Screening

Karl R. White, Ph.D.,[1] and Karen Muñoz, Ed.D.[2]

ABSTRACT

Most hearing screening programs have historically targeted children with moderate or more severe bilateral hearing loss. Children with unilateral or mild bilateral permanent hearing loss represent a substantial proportion of all children with hearing loss, and there are serious negative consequences for these children if they are not identified early and given appropriate help. Many children, particularly those with unilateral or mild bilateral hearing loss, acquire hearing loss after the newborn period. Although virtually all newborns are now screened for hearing loss before leaving the hospital, there are very few opportunities for periodic hearing screening after the newborn period. Effectively identifying those children who have late-onset loss or who are missed during newborn hearing screening will require modifying some of the procedures currently employed in hospital-based newborn hearing screening programs, as well as establishing better hearing screening procedures for early childhood and elementary school programs. Existing state Early Hearing Detection and Intervention systems are a resource for establishing and improving screening programs for infants and children with unilateral or mild bilateral hearing loss.

KEYWORDS: Unilateral hearing loss, mild bilateral hearing loss, screening

Learning Outcomes: As a result of this activity, the participant will be able to describe how hearing screening programs during the newborn, early childhood, and school-age periods can be improved to better identify children with unilateral and mild bilateral hearing loss.

The first 36 months of a child's life are a critical period for language learning. Research has shown that undetected hearing loss leads to delays in speech, language, cognitive, and social-emotional development, negatively affecting academic performance and potential.[1,2]

[1]Professor of Psychology, Director, National Center for Hearing Assessment and Management, Utah State University, Logan, Utah; [2]Assistant Professor of Communicative Disorders and Deaf Education, Deputy Director, National Center for Hearing Assessment and Management, Utah State University, Logan, Utah.

Address for correspondence and reprint requests: Karl R. White, Ph.D., Director, National Center for Hearing Assessment and Management, Utah State University, 2880 Old Main Hill, Logan, UT 84322 (e-mail: Karl.White@usu.edu).

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Conversely, children who are identified early and begin intervention shortly thereafter have significantly better language development than do those children identified later.[3–5] Most of the attention in the past was focused on children with hearing losses in the better ear of 40 dB hearing level (HL) or worse.[6,7] Recently, however, there has been increasing concern about the consequences of not identifying children with unilateral hearing loss (UHL) and mild bilateral hearing loss (MBHL). Unfortunately, these children are an often overlooked population within the larger population of children with hearing loss even though they are at increased risk for academic grade failures compared with children with normal hearing and often need supplemental educational assistance.[8–10]

All states have established Early Hearing Detection and Intervention (EHDI) programs with a goal of screening all newborns and identifying all infants and young children with permanent hearing loss to provide them with timely and appropriate early intervention services.[11] As a result of these initiatives, hearing screening in infancy has become the medical-legal standard of care in the United States.[12] Indeed, more than 95% of babies are currently screened for hearing prior to hospital discharge compared with less than 20% in 1997.[13]

States and territories have implemented EHDI programs to ensure all children receive hearing screening, diagnostic audiologic evaluations to confirm a loss when indicated by a positive screen, and early intervention services when needed. To help ensure children receive the above-mentioned services, many EHDI programs are developing tracking and surveillance systems that include information about hearing screening results and subsequent follow-up testing. In addition, the information in these systems is used by a majority of states and territories to provide aggregate data that helps calculate national level EHDI statistics.[13,14]

As discussed at the 2005 National Workshop on Mild and Unilateral Hearing Loss, although great strides have been made in screening for hearing in newborns, several challenges exist related to ensuring that all infants and young children with hearing loss receive the services they need to make appropriate developmental progress. This also was noted recently in a letter sent to all state early intervention programs from officials at the U.S. Department of Education and U.S. Department of Health and Human Services,[15] which stated that there is a "growing national crisis in the provision of essential early intervention and health care services for infants and toddlers with hearing loss." That letter continued:

> Studies have demonstrated that when hearing loss of any degree, including mild bilateral or unilateral hearing loss, is not adequately diagnosed and addressed, the hearing loss can adversely affect the speech, language, academic, emotional, and psychosocial development of young children. Although efforts to identify and evaluate hearing loss in young children have improved, there is still anecdotal evidence to suggest that many young children with hearing loss may not be receiving the early intervention or other services they need in a timely manner that will enable them to enter preschool and school ready to succeed.

It is noteworthy that this letter highlighted the importance of "mild bilateral or unilateral hearing loss" because until the 1990s, most efforts to identify hearing loss among infants and young children were limited to identifying children with bilateral losses of 40 dB HL or greater in the better ear.[6,7] Given the growing consensus that children with milder forms of permanent hearing loss also need to be identified and treated as early as possible, it is of concern that infants and young children with less severe degrees of hearing loss are not being identified at a rate anticipated based on documented incidence.[9,16–21] For example, reports in the literature suggest that the incidence among newborns for UHL ranges from 0.8 to 2.7 per 1000 and from 0.4 to 1.3 per 1000 for MBHL.[9,16–20] However, state EHDI programs are reporting the identification of significantly fewer infants and young children with UHL and MBHL.[14]

The incidence of UHL and MBHL appears to increase significantly as children age. Prevalence estimates in school-aged children (~6 through 19 years) range from 30 to 56 per

1000 for UHL and 10 to 15 per 1000 for MBHL[9,18] (see the article by Ross et al,[22] this issue, for a more in-depth discussion). This represents a significant difference in incidence between the newborn period and school age. The reasons for this difference are unclear and are most likely due to a combination of factors, including children with late-onset hearing loss. Causes of late-onset hearing loss include genetic and teratogenic factors as well as acquired environmental factors such as infections or illnesses, trauma, and noise-induced hearing loss.[23]

Participants at the 2005 National Workshop on Mild and Unilateral Hearing Loss pointed out the identification of more children with UHL and MBHL loss at an earlier age will require increased attention to how screening is done during the newborn period, the preschool years, and during the early elementary school years. Issues related to screening during each of these time periods are discussed in the sections that follow.

SCREENING FOR UHL AND MBHL DURING THE NEWBORN PERIOD

During the newborn period, there are many factors that can affect identification of less severe degrees of hearing loss. One factor is the limitation of current technology and protocols used for screening. Protocol selection should involve consideration of the type and degree of hearing loss targeted for identification, the population to be screened, the geographic location of the program, the likelihood that families will complete recommended outpatient testing, and the availability of trained and experienced pediatric audiologists to do complete diagnostic evaluations.

If these factors are carefully considered, screening program administrators can make decisions that will best fit the specific needs of their population. Because these needs vary from site to site, screening protocols are often quite different among hospitals. Most programs and policy statements have historically focused on detecting either bilateral and/or more severe forms of hearing loss.[6,7,23] An important factor that is frequently overlooked is the inability of the chosen protocol to screen for milder degrees of hearing loss. The focus on more severe hearing loss coupled with a desire to minimize the number of false-positive results[7] also has led to most equipment manufacturers setting presentation levels for automated auditory brain-stem response screening equipment at 35 or 40 dB normalized hearing level (nHL).[24] As a result, an infant with a mild degree of hearing loss may be missed if hearing levels are better than the minimum level that can be detected by the screening equipment being used.

In an effort to produce a low refer rate prior to hospital discharge, many hospitals use a two-stage protocol. In this protocol, all infants are screened first with otoacoustic emissions (OAEs), and no additional testing is done for those passing the OAE screening. Infants failing the OAE proceed to the second stage where they are screened with automated auditory brain-stem response (A-ABR). A recent multicenter study by Johnson et al[17] evaluated how many infants who failed the OAE and passed the A-ABR had permanent hearing loss at ~9 months of age. From a birth cohort of 86,634 infants who were screened at seven geographically dispersed birthing centers using a two-stage OAE/A-ABR hearing screening protocol, 1524 infants who failed the OAE but passed the A-ABR were enrolled in the study. Diagnostic audiologic evaluations were completed for 64% of the enrolled infants (1432 ears from 973 infants) when they were an average of 9.3 months of age. The study found 21 infants (30 ears) who passed the newborn A-ABR hearing screening had permanent hearing loss when they were 8 to 12 months of age.

When the results for those infants who failed the OAE but passed the A-ABR screening were combined with those of the infants who failed the OAE and failed the A-ABR, it was determined that the incidence of permanent hearing loss in this cohort of 86,634 newborns was 2.37 per 1000 (this incidence is a little lower than what would be expected in the general population because only one of the seven participating centers enrolled children from the neonatal intensive care unit). Alarmingly, 23% of the infants with permanent hearing loss in this cohort would have been

missed if babies who failed the OAE but passed the A-ABR were screened as normal and had not been followed. Most (71.4%) of the infants with hearing loss who failed the OAE but passed the A-ABR screening test had mild hearing loss. Of those with hearing loss who failed both OAE and A-ABR, 19.6% had mild hearing loss (20 to 40 dB as measured in the worse ear).

The results of this study have important implications for newborn hearing screening programs, particularly with respect to identifying infants with MBHL. First, these results emphasize the need for administrators of newborn hearing screening programs to carefully evaluate which screening protocol and equipment is best for their situation and objectives. In particular, they should explicitly consider whether they want to detect MBHL. In making such decisions, it is important to remember that this is not an issue of whether or not to use A-ABR hearing screening equipment. Instead, it is an issue of how the stimulus presentation for that equipment is set. If a different intensity stimulus had been used (e.g., a 25 dB nHL click stimulus instead of the 35 dB nHL stimulus that was used in this study), the results would almost certainly have been quite different. All A-ABR equipment that is commercially available at the present time uses a 35 dB nHL or greater click stimulus.

Second, parents and health care providers need to be reminded frequently that passing a newborn hearing screening test does not guarantee that the child does not and will not have a permanent hearing loss. This was emphasized more than 20 years ago by Mason et al[25]: "Passing a neonatal screening test, therefore, does not exclude the possibility of subsequent [permanent hearing loss] and highlights the need for further surveillance."

Third, as will be discussed in the next section of this article, hospital-based newborn hearing screenings are not sufficient to detect all permanent hearing loss that occurs during childhood. In addition to making hospital-based screening programs as efficient as possible, public health officials should consider the pros and cons of doing systematic hearing screening during the early childhood years in day care, preschool programs, or well-child visits in health care provider offices.[26] Such screening may be a useful tool for detecting late-onset permanent hearing loss, as well as hearing loss that is missed during newborn hearing screening (which is particularly likely with UHL and MBHL).

In addition to the issues highlighted by the study conducted by Johnson et al,[17] there are several other issues that need to be considered with regard to hospital-based screening. For example, there is an unfortunate lack of national or international standards for the calibration of otoacoustic emission screening devices. Only one international standard (IEC 60645–3[27]) is available for the calibration of clicks (such as those used in A-ABR testing). It is not clear, however, that manufacturers of screening devices used in the newborn period calibrate their instruments using a uniform standard. In addition, screening algorithms and pass-refer criteria vary among manufacturers. Therefore, the screening outcome obtained with one screening unit may not yield the same outcome (pass-refer) as a device from another manufacturer.[28] Related to this issue, otoacoustic emission technology presents stimuli at a known sound pressure level "adjusted" in the ear canal of the individual infant/child. However, the level of the A-ABR click stimulus at the plane of the tympanic membrane varies to some degree depending on the earphone type (i.e., circumaural or insert) and the size of the infant's ear canal. Thus, the actual level of the test signal may vary appreciably among infants regardless of the audiometric hearing level in dB nHL referenced as the pass-fail criterion level by the manufacturer.[29,30] This is particularly concerning because the stimulus will frequently be higher, with the potential result of infants with UHL or MBHL being missed.

SCREENING FOR UHL AND MBHL DURING EARLY CHILDHOOD

It is estimated that the incidence of permanent hearing loss more than triples from 3 per 1000 to 3 per 300 by the time children are 5 years of age.[31] For these children to be identified in a timely manner, systematic hearing screening at periodic intervals will be necessary because

there is evidence that parents are not good at detecting when their child has a hearing loss.[32,33] As noted by the American Academy of Pediatrics (AAP), "Reliance on physician observation and/or parental recognition has not been successful in the past in detecting significant hearing loss in the first year of life."[7] The inability of parents or health care providers to detect hearing loss is particularly true for UHL and MBHL.

Unfortunately, after the newborn period, very few newborn to 5-year-old children are regularly screened for hearing loss using objective screening tools. Indeed, the Pediatric Periodicity Schedule[34] of the AAP calls only for subjective screening of hearing during well-child visits from birth until 4 years of age, and the 2007 Joint Committee on Infant Hearing (JCIH) Position Statement[23] recommends only that there be "regular surveillance of developmental milestones, auditory skills, parental concerns, and middle-ear status. . .in the medical home, consistent with the American Academy of Pediatrics pediatric periodicity schedule." In a positive step toward the goal of systematic hearing screening during the early childhood years, the 2007 JCIH statement does call for "an objective standardized screening of global development with a validated assessment tool at 9, 18, and 24 to 30 months of age or at any time if the health care professional or family has concern. . .Infants who do not pass the speech-language portion of a medical home global screening or for whom there is a concern regarding hearing or language should be referred for speech-language evaluation and hearing assessment."

However, implementing such office-based screening will not be easy as shown by how difficult it has been to get health care providers to do a quick, subjective hearing screen in a context where they have a high likelihood of being reimbursed. Specifically, more than 35% of all newborn to 5-year-old children in the United States are covered by Medicaid. Since its inception, Medicaid has mandated that all eligible children receive early preventive health care through the Early and Periodic Screening Diagnosis and Treatment (EPSDT) program, but this has never been achieved. A national review of EPSDT revealed that of the 22.9 million children eligible for these services, only 36% received a medical screen and only 13% received a hearing screen.[35] The report also concluded that the poorest children and those from minority families (which are the children most likely to acquire hearing loss during early childhood) were disproportionately less likely to receive these services.

Another problem has been identified when children are screened for hearing in their health care provider's office. In one recent study,[36] nine pediatric practices were provided with equipment and staff to do hearing screening during well-child visits for those 3 to 19 years of age. Of the 1061 children screened, 10% failed. Of these, 59% had no further evaluation indicating how difficult it is to get health care providers and families to follow up when a child fails the hearing screening test.

Despite such challenges, there have been successful programs designed to screen hearing loss during the early childhood years. Eiserman et al[26] evaluated the feasibility of performing hearing screening in Migrant, American Indian, and Early Head Start programs using objective OAE technology. Staff at the Head Start programs were trained to screen newborn to 3-year-old children for hearing loss using hand-held OAE equipment and a multistep screening and referral protocol. Of the 3486 children screened, 95% passed, and 5% (183 children) were referred for a diagnostic evaluation. The median time required to complete a single OAE screening session was 4 minutes per child. Of the 119 children who completed a diagnostic evaluation (64 children, or 35%, were lost to follow-up), 6 had a permanent hearing loss, and 74 more had a chronic fluctuating conductive hearing loss that had not previously been detected or treated. These results demonstrate that OAE-based hearing screening of young children can be practical and effective if there is appropriate training, use of the specified protocol, and audiologic support.

Prior to the evolution of OAE technology, early childhood educators and health care providers had no choice but to rely on informal behavioral observations, checklists, and questionnaires to screen for hearing loss. Fortunately, OAE screening technology has proved

to be a reliable, easy-to-use screening tool that can be effective in identifying newborn to 5-year-old children with permanent hearing loss. Though not used much in health care providers' offices at the current time, the success of OAE-based hearing screening in Early Head Start programs suggests that this might be an effective tool for health care providers to use during well-child visits.

SCREENING FOR UHL AND MHL AMONG ELEMENTARY SCHOOL-AGED CHILDREN

The number of children affected with hearing loss continues to increase as children grow older. An excellent source of data about this is the National Health & Nutrition Examination Survey (NHANES). The NHANES surveys are broad, multipurpose surveys conducted with a target population of civilian, noninstitutionalized members of the U.S. population. The sample size for audiometry in children, 6 to 19 years old, was 6166 for the NHANES study conducted from 1988 to 1994. According to the NHANES data, the prevalence of hearing loss for 6- to 19-year-old children in the United States is 73.31 per 1000.[37] Ninety-six percent of these children had UHL (77.8%) or MBHL (18.8%).* Unfortunately, given the way the data were collected, it was not possible to distinguish between permanent hearing losses and those caused by fluctuating conditions such as otitis media with effusion.

Given the prevalence of hearing loss among school-aged children (almost 25 times as many school-aged children have hearing loss as do newborns), it is not surprising that most states have legislative mandates requiring school screening.[38] But even though school-age hearing screening has been occurring for decades, there has been no systematic national evaluation of these programs, and little is known about their effectiveness.[39] It is likely that the situation in the United States is very similar to what was recently reported from a national evaluation of the school-entry hearing screening program in England.[40,41]

> Referral rates are variable. . .The test used for the screen is the pure tone sweep test but with wide variation in implementation, with differing frequencies, pass criteria and retest protocols; written examples of protocols were often poor and ambiguous. There is no national approach to data collection, audit and quality assurance, and there are variable approaches at the local level. The screen is performed in less than ideal test conditions and resources are often limited, which has an impact on the quality of the screen.

The dramatically higher prevalence of hearing loss among school-aged children (most of whom have UHL and MBHL) and the documented deleterious effects of undetected hearing loss, coupled with the difficulty of detecting UHL and MBHL, raises concerns about why there has not been a more systematic approach to school-based hearing screening. One of the reasons that EHDI programs have been relatively successful with the newborn population is that there have been widely accepted standards and protocols promulgated by groups such as the AAP, the JCIH, and the Centers for Disease Control and Prevention (CDC).[42] Nothing similar has occurred for hearing screening for school-aged children.

Of course, there are challenges that make it difficult to implement effective hearing screening programs in a school setting—particularly if the goal is to detect children with UHL and MBHL. Ninety-six percent of children enroll in the public school system at 5 to 6 years of age.[43] Currently, this is the only opportunity for an additional universal hearing screening beyond the newborn period, although the acoustic environments in schools for the entry screen and any subsequent hearing screenings are often inadequate to conduct appropriate and accurate testing.[9] Behavioral audiometric and tympanic screening tests applied at school-age are intended to identify temporary as well as permanent hearing loss, including UHL and MBHL. Yet the acoustic conditions under which school-based identification programs

*NHANES analysis uses definitions of ''slight'' (16 to 25 dB HL) and ''mild'' (26 to 40 dB HL) for low-frequency hearing loss (pure-tone average 0.5, 1, and 2 kHz) and high-frequency hearing loss (pure-tone average 3, 4, and 6 kHz).

are administered as well as pass-fail criteria are not always conducive to achieving that goal. Unless controlled acoustic conditions that reduce the levels of background noise (e.g., a portable sound-treated test booth) are available, cases of UHL and MBHL will likely be underidentified.

DIAGNOSING UHL AND MBHL

Another factor affecting the identification of hearing loss at all ages is the ability to successfully link the family to appropriate outpatient services with a well-trained pediatric audiologist. During the newborn period, state EHDI programs often have difficulty determining if an appropriate diagnostic assessment has been completed because many audiologists do not consistently report their findings to the state EHDI program, even though many are required to do so by their state mandate.[44] According to national statistics from the CDC,[14] almost 60% of infants who are referred from newborn hearing screening programs have no documented follow-up. However, it is difficult to determine how many of these infants never received needed follow-up testing compared with how many completed diagnostic testing, but the findings were not reported to the state EHDI program. State EHDI programs also have consistently reported a severe shortage of well-trained pediatric audiologists. This means that families often have difficulty in identifying where to go for recommended services.[42] There is currently no national effort to track older children, and therefore there is no information about audiologic follow-up among these children.

Audiologists who are not familiar or comfortable with including frequency-specific auditory brain-stem response (ABR) stimuli (tone bursts of low, mid, and high frequency[†]), bone conduction ABR,[‡] high-frequency acoustic immittance assessment, and determining hearing sensitivity for each ear as part of recommended protocols[45] may inadvertently miss UHL and MBHL. Too often, the presence of middle ear effusion has delayed the diagnosis of permanent hearing loss. Audiologists who postpone diagnostic testing until the middle ear effusion has cleared risk delaying diagnosis by many months. Delaying the diagnostic assessment puts the child at risk of being lost, not diagnosed in a timely manner, and subsequently not referred for appropriate follow-up services (see the article by Gabbard et al,[46] this issue, for a more in-depth discussion).

CONCLUSION

Now that newborn hearing screening is a standard of care, there is an opportunity to create a seamless system of care for infants and young children with hearing loss from birth through school age. For this to happen, greater attention must be given to ensuring that all children with congenital hearing loss (including those with UHL and MBHL) are identified and provided with appropriate services during the first months of life. Additionally, the lessons learned about implementing effective newborn hearing screening programs must be adapted and applied to screening children for hearing loss in early childhood and elementary school settings. For example, protocols and procedures for successful screening programs need to be developed and disseminated, more attention needs to be given to training, quality assurance, and documentation of program activities and outcomes, and national goals and standards need to be established.

Many state EHDI programs have developed an infrastructure for supporting hospital-based newborn hearing screening programs, providing quality assurance, training, and technical support, and collecting data to ensure that children receive the services they need and that the system can be continually evaluated and improved. These EHDI systems could be expanded to assist in doing hearing screening during early childhood and elementary school.

Identification of consistent referral and follow-up systems for children with less severe degrees of hearing loss is needed. Research has shown that a substantial percentage of

[†]Recording epochs of 20 to 25 milliseconds are necessary for adequate ABR threshold detection measures in infants, especially when tonal stimuli are used.[45]
[‡]Bone-conduction testing should be completed if air-conduction thresholds are greater than 20 dB nHL.[45]

children with UHL or MBHL have significant speech-language delays, negative educational consequences, and behavioral problems associated with the presence of their hearing loss.[8] Identification of UHL and MBHL through improved hearing screening programs during the newborn, early childhood, and school-age period offers an opportunity to provide intervention, parent education, and preventative measures to improve outcomes for children before problems arise. Similar to what happened in the development of hospital-based newborn hearing screening programs, achieving these goals will require input and participation from many different stakeholders, including public health and education officials at the local, state, and national level, health care providers, parents, and professional and advocacy organizations.

ABBREVIATIONS

A-ABR	automated auditory brain-stem response
AAP	American Academy of Pediatrics
ABR	auditory brain-stem response
CDC	Centers for Disease Control and Prevention
EHDI	Early Hearing Detection and Intervention
EPSDT	Early and Periodic Screening Diagnosis and Treatment
HL	hearing level
JCIH	Joint Committee on Infant Hearing
MBHL	mild bilateral hearing loss
NHANES	National Health & Nutrition Examination Survey
nHL	normalized hearing level
OAE	otoacoustic emission
UHL	unilateral hearing loss

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