in OAE analysis are selected with the objective of

Table 3. A New Feasible Evidence-Based Strategy for Effective and Efficient Hearing Screening in Preschool Children

6 Months to 4 Years Primary Screening Technique: Distortion product otoacoustic emissions (DPOAEs) Stimulus intensity: L1 = 65 dB SPL; L2 = 55 dB SPL • F2 frequency region = 2000 to 5000 Hz • Frequencies per octave = 4 Pass Criteria o DPOAE amplitude = >0 dB SPL o DPOAE – noise floor = > 6 dBSecondary Screening Techniques for Refer Outcome Tympanometry · Acoustic reflex for broadband noise signal as indicated Otoscopy as indicated ≥ 4 Years Primary Screening Technique: Distortion product otoacoustic emissions (DPOAEs) Stimulus intensity: L1 = 65 dB SPL; L2 = 55 dB SPL F2 frequency region = 2000 to 5000 Hz • Frequencies per octave = 4 Pass Criteria o DPOAE amplitude = >0 dB SPL o DPOAE – noise floor = > 6 dBFollow-up Techniques for Children Who Do Not Pass DPOAE Tympanometry • Pure tone hearing screening at 20 dB HL if possible · Acoustic reflex for broadband noise signal if indicated Otoscopy as indicated

identifying children with hearing loss equal to or greater than 20 dB HL, a screening objective common also to the pure tone method. Tympanometry is performed for all children who do not pass the initial OAE screening in order to identify those with middle ear dysfunction that is often transient or successfully treated medically. The specific technique selected for follow-up to screening is age-dependent for children who do not pass an initial OAE screening who also have normal tympanograms and probably normal middle ear function.

For younger children under the age of 4 years, the followup should be done using acoustic reflex measurement. Acoustic reflex screening is conducted with a broadband noise (BBN) stimulus. BBN-evoked acoustic reflexes offer a quick and objective method for detection of likely sensory hearing loss in children with normal middle ear function as inferred from tympanometry (Hall, Berry, & Olson, 1982; Hall & Swanepoel, 2010; Kei, 2012). Pure tone hearing screening testing is the follow-up technique of choice for children of 4 years or older who do not pass OAE screening but who have normal tympanograms. Technological advances in pure tone hearing instrumentation (Wenjin et al., 2014) offer an opportunity to avoid some of the wellappreciated drawbacks associated with conventional pure tone hearing screening of preschool children detailed above.

Upon the completion of accurate OAE screening and follow-up of preschool children as just reviewed, recommendations in existing documents (e.g., JCIH, 2007; American Academy of Audiology, 2011; American Academy of Audiology, 2013) provide ample guidance on protocols for medical and audiological referral of infants and hearing screening program management.

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