



A Multisite Study to Examine the Efficacy of the Otoacoustic Emission/Automated Auditory Brainstem Response Newborn Hearing Screening Protocol: Recommendations for Policy, Practice, and Research

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Purpose: This article examines whether changes in hearing screening practices are warranted based on the results of the recent series of studies by J. L. Johnson, K. R. White, J. E. Widen, J. S. Gravel, B. R. Vohr, M. James, T. Kennalley, A. B. Maxon, L. Spivak, M. Sullivan-Mahoney, Y. Weirather, and S. Meyer (Johnson, White, Widen, Gravel, James, et al., 2005; Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005) that found a significant number of infants who passed an automated auditory brainstem response (A-ABR) screening after failing an initial otoacoustic emission (OAE) screening later were found to have permanent hearing loss in one or both ears. **Method:** Similar to the approach used by F. H. Bess and J. Paradise (1994), this article

addresses the public health tenets that need to be in place before screening programs, or in this case, a change in screening practice (use of a 2-step screening protocol) can be justified.

Results: There are no data to suggest that a 2-step OAE/A-ABR screening protocol should be avoided.

Conclusion: Research is needed before any change in public policy and practice surrounding current early hearing detection and intervention programs could be supported.

Key Words: newborn hearing screening, otoacoustic emissions, automated auditory brainstem response, visual reinforcement audiometry

A key event in the advancement of universal newborn hearing screening (UNHS) programs in the United States occurred in 1993 when the National Institute on Deafness and Other Communication Disorders within the National Institutes of Health (NIH) convened a consensus conference that resulted in the publication and dissemination of the NIH consensus statement *Early Identification of Hearing Impairment in Infants and Young Children* (NIH, 1993). This document supported (a) the screening of all neonatal intensive care unit infants before hospital discharge; (b) the screening of all infants by 3 months of age, preferably before hospital discharge; (c) use of a hearing screening protocol that consisted of testing using evoked otoacoustic emission (OAE) technology followed by testing using auditory brainstem response (ABR) technology for all infants who failed the OAE screening; (d) comprehensive intervention programs for infants and children with hearing loss; (e) ongoing surveillance for hearing loss throughout childhood; and (f) education of primary care providers on early signs of hearing loss in children.

Early hearing detection and intervention (EHDI) programs developed in later years have incorporated most of the consensus statement recommendations. In particular, many newborn hearing screening programs have adopted the general recommendation of the consensus statement to use both OAE and ABR in a newborn hearing screening protocol. The use of a two-step, two-technology protocol has resulted in a lower hearing screening failure rate at hospital discharge than OAE screening alone (e.g., Gravel et al., 2000) by separating infants with temporary auditory deficits associated with debris or fluid in the middle ear in the newborn period from those who were likely to have permanent congenital or neonatal onset bilateral or unilateral hearing loss of 30–40 dB HL or greater (Dalzell et al., 2000; Finitzo, Albright, & O'Neal, 1998; Joint Committee on Infant Hearing, 2000; Prieve et al., 2000).

The increase in newborn hearing screening programs and the resultant issues that have emerged surrounding the direct screening of hearing before hospital discharge as well as the follow-up, comprehensive audiologic assess-

ment and long-term management of neonates and infants with hearing loss have continued to challenge professionals interested in the implementation of quality EHDI programs. Now that more than 90% of all newborns are screened for hearing loss before leaving the hospital (National Center for Hearing Assessment and Management [NCHAM], 2005), concern has been raised about whether an automated ABR (A-ABR) screening pass following the fail of an OAE test may miss some infants with permanent hearing loss (PHL) in one or both ears who may benefit from early intervention.

Based on individual case reports as well as published outcomes from a clinical trial of hearing screening technologies (Norton et al., 2000b), the Association of Teachers of Preventive Medicine through the Centers for Disease Control and Prevention funded a multicenter prospective study of the audiologic status of infants who as neonates failed OAE but passed A-ABR hearing screening. The results of that investigation (Johnson, White, Widen, Gravel, James, et al., 2005) and subsequent articles (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005) supported previous reports that neonatal hearing screening tests will miss a proportion of infants with permanent forms of hearing loss (Norton et al., 2000a). White et al. (2005) estimated that 23% of infants with PHL in infancy would not be identified using the common OAE/A-ABR protocol.

The majority of the infants (over 80%) who were not identified by the OAE/A-ABR protocol had confirmed mild bilateral and unilateral PHL (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005). There has been growing concern among professionals over school-age children with mild bilateral or unilateral forms of PHL. Evidence suggests that for a sizeable proportion, auditory-based communication, social, and academic deficits may be present (Bess, Dodd-Murphy, & Parker, 1998; Bess & Tharpe, 1984; Davis, Reeve, Hind, & Bamford, 2002). While there is increasing evidence supporting the importance of early intervention for children with moderate and greater degrees of PHL (Moeller, 2000; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998), as well as

the existence of early sensitive periods for auditory input (Sharma, Dorman, & Kral, 2005), there is little known about the effects of mild forms of bilateral or unilateral hearing loss on the development of infants and young children.

We suggest that the findings of the series of studies presented here (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005) require clinicians to examine current public policies and practices directed at the early identification of hearing loss. These studies necessitate that professionals ask the question of whether missing more than 20% of infants with congenital and early onset hearing loss in infancy, albeit primarily mild in degree, is acceptable. The answer to this question, however, is not straightforward.

Method

A commentary written by Bess and Paradise in 1994 to the editor of *Pediatrics* following the publication of the NIH 1993 consensus statement is a relevant framework for the examination of whether a change in hearing screening policy is warranted. Their critique, titled “Universal Screening for Infant Hearing Impairment: Not Simple, Not Risk-Free, Not Necessarily Beneficial, and Not Presently Justified,” explored the evidence supporting the NIH consensus statement (1993) that advocated UNHS. Their article questioned whether the available literature and practical experiences of screening programs at that time were sufficient for a recommendation for UNHS and whether the practice was justified on “grounds of practicability, effectiveness, cost and harm-benefit ratio” (Bess & Paradise, 1994, p. 330).

Using the same criteria and public health screening tenets as Bess and Paradise (1994), we address whether changes in our current UNHS practices are warranted based on the Johnson, White, Widen, Gravel, James, et al. (2005) study and the reports in Johnson, White, Widen, Gravel, Vohr, et al. (2005); White et al. (2005); and Widen et al. (2005). The successful implementation of newborn hearing screening and other EHDI initiatives across the country has increased the desire of professionals to identify all cases of PHL in early life so that families have accurate information about their child’s hearing status and can make informed decisions about their various opportunities for intervention. Examining our current EHDI practices following the Bess and Paradise approach is useful in directing our thinking about whether a modification of the now common OAE/A-ABR screening protocol is warranted, or whether other approaches to identifying children with PHL in infancy might be more effective and practical.

Importance of Early Mild Sensorineural Hearing Loss

Since 71% of the infants with hearing loss that failed OAE and passed A-ABR in the Johnson, White, Widen, Gravel, James, et al. (2005) study were classified as having mild auditory deficits, this section focuses on whether it is

important to detect this degree of PHL during infancy. The relevant public health tenet examined by Bess and Paradise (1994) with regard to UHNS is whether the disorder for which screening is being considered is important. In this section, we explore whether mild hearing loss has important consequences for the developmental outcomes of children.

Studies of school-age children with bilateral mild or unilateral hearing loss suggest that several domains may be affected, including speech and language abilities, speech perception, functional auditory abilities, academic performance, listening effort, social-emotional development, motor abilities including early locomotion and later balance and coordination, and families’ quality of life (e.g., Bess et al., 1998; Bess & Tharpe, 1984; Davis et al., 2002; Hicks & Tharpe, 2002). If grade retention and support through resource service rates are considered as indicators of the incidence of children with milder forms of hearing loss who experience negative consequences, as many as 50% of school-age children with these types of PHL may be affected (e.g., Bess et al., 1998; Bess & Tharpe, 1984; Lieu, 2004).

Limited information is available on the effects of mild bilateral or unilateral hearing loss in infants and pre-school-age children. Young children with moderate to profound degrees of unilateral hearing loss are the subjects of available reports, with some but not all authors citing delays in language development as sequelae (see Lieu, 2004, for a review). A recent report (Stredler-Brown, 2004) on a small number of children from the Colorado Children’s Hearing Intervention Project suggests that unilateral hearing loss may negatively affect emerging speech and language development in some children with unilateral hearing loss in the infant-to-3-year age group.

Thus, while a body of work suggests that about half of elementary and middle school-age children with mild bilateral or unilateral hearing loss have some degree of academic or social-emotional difficulties, as yet there is little direct evidence in larger sample sizes using prospective research designs that mild forms of bilateral or unilateral hearing loss are related to developmental delays in the infant and pre-school-age population. There are no studies concerning which individual infants and toddlers with mild forms of hearing loss are more likely to experience school-related problems at older ages and what the influence of other intrinsic and extrinsic variables is on these children’s developmental outcomes.

Screening Tests for Hearing Loss in Infants

This section examines whether existing screening procedures are “safe, acceptable, simple, reliable, valid, reasonably low in cost and practicable” (Bess & Paradise, 1994, p. 331). Currently, there are two technologies used in hearing screening: OAE (either transient evoked [TEOAE] or distortion product [DPOAE]) and ABR. Both technologies are now available in screening devices that are automated and present screening results as a pass-fail outcome. The presence of evoked OAEs indicates healthy

outer hair cells in the cochlea, and recordable OAEs are highly (but not always) correlated with normal hearing in adults and are not always absent when cochlear hearing loss exists (Gorga et al., 1997). Generally, however, a screening OAE pass is interpreted as indicative of a generally intact sensory, middle, and outer ear mechanism, as the test requires that acoustic signals be transmitted unobstructed forward to and the acoustic emission back from the cochlea.

The ABR, an electrophysiological response, is typically in the submicrovolt range and arises from the synchronous activity of the auditory nerve and auditory brainstem. With presently available A-ABR equipment, the presence of sensory hearing loss of moderate degree or worse or the presence of a neural disorder usually results in a failed screening test. External and middle ear conditions are less likely to affect the recording of an ABR than the recording of OAEs in newborns (Chang, Vohr, Norton, & Lekas, 1993; Doyle, Burggraaff, Fujikawa, Kim, & MacArthur, 1997; Sutton, Gleadle, & Rowe, 1996; Thornton, Kimm, Kennedy, & Cafarelli-Dees, 1993). Aspects of the OAE and A-ABR screening tests are examined below and discussed relative to their usefulness for the detection of PHL in the neonate and, in particular, the identification of permanent mild bilateral or unilateral forms.

Safety

Similar to Bess and Paradise's conclusion in 1994, no issues or concerns have been raised about the safety of either of the two technologies (OAE and A-ABR) for screening hearing in neonates. None of the SPLs of test stimuli generated by either technology used in screening devices are harmful. Universal precautions with regard to infection control are practiced; ear tips, earphones, and electrodes used to deliver and record test signals are disposable.

Acceptability and Simplicity

There are multiple automated OAE and A-ABR devices now available for use in newborn hearing screening programs. The screening devices are simple and easy to use, and the test itself is not time consuming (depending on various factors, taking minutes to administer). Because most of these screening devices provide only a pass-fail outcome, test interpretation that in the past required a professional is now not an issue. No changes can be made in the level of the stimulus or the pass-fail criterion of some devices; only a professional can modify these characteristics in others.

Acceptability of present automated screening tests is not an issue with regard to neonates; neither technology is invasive or traumatic (Bess & Paradise, 1994). The acceptability of the test for screening personnel also is not an issue. The OAE screening test requires the placement of a small probe tip into the external ear canal of the infant. The A-ABR requires the preparation of the skin for the placement of electrodes and the placing of earphones or a plastic ear tip into the external auditory canal. Higher fail rates are encountered when less experienced screening

personnel administer the test, when the screening is conducted in spaces with high noise levels (OAE technology is generally more affected than A-ABR; see, e.g., Headley, Campbell, & Gravel, 2000, but see Norton et al., 2000a, and Gorga et al., 2000, who found no effect of test environment), and when screening is conducted with infants less than 24 hr old (Doyle, Burggraaff, Fujikawa, & Kim, 1997).

Reliability

Both technologies are reliable. In other words, they yield consistent results when they are repeated under exactly the same conditions. Not surprisingly, in a clinical setting where it is impossible to duplicate the exact conditions from one time to another, an infant may fail the screening test on the first attempt and pass it a few hours later. This is almost always due to changes in the circumstances of the test (e.g., how quiet/relaxed the infant is, the placement of the probe, the ambient noise in the room). Whenever statistical probability is being used to make a pass/fail decision, the chance of obtaining a false-negative result (a pass by chance alone) is increased to some degree when a test is repeated over and over again (Benjamini and Hochberg, 1995; Zhang, Chung, & Oldenburg, 2000). However, given the statistical algorithms and cutoff points used in these hearing screening units, the tests would have to be repeated dozens of times before the reliability of the screening test was noticeably affected.

Validity

The purpose of neonatal hearing screening is to identify those infants that are most likely to have PHL so that they can be referred for diagnostic evaluation. The ideal screening procedure would be both highly sensitive (fails infants with PHL) and highly specific (passes infants with normal hearing). However, no screening test is perfect. An individual screening program must select a screening technology whose performance characteristics are known and have been evaluated against a "gold standard" test (e.g., Gorga & Neely, 2003).

Norton et al. (2000a) conducted a large research study to compare three potential hearing screening tests (TEOAE, DPOAE, and ABR) with such a comparison measure. Behavioral audiometry at 8–9 months of age was used as the gold standard test and was completed on infants regardless of their neonatal test results (95% provided reliable tests in 64% of the original study cohort that were evaluated; Widen et al., 2000). The investigators reported that for the specific test conditions, parameters evaluated, and definitions of hearing loss and normal hearing used, the three screening tests had similar performance characteristics (were able to accurately identify infants with PHL as impaired and those with normal hearing as normal; Norton et al., 2000a).

Several large demonstration projects (Dalzell et al., 2000; Finitzo et al., 1998; Prieve & Stevens, 2000; Spivak et al., 2000; Vohr, Carty, Moore, & Letourneau, 1998) have reported on the feasibility and practical aspects of using the

two-step OAE/A-ABR screening protocol. However, these programs evaluated only the hearing of infants who had failed the in-hospital screening. While the Norton et al. (2000a) study evaluated individual technologies, like the Johnson, White, Widen, Gravel, Vohr, et al. (2005) investigation, the protocol was not designed to examine the performance characteristics of the two-step hearing screening procedure per se. However, it is possible to use some data obtained in both studies to address issues of the validity of the OAE/A-ABR protocol in the identification of infants with mild or greater degrees of hearing loss. For example, based on the data obtained in the Johnson, White, Widen, Gravel, James, et al. (2005) and Johnson, White, Widen, Gravel, Vohr, et al. (2005) studies, it was possible to conservatively estimate that the prevalence of hearing loss among infants who passed the two-step OAE/A-ABR screening protocol and were later confirmed with PHL was 0.55/1,000 (see White et al., 2005, for details of the calculation).

Because the second-step A-ABR screening missed some (but not all) cases of PHL in infancy, clinicians might question whether OAE screening alone would be a more sensitive test for identifying all cases of PHL in later childhood. Recall that only those infants who failed OAE and passed A-ABR received audiologic evaluation in infancy (Widen et al., 2005). While it is the case that none of the ears of the study participants who passed OAE screening were found to have hearing loss at 8–9 months of age, test performance characteristics for OAE screening alone for identifying mild and greater degrees of PHL were not part of the study design of the Johnson, White, Widen, Gravel, Vohr, et al. (2005) investigation (White et al., 2005). The performance characteristics of the OAE test would need to be evaluated in a study that followed both OAE pass and fail cases, as was done by the Norton et al. (2000a, 2000b) NIH investigation. Based on the Norton study, there is some evidence that if OAE testing were applied alone, the technology would still miss cases of mild hearing loss (Norton et al., 2000a). The Norton NIH study data (Norton et al., 2000a) would suggest that OAEs are no more sensitive to mild hearing loss than ABR.

It is important to remember in the Norton NIH investigation that for cases of moderate and greater hearing loss, the performance characteristics of ABR and both OAE tests (DPOAE and TEOAE) were essentially the same. However, the problem of detecting mild hearing loss is evident in the data reported from the NIH study presented in Cone-Wesson et al. (2000) for infant ears that were determined to have mild PHL ($n = 22$) in infancy and that had all tests (A-ABR, DPOAE, and TEOAE) in the perinatal period. Ten of those ears failed both OAE and A-ABR tests, 4 ears passed both OAE and A-ABR tests, 4 passed ABR and failed both OAE measures, and 2 failed ABR and passed OAE tests. Finally, 2 ears that failed ABR passed DPOAE but failed TEOAE. These findings from the Norton NIH study (Cone-Wesson et al., 2000; Norton et al., 2000a), as well as those of our current studies (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005), remind us that the

identification of mild hearing loss in the neonatal period is not straightforward and that no screening test performs perfectly.

The validity of a screening test also could be affected by variability among the various screening test parameters used for infant screening. The dB nHL levels used in ABR testing are calibrated and referenced to adult behavioral thresholds for the stimuli (e.g., clicks; Stapells, Picton, & Smith, 1982). The click intensity used in the NIH study was 30 dB nHL (0 dB nHL = 33.8 dB SPL) calibrated similarly across sites. The click level used in the A-ABR testing in the Johnson, White, Widen, Gravel, James, et al. (2005) study was 35 dB nHL, and no attempt was made to calibrate devices to the same SPL referent across sites in that investigation (see White et al., 2005). Importantly, neither differences in the size of the external ear and auditory canal (in the case of supra-aural-type earphones) nor the volume of the closed ear canal (in the case of insert-type earphones) of individual infants are accounted for by the ABR screening instruments used in either the Norton study or the Johnson study (Johnson, White, Widen, Gravel, James, et al., 2005; Sininger et al., 2000; White et al., 2005). Said differently, the actual dB SPL of the stimulus at the eardrum used for screening differs from the adult standard referent, among infants, and indeed among equipment and earphone type (e.g., Sininger, Abdala, & Cone-Wesson, 1997; Stapells, 2000; Stevens & Wood, 2004). This may account for the difficulty in identifying infants with mild degrees of hearing loss as well as the challenge in developing a reasonable screening procedure that would be more sensitive to mild hearing loss than the currently used two-step OAE/A-ABR protocol. (Note that OAE stimulus level is adjusted in the ear canal of the infant, and test outcomes are less likely to be affected by this issue.)

Another consideration affecting the validity of the technologies and therefore the devices currently available for hearing screening is that there are no national standards for the calibration of OAE or ABR instrumentation. Compounding this lack of uniform standards, manufacturers of hearing screening devices may not provide sufficient supporting evidence for professionals that would allow them to determine the validity of the specific pass-fail criteria and/or automated algorithms incorporated in the instruments they purchase. Consequently, there is potential variability of screening results within and between technologies and across manufacturers' devices. Finally, the broad spectrum of the click stimulus used for ABR screening tests could result in a pass outcome in later confirmed cases of high-frequency or unusual configurations of hearing loss (Widen et al., 2000). As suggested previously, all of these circumstances may complicate the development of early identification programs whose goal is to detect milder forms of hearing loss.

Predictive Value

The positive and negative predictive value of a screening test (the probability of having a hearing loss when the infant fails the screening test and the probability of having

normal hearing when the infant passes the screening test, respectively) is affected by the prevalence of the condition within the population. When the prevalence of the condition is low, as is the case with PHL among infants, even a highly specific test will have a low positive predictive value, thereby requiring many infants to be brought back for diagnostic testing to find the few cases of true hearing loss (Bess & Paradise, 1994).

Population-based estimates indicate that milder forms of PHL exist in a substantial proportion of school-age children. Depending on the definition, the prevalence rate for permanent mild bilateral and unilateral hearing loss in school-age children has been estimated at 54/1,000 (5.4%; Bess et al., 1998) and at nearly 71/1,000 (7.1%) in the school-age and adolescent population (6–19 years) in a recent preliminary analysis of the audiologic data from the National Health and Nutrition Examination Survey III (1988–1994) national population-based cross-sectional survey by Ross and colleagues (Ross, Visser, Holstrum, & Kenneson, 2005).

To examine prevalence of mild PHL in the Johnson, White, Widen, Gravel, James, et al. (2005; Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005) birth cohort ($n = 86,634$), we considered the number of infants from this larger group who were enrolled in the study and who failed OAE and passed the A-ABR screening and were confirmed with mild PHL in one or both ears in infancy ($n = 17$ of 21; White et al., 2005). We also considered the number of infants from the larger comparison group identified through neonatal screening (failed both OAE and A-ABR screening) who were referred for diagnostic tests and confirmed as having mild PHL ($n = 31$ of 158; White et al., 2005). Based on the total numbers of children confirmed with mild forms of PHL, we calculated a conservative estimate of the prevalence rate of mild PHL (bilateral mild or mild unilateral hearing loss) in infancy at 0.55/1,000 (0.06%).

If this estimate (0.06%, not adjusting for loss to follow-up) is correct, it appears that our newborn screening efforts may be missing large numbers of children with later permanent mild bilateral or unilateral hearing loss. Consider that Bess et al. (1998) reported a prevalence rate of mild PHL of 5.4% in elementary and middle school-age children (assessed at third, sixth, and ninth grade). Thus, although we may identify some of the infants with mild hearing loss through standard as well as intensive follow-up of infants failing only one screening measure (i.e., those who failed OAE only), it appears that we cannot account for the greater numbers of children with mild bilateral or unilateral hearing loss later in childhood (see Davis et al., 2002, for further discussion). Possible factors for this disparity include that mild hearing loss is missed because neonates pass OAE and/or A-ABR screening but have the following: (a) unusual configurations of hearing loss (Widen et al., 2000); (b) regions of normal hearing sensitivity along with hearing loss in other frequency regions (Widen et al., 2000); (c) late onset hearing loss and normal hearing at birth; (d) hearing loss undetected by standard click- and frequency-specific ABR threshold

measures below the age of 6 months (Widen et al., 2000); (e) mild hearing loss that exhibits normal thresholds during behavioral audiometric assessment in infancy when insert earphones are used (see discussion below); (f) mild hearing loss but the children could not be tested reliably using behavioral measures (the “raised suspicion group”; see White et al., 2005, and Widen et al., 2005); or (g) late-onset hearing loss and the children eventually are diagnosed with more moderate and severe forms of auditory impairment.

Regardless of the reason, this apparent disparity must be considered as clinicians contemplate both the usefulness of screening for mild hearing loss in the neonatal period and the implications of that decision for follow-up. In addition, clinicians, educators, and public health officials need to appreciate this difference in prevalence rates as they develop policies and plan audiologic and educational service provision throughout childhood (Davis et al., 2002). Finally, these data support the development of organized early identification programs before school entry (e.g., Allen, Stuart, Everett, & Elangovan, 2004), if mild bilateral and unilateral hearing losses are considered important educational and public health conditions.

Costs

Costs associated with newborn hearing screening programs are calculated from capital and operating expenses including costs of the screening equipment, personnel, disposables, and follow-up testing including an outpatient rescreen and diagnostic testing to confirm the presence of hearing loss (Gorga, Preissler, Simmons, Walker, & Hoover, 2001). Costs associated with a two-step OAE/A-ABR screening protocol initially are higher, because the combined technologies are more expensive to purchase than either an OAE or A-ABR screener alone (NCHAM, 2005). In addition, in-hospital screening using the OAE/A-ABR protocol is more expensive than OAE screening alone because of the additional disposable costs and additional personnel time required to complete both procedures (Gorga et al., 2001). Costs associated with follow-up and diagnostic testing are also a consideration. A screening program using OAE alone generally has a greater number of children who require follow-up than a screening program that uses A-ABR technology alone or when an OAE/A-ABR protocol is used (e.g., Gorga et al., 2001; Gravel et al., 2000).

However, costs associated with follow-up can be significant because of the resources that need to be expended on bringing back all infants who failed or were missed by the inpatient neonatal screening for testing on an outpatient basis. Thus, keeping inpatient screening referral rates as low as possible is desirable. In addition to cost, lower referral rates at discharge could increase the concern of primary care providers over a fail outcome. That is, the providers may be more likely to see the need and ensure that follow-up testing is completed if very few infants require it.

A second-stage outpatient screening also reduces appreciably the number of infants referred for expensive diagnostic tests (e.g., Prieve et al., 2000). When an outpatient

rescreening occurs, often the same test (OAE or A-ABR) that the infant failed in the neonatal period is repeated. Some programs increase the amount of audiologic data collected at the outpatient visit and include tympanometry, for example, in the screening test protocol. Consider that second-stage outpatient screening offered to neonates who have OAE fail/A-ABR pass outcomes may offer an opportunity to examine the reason for that screening pattern more completely. This follow-up strategy could provide the opportunity to begin the delineation of those infants who continue to have an OAE fail outcome in the presence of normal tympanograms (using a high-frequency 1000-Hz probe tone before 4 months of age; Kei et al., 2003; Margolis, Bass-Ringdahl, Hanks, Holte, & Zapala, 2003); this would suggest sensory involvement, from those who fail an outpatient OAE screening and have abnormal tympanometry, consistent with a conductive component contributing to the OAE screening failure. In these two outcome examples, follow-up would be indicated; however, the type of referral (audiologic vs. medical) would differ, at least initially.

Would costs of a newborn hearing screening program increase significantly if infants with mild PHL (<40 dB HL) were included in the target population for identification in the neonatal period? This can be examined by considering the potential costs of possible approaches to identifying infants with mild hearing loss, for example: (a) using a single-technology (OAE) screening protocol and following up all those who fail OAE screening with diagnostic testing, (b) lowering the intensity of the second-step A-ABR level to 25 dB nHL from the current 35 or 40 dB nHL, or (c) bringing all children who pass a two-step screening protocol (fail OAE/pass A-ABR) back for behavioral testing before the end of the 1st year as was done in the Johnson, White, Widen, Gravel, Vohr, et al. (2005) study (White et al., 2005). Such cost-benefit studies are critical before changes in approaches to early detection are made; to our knowledge, such analyses of these alternative approaches have not been completed to date.

Practicability

The concerns raised by Bess and Paradise (1994) regarding the achievability of UNHS have essentially been alleviated by the results of newborn hearing screening programs in the United States as well as around the world. The establishment of approximately 2,500 UNHS programs here in the United States (NCHAM, 2005), as well as the development of quality provincial and national UNHS programs in other parts of the world, supports the practicability of screening infants for hearing loss before discharge from the hospital.

Another practical concern raised by Bess and Paradise (1994) was the potential high referral rate they thought possible from hospitals with 24-hr nursery discharge policies, specifically those that used OAE screening technology only in the neonatal period. Indeed, as discussed previously, this is a concern (e.g., Doyle, Burggraaff, Fujikawa, & Kim, 1997), and reports (NCHAM, 2005) from programs using current OAE screening devices

suggest that average fail rates at discharge can range from 3% to 15%. However, when a second-stage screening using the same technology before referral for comprehensive audiologic assessment is included in the protocol, referral rates can be decreased to 0.5%–1% (NCHAM, 2005).

The final concern raised by Bess and Paradise (1994) was regarding the availability of “qualified audiological professionals and sophisticated audiometric equipment [for the] second-stage screening and follow-up evaluation of infants” (p. 332). Indeed, these concerns are among the most important issues facing the provision of quality EHDI programs in the United States today (White, 2003). Bess and Paradise also were concerned about access to quality audiologic services for those “estimated 25% of births ... that occur in rural or remote areas” (p. 332). There is also evidence that even when availability and proximity of audiologic follow-up services are not a concern, socioeconomic circumstances and/or geographic location may reduce the number of infants returning for follow-up diagnostic tests (Prieve et al., 2000). We address these important matters, specifically with regard to the identification and follow-up of infants and young children with permanent bilateral mild and unilateral hearing loss, in the section below titled “Availability and Accessibility.”

Treatment of Mild Hearing Loss in Infants

This section examines questions surrounding the efficacy, availability, accessibility, and compliance with treatment (early intervention) following confirmation of mild hearing loss and whether early intervention of children with mild hearing loss is more effective than later treatment.

Efficacy

Unlike the extant data available at the time of the Bess and Paradise (1994) report, today there is evidence that early intervention for infants with hearing loss is efficacious (Moeller, 2000; Yoshinaga-Itano et al., 1998). In addition, there are no reports of harm associated with early intervention efforts per se. There appear to be no published data that have specifically addressed the efficacy of early intervention for children with mild bilateral hearing loss or with unilateral hearing loss. Studies examining the long-term benefit and timing of enrollment in a school- or home-based language stimulation program are needed for appropriate management to occur. For example, evidence is needed on the benefit of interventions such as the fitting of personal hearing aids (in one or both ears) and/or the use of a sound-field FM amplification system by young children with bilateral mild or unilateral PHLs. Currently in many states, infants with PHL are automatically eligible for early intervention services before evidence of delay (in communication development) is apparent. As in all aspects of pediatric audiology, an evidence base for practice is critical in order that limited public health and educational resources are allocated most effectively (Gravel, 2005).

Availability and Accessibility

Bess and Paradise (1994) cautioned that there must be a “certainty that facilities for suitable follow-up care of individuals who fail the screen are both available and accessible” (p. 332). This tenet of public health screening applies to diagnostic testing as well as to intervention including amplification fitting and early habilitation programs. Certainly, availability and accessibility, as well as the quality of pediatric audiologic services, are still concerns today. Indeed, the shortage of qualified and experienced pediatric audiologists has been cited as a major problem by state EHDI coordinators in annual surveys conducted by NCHAM (White, 2003). The availability and quality of audiologic testing facilities with the requisite equipment and personnel necessary to diagnosis any type and degree of hearing loss (American Speech-Language-Hearing Association, 2004) are particularly relevant for the accurate and timely confirmation of mild forms of hearing loss (see Widen et al., 2005). “Audiological uncertainty” has been cited as the reason for delays in confirmation and amplification fitting in cases of mild forms of hearing loss (Dalzell et al., 2000), and it is clear, based on national surveys, that infants with mild and moderate hearing loss have later ages of confirmation and hearing aid fitting than those with severe to profound hearing loss (Harrison & Roush, 1996), regardless of their access to newborn hearing screening (Harrison, Roush, & Wallace, 2003). Thus, evidence suggests that the concern of Bess and Paradise (1994) remains, as centers and personnel who can complete comprehensive audiologic assessment of young children are still not readily available or accessible (White, 2003).

These concerns are particularly important when we consider what facilities and “best practices” would be needed to accurately distinguish between mild hearing loss and normal hearing, as well as between mild and greater degrees of hearing loss in infants. We spent considerable time specifying what combination of audiologic test characteristics (including tympanometry, OAE, behavioral audiometry, and evoked potential test results) would be considered sufficient to deem an ear as having mild PHL (Widen et al., 2005). In addition, in many cases repeated visits were needed to confirm the presence, degree, configuration, and type of hearing loss. Infants in the present study (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005) were assessed at a developmental age of 8–9 months (Widen et al., 2005), an age at which behavioral testing is generally considered reliable (Widen, 1993), as well as at a time within the 1st year of life, a period that has been shown to be important for the initiation of early intervention (Moeller, 2000). Whether all cases (that is, degrees and configurations) of mild hearing loss could be adequately delineated using diagnostic electrophysiological measures alone (e.g., frequency-specific air- and bone-conduction ABR) to estimate behavioral thresholds before conditioned response audiometry is possible (<6 months) is not clear. To adequately delineate mild hearing loss using electrophysiological or behavioral measures, when test stimuli are delivered using insert earphones, a measured or derived estimate of the SPL at the

plane of the tympanic membrane should be available. This is necessary because of the difference in SPL associated with the dial-setting value (in dB HL or dB nHL) on audiometric equipment that has been calibrated to an adult reference standard versus the actual SPL in the infant ear canal (see, e.g., Seewald & Scollie, 1999). In the case of confirming hearing status, a 10-dB underestimate or overestimate of hearing sensitivity can make a significant difference in whether an infant is considered to have normal hearing or mild hearing loss and either cleared from further follow-up or referred for early intervention and potentially even early amplification provision. Consider that the consequences of a 10-dB over- or underestimation of threshold sensitivity may not be as significant for children with moderate or greater degrees of PHL. Clearly, the ready availability and the validity of diagnostic audiologic assessment methods needed to confirm the presence of normal hearing from mild degrees of hearing loss should factor into our decisions regarding what is the best age to initiate early identification efforts.

Compliance

While UNHS has been implemented for the majority of birthing hospitals across the United States, tracking and follow-up continue to be major challenges (White, 2003). Indeed, it remains difficult in many places for facilities and states to track those infants who failed screening in the newborn period and return them for follow-up regardless of the protocol being used. Evidence suggests that in some localities, despite concerted efforts at bringing infants back for rescreening or diagnostic audiologic evaluation, as many as 50% of infants may be lost to follow-up (e.g., Prieve et al., 2000). Consider that in our study, only 64% of families whose infants had failed OAE and passed A-ABR screening returned for comprehensive audiologic testing at 8–9 months of age, even though the reason for the study (that is, the potential for hearing loss despite the hearing screening pass outcome) had been explained to them (White et al., 2005).

Would identifying infants at risk for late onset and progressive hearing loss (Joint Committee on Infant Hearing, 2000) detect infants who had mild hearing losses in later childhood? Although the losses confirmed at 8–9 months of age could have been adventitious, the results of the Johnson, White, Widen, Gravel, Vohr, et al. study (2005) indicate that only 8 of 21 infants with confirmed PHL in infancy had risk factors for late onset or progressive hearing loss (White et al., 2005; Widen et al., 2005).

The efforts of the American Academy of Pediatrics (AAP) in educating physicians about the importance of early identification of hearing loss as part of ongoing surveillance in the child’s “medical home” (AAP, 2002) may represent an alternative to expending resources directly on diagnostic audiologic follow-up of large numbers of infants. The limitation of this approach is that it is not until parents or the physician become concerned about communication development that a referral for audiologic assessment is made. Speech and language delays may not be as readily apparent in cases of mild bilateral or unilateral

hearing loss as they are in children who have more severe hearing deficits. Moreover, a convenient, objective screening tool (such as an OAE screening device) that could be used in routine office-based screening currently is not widely available in primary care practice settings. Thus, while monitoring hearing status in the child's medical home is a desired goal, the approach is only one of several that may be useful in identification of infants with mild bilateral and unilateral hearing loss.

Early Versus Late Treatment

At the current time, there is limited evidence about whether early intervention leads to better outcomes for infants with confirmed mild hearing loss. This includes the efficacy of various forms of amplification devices including hearing aids and FM systems for use with this population in early life. Questions remain as to whether technology-based or non-hearing-instrument-supported early interventions are most useful for those children with mild forms of PHL who are experiencing communication delays or whether the devices should be used as preventative measures. If, when, and how to intervene with infants diagnosed with mild forms of hearing loss are important questions that need to be addressed as states consider the expenditure of resources in early intervention programs.

It may be that identification efforts for mild hearing loss should be directed at those children already receiving early intervention—for example, direct objective screening of pre-school-age children enrolled in early education programs—whose eligibility for early intervention services was based on significant speech and language delay or on other developmental risk factors such as living in poverty (e.g., Head Start programs; Allen et al., 2004). Children with mild forms of hearing loss and with one or more intrinsic or extrinsic risk factors are likely to be more vulnerable to the development of communicative, social-emotional, and later academic sequelae because of co-occurrence of such variables with an auditory impairment.

Research Recommendations

Based on the Johnson, White, Widen, Gravel, James, et al. (2005; Johnson, White, Widen, Gravel, Vohr, et al., 2005) investigation and the other articles in this series (White et al., 2005; Widen et al., 2005), there are a number of important research questions that require study and issues that need to be addressed before a change in current screening practice or use of a specific protocol is warranted. However, we have the following recommendations:

- Implement a large, multisite prospective study of the development of infants and toddlers identified early in life with mild forms of PHL.
- Measure SPL in the ear canal of newborns using various test signal delivery methods, in particular insert and supra-aural earphones, to determine the range of

variability associated with test signals used in A-ABR screening devices.

- Require that all hearing screening studies clearly and accurately describe the stimuli (e.g., level), as well as the instrumentation and methods used to determine those acoustic characteristics.
- Determine the pass-referral rates that result from using different OAE (TEOAE vs. DPOAE) and A-ABR equipment for identifying mild forms of hearing loss.
- Examine the usefulness of adding a screening for middle ear function at the time of the newborn screening and, at the second stage, outpatient rescreen for identifying cases of OAE fails that are more likely to be associated with middle ear/external ear canal debris. Study the usefulness of the measure to increase the sensitivity of screening for detection of mild hearing loss and for reduction of the costs associated with follow-up.
- Determine what percentage of infants with confirmed mild hearing loss in infancy had hearing loss in the newborn period through immediate diagnostic assessment (i.e., determine the true false-negative rate).
- Develop a more cost-efficient and effective screening protocol that modifies one or more existing screening technologies. For example, determine the effectiveness and costs associated with protocols that (a) lower the A-ABR screening level to 25 dB nHL, (b) incorporate a two-level OAE screening test to detect cases of mild hearing loss (Hall, 2005; Hall & Lutman, 1999; Widen & O'Grady, 2002), or (c) include a screen of middle ear function (Keefe et al., 2000; Keefe, Gorga, Neely, Zhao, & Vohr, 2003; Keefe, Zhao, Neely, Gorga, & Vohr, 2003; Kei et al., 2003; Margolis et al., 2003) in addition to OAE and A-ABR screening in the neonatal period.

Conclusions

The results of the series of studies in this issue (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005) are enlightening and thought-provoking. Collectively, the series reminds us that no screening program is perfect and that the detection and confirmation of mild forms of PHL are not straightforward. The frequency with which the OAE/A-ABR newborn hearing screening protocol is used, coupled with the successful outcomes of infants and young children with hearing loss who are identified in newborn hearing screening programs, has led many people to believe that such screening programs are identifying all newborns with PHL. However, based on this series of reports, it is clear that a substantial number of infants with PHL will pass a commonly used OAE/A-ABR hearing screening protocol. These findings remind us of the need to continually monitor the results of any screening program and to look for ways to improve on current "best practices." The results also emphasize the importance of ensuring that the equipment and protocols used in a screening program are consistent with the goals of that program. It is not surprising that a protocol

based on equipment that screens for hearing loss using a 35-dB nHL click, for example, will miss a significant number of infants with hearing losses ≤ 30 dB. The results of this series of reports are also thought-provoking because of the directions suggested for further refinement and improvement in how we identify PHL among infants and young children and confirm its presence as early as possible. Providing answers to the research questions outlined above is important and deserves our sustained attention and resources.

The studies presented here provide important information about one piece of the newborn hearing screening puzzle that was missing; that is, an estimate of the proportion of infants with mild PHL missed by a widely used two-step screening protocol. However, there is much more information needed in order that clinicians may continue to improve newborn hearing screening programs and early identification efforts in childhood, in general. Although all of the research issues outlined above are important, one area that has received relatively little attention is how to supplement hospital-based newborn hearing screening programs with continued opportunities for screening in other settings where infants and young children are frequently present and thus accessible (e.g., health care providers, early childhood programs). Given the findings of the series of studies presented here (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005), early identification efforts that continue beyond the newborn period targeted at identifying young children with PHL who were not identified by newborn hearing screening programs would be desirable.

By considering the results of this study in light of the tenets of good public health programs suggested by Bess and Paradise in 1994, it is clear that there are not definitive data to suggest that the two-step OAE/A-ABR newborn hearing screening protocol should be avoided. In those situations where it is important to minimize referral rates at the time of hospital discharge or where identification of mild hearing loss is not a priority, a two-step screening protocol using currently available screening equipment may well be the best approach. In cases where identification of mild hearing loss is a priority, current OAE/A-ABR hearing screening protocols may not be the best option. As suggested previously, modifications of the existing two-step protocol, such as lowering the A-ABR stimulus from 35–40 dB nHL to 25 dB nHL, may improve the appropriateness of this protocol for identifying cases of mild hearing loss. However, there are trade-offs when any test (such as OAE and A-ABR) that does not perform perfectly (Norton et al., 2000a, 2000b) is used in screening. For example, a change in the stimulus level of the A-ABR may identify a significant number of additional cases of mild hearing loss (that is, increase the hit rate) over the current two-step protocol, but the change would almost certainly increase the false-positive rate. The costs associated with the follow-up of an increased number of infants who fail screening but who have normal hearing may divert resources now allocated toward EHDI programs targeting infants with congenital and neonatal onset hearing loss of moderate degree and greater who are known to benefit from

early intervention (Moeller, 2000; Yoshinaga-Itano et al., 1998). These factors would have to be considered, and studies of the efficiency and effectiveness of any such change of procedures would be necessary, before any decision regarding the optimal hearing screening protocol for use in the newborn period could be made.

One of the most important conclusions from this study is that there are many different factors that contribute to a decision about which protocol and equipment are best for a particular hearing screening program designed to identify infants and young children at risk for hearing loss. Clearly, the advent of new technology and screening devices, including those that examine middle ear function in addition to sensory and neural integrity, will continue to allow for new and different opportunities for detection of hearing loss across childhood. Unfortunately, little is known about the many factors that will affect the development of optimal screening protocols across the pediatric age range. However, it is clear that the evidence obtained from studies of hearing screening protocols similar to those reported here (Johnson, White, Widen, Gravel, Vohr, et al., 2005; White et al., 2005; Widen et al., 2005) will be needed to address many of the issues raised in this report, as well as other important matters related to hearing loss in children.

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