Issues and Concerns Associated with Universal Newborn Hearing Screening Programs

Questions et préoccupations associées aux programmes de dépistage universel de la surdité chez les nouveau-nés

Fred H. Bess and Terrey Oliver Penn
Vanderbilt University Medical Center
Nashville, Tennessee

Abstract

Since the initial call for universal newborn hearing screening (UNHS), an intense professional debate regarding the possibility, effectiveness, efficiency, and risks has ensued in the professional literature. Despite the earlier controversies and continuing concerns surrounding UNHS, more recent and widely publicized support for UNHS has lead to a groundswell of new programs. This paper is intended not to take a position on the value of UNHS, but to remind professionals involved in UNHS that critical issues remain that warrant serious discussion. This paper focuses on several important issues and concerns that program leaders will encounter in the development and implementation of UNHS. These include programmatic (i.e., predicting screening outcomes, improving screening outcomes, and optimizing performance), cost, legal, follow-up, and training issues associated with UNHS. By attending to and selecting the most successful aspects of programs one can establish a high-quality standard that will provide momentum for UNHS growth.

Abrégé

Depuis qu'a été proposé le dépistage universel de la surdité chez les nouveau-nés (DUSN), un intense débat concernant les possibilités, l'utilité, l'efficacité et les risques d'un tel programme a émergé dans la littérature professionnelle. Mais malgré les controverses initiales et les préoccupations toujours actuelles au sujet du DUSN, le large engouement public récent en sa faveur a favorisé l'apparition de nouveaux programmes. Cet article ne vise pas à prendre position quant à l'utilité du DUSN, mais plutôt à rappeler aux professionnels que des questions importantes restent à étudier. Il met l'accent sur les questions et préoccupations fondamentales auxquelles les responsables de programmes devront faire face lors du développement et de la mise en œuvre du DUSN. Citons notamment les questions liées à l'exécution des programmes (c.-à-d. la prévision et l'amélioration des résultats du dépistage ainsi que l'optimisation de la performance), aux coûts, à l'aspect juridique, au suivi et à la formation. En veillant à sélectionner les éléments les plus performants des programmes, une norme de qualité élevée pourra être établie, ce qui donnera un élan au développement des programmes de DUSN.

Key words: universal newborn hearing screening, screening outcomes

In 1993, the National Institutes of Health (NIH) hosted a conference that concluded with a Consensus Statement on Early Identification of Hearing Impairment in Infants and Young Children (NIH, 1993). The statement called for the "screening of all newborns, both high and low risk, for hearing impairment prior to hospital discharge." An intense professional debate ensued regarding the possibility, effectiveness, efficiency, and risks associated with universal newborn hearing screening (UNHS; Bess & Paradise, 1994a; 1994b; Northern & Hayes, 1994; White & Maxon, 1995). In fact, one year after the NIH Consensus Conference, the representatives from the member organizations of the Joint Committee on Infant Hearing Screening (JCIH; 1994) found themselves unable to agree unanimously with the 1993 NIH Consensus Committee recommendation. Apparently, the empirical evidence available at that time was inadequate to support such a recommendation by the JCIH. They instead agreed to endorse "the goal" of universal detection of hearing loss in newborns in the 1994 JCIH Position Statement. Some continue to suggest that the implementation of UNHS is premature and not cost effective (Berg, 1999; Clayton & Tharpe, 1998; Kemper & Downs, 2000; Kileny & Jacobsen, 2000a, 2000b; Newman, 1998; Paradise, 1999; Stein, 1999; Tharpe & Clayton, 1997; Van Riper & Kileny, 1999).

Since the initial call for universal screening, a growing number of articles, both subjective and objective, have appeared in the professional literature. Content has ranged from descriptions of technologies and procedures to broad discussions of policies and ethics of hearing screening, to outcomes research aimed at program assessment and quality assurance.

Today, a number of reports from large multi-site UNHS programs are available for review. One result of this growing body of information has been endorsement of UNHS, without reservation, by the current representatives of the member organizations of JCIH (2000). Furthermore, over $4 million (US) in federal funds were appropriated for universal newborn hearing screening and early intervention grants to states for the fiscal year 2000.

Despite the earlier controversies and continuing concerns surrounding UNHS, the more recent and widely publicized support for UNHS inevitably will extend the groundswell of new programs. Thus, this paper is intended not to take a position on the value of UNHS, but to remind professionals involved in UNHS that critical issues remain, issues that warrant serious consideration. Inevitably, additional issues will surface as UNHS momentum builds and programs increase in number, size, and complexity. This paper focuses on some of the important issues and concerns that program leaders will no doubt encounter in the development and implementation of UNHS.

Programmatic Issues Associated With UNHS

Predicting Screening Outcomes

Congenital hearing loss is a relatively low prevalence disorder. Approximately one to three in 1,000 newborns will have congenital hearing loss. Ideally, one wants a screening tool that identifies all newborns with a hearing loss, yet keeps the number of false positive outcomes to a minimum. Performance characteristics of tests and screening tools help one to determine the effectiveness of any screening protocol. Remember that sensitivity and specificity quantify a test's accuracy in the presence of a known diseased or disordered condition. A positive predictive value (PPV) is the probability of having a disorder when the test is positive. A negative predictive value (NPV) is the probability of not having a disorder when the test is negative. Stated otherwise, sensitivity and specificity quantify a test's accuracy given the *known condition of the patient.* Predictive values quantify a test's accuracy given *only the test results.* Using these performance characteristics of a screening tool one can predict screening outcomes for a given population.

Screening for rare conditions, even with tests high in sensitivity and specificity, can lead to a significant number of false positive outcomes. Figure 1 shows how sensitivity, specificity, and the prevalence rate influence predictive values. This figure includes outcomes for a screening across three populations with disorders varying in degree of prevalence. The sensitivity and specificity of the test are held constant at 99% and 98%, respectively. A relatively high prevalence rate of 100/1,000 for Population A results in a PPV of 0.85. That is, given a positive screening outcome, the probability that this person has the disorder is 85%. For Population B with a lower prevalence rate of 10/1,000, the PPV drops to 0.36 or a 36% probability. Population C has a prevalence rate similar to estimates for congenital hearing loss, 3/1000. In this case, the PPV drops to a level of 0.13 or a 13% probability. Stated otherwise, 87% of the positive test results come from people who do not have the disorder. On the other hand, the NPV is high at 99% for all three levels of prevalence. If someone passes the screening, it is highly unlikely that they have the

Figure 1. Effect of three prevalence rates on screening results. Sensitivity = 0.99, Specificity = 0.98.

Population A

Prevalence = 100/1,000 = 0.10

	Disorder +	Disorder –	Total
Fail	99,000	18,000	117,000
Pass	1,000	882,000	883,000
	100,000	900,000	1,000,000

PPV = 99,000 / (117,000) = 0.85

NPV = 882,000 / (883,000) = 99.9

Population B

Prevalence = 10/1,000 = 0.01

	Disorder +	Disorder –	Total
Fail	9,900	19,800	29,700
Pass	100	970,200	970,300
	10,000	990,000	1,000,000

PPV = 9,900 / (29,700) = 0.36

NPV = 970,200 / (970,300) = 99.9

Population C

Prevalence = 3/1,000 = 0.003

	Disorder +	Disorder –	Total
Fail	2,970	19,940	22,910
Pass	30	977,060	977,090
	3,000	997,000	1,000,000

PPV = 2,970 / (22,910) = 0.13

NPV = 977,060 / (977,090) = 99.9

disorder. In general, the prevalence of a disorder has a profound effect on the usefulness of a test. For even the best newborn hearing screening tool, the PPV will remain low due to the relatively low prevalence of congenital hearing loss, unless some consideration is given to improve outcomes. High levels of false positive outcomes should be expected in a UNHS program, and this probability should be a critical concern for UNHS program directors.

Improving Screening Outcomes

Several program characteristics can be selected to improve the screening outcomes. First, employing technology with superior performance characteristics is the single best way to improve the chances of an efficient and effective screening program. Technological advancements in program protocols and procedures have been instrumental in the improved false positive rates reported by many UNHS programs over the past decade.

The earliest report of infant hearing screening in the United States appeared nearly 40 years ago (Downs & Sterritt, 1964). This early program used behavioral observation of infants in response to intense auditory stimuli in order to screen for hearing status. Although this subjective technique was easy to use and inexpensive, ultimately it was found to have inadequate sensitivity and specificity rates. Since that initial report, a number of screening protocols and techniques have been proposed and evaluated (Hayes & Northern, 1997). Today, the emphasis is placed on techniques that provide objective, physiologic measures to screen for hearing status. Specifically, two measures dominate the field of newborn hearing screening: evoked otoacoustic emissions (EOAEs) and the auditory brainstem response (ABR). Two types of EOAEs, transient (TEOAEs) and distortion product (DPOAEs), are used in newborn hearing screening programs. Automated ABR (AABR) is more commonly used than conventional (CABR) in UNHS programs. Both technologies are straightforward, noninvasive, and allow for quick measurement of a newborn's hearing status within the hospital nursery environment. Conventional and automated test instruments that incorporate either technique are available. The placement of these two devices in existing UNHS programs is fairly evenly distributed. Further, most legislative mandates do not recommend a specific technology, but are often written to require that non-specific, objective, and physiologic measures be used. Undoubtedly, the recent advances in hearing screening technology support the acceptance and practice of universal newborn hearing screening and, in turn, the increase in acceptance and practice stimulates refinement in technology. Continued improvements in technology certainly will occur, always with improvements in false positive percentages.

A second method to improve false positive rates is to adjust the cut-off criterion for failure. The number of false positives will be reduced at the expense of a few true positives when the failure criterion is raised and the condition being screened for is rare. In other words, only a few additional children with congenital hearing loss would be missed, but many more children who would have failed the screening will now pass. Hyde, Riko, and Malizia (1990) showed this effect by increasing the ABR intensity cut-off by 10dB. When the level changed from 30 to 40dB HL, 67 false positive outcomes became true negatives at the expense of two true positives.

The third method often used to address this inherent problem in screening rare disorders is to provide sequential testing strategies (Truman & Teutsch, 1998). There are two sequential testing strategies, parallel testing and serial testing. In the case of parallel testing, two or more tests are given regardless of the result of the initial screen. A patient would be referred for diagnostic testing after failing *any* of these tests. In serial testing, the second test is completed *only if* the first test is failed. In both strategies, the multiple tests could include a new technique or tool, or it could mean simply

Figure 2. The effect of a sequential screening strategy on screening results. Sensitivity = 0.99, Specificity = 0.98.

A

Initial Screen

Prevalence = 3/1,000 = 0.003

	Disorder +	Disorder –	Total
Fail	2,970	19,940	22,910
Pass	30	977,060	977,090
	3,000	997,000	1,000,000

B

Second Screen

Prevalence = 2,970/22,910 = 0.13

	Disorder +	Disorder –	Total
Fail	2,910	399	3,309
Pass	60	19,541	19,601
	2,970	19,940	22,910

C

Final Screening Outcome

Prevalence = 3/1,000 = 0.003

	Disorder +	Disorder –
Fail	2,910	399
Pass	90	996,601

Sensitivity = 2,910 /3,000 = 97%
Specificity= 996,601/997,000 = 99%
PPV = 2,910 / 3,309 = 0.88
NPV = 996,601 / 996,691 = 0.99

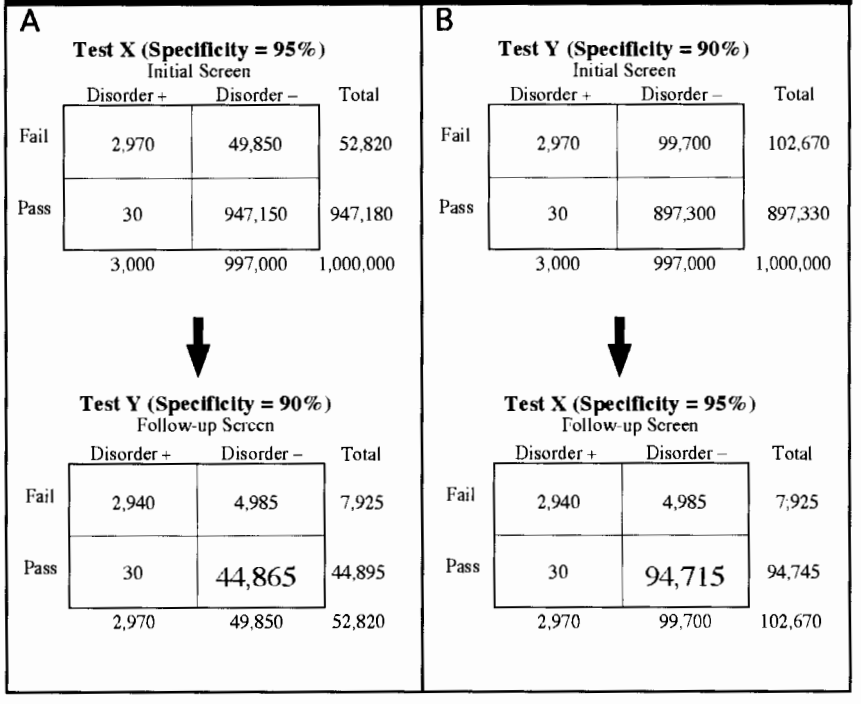
rescreening with the same tool. Parallel testing is often used when available tests are relatively insensitive, because this strategy increases sensitivity and PNV. However, it will decrease specificity and PPV. Therefore, this strategy is not recommended in screening for congenital hearing loss. Serial testing, on the other hand, results in the converse. Specificity and PPV are increased, although sensitivity and NPV may be decreased. This strategy is used in screening programs such as screening for prostate cancer, HIV, or screening for Down syndrome during pregnancy. The initial test in serial screening typically is less risky to the patient, less expensive, or less complicated procedurally. Figure 2 illustrates this method used for a disorder with a prevalence rate similar to congenital hearing loss. In Panel A, the initial screening results are shown. Panel B shows the results of the second screening for the 22,910 patients who failed the initial screen. Because the prevalence of hearing loss in this second screened group is higher than in the general neonatal population, one would expect the PPV to increase. Panel C shows the combination of both screenings. After failing both screenings, there is an 88% chance that the patient would have the disorder (i.e., PPV). The NPV remains high at 99%. Notice that the sensitivity drops to 97% from 99%, but the specificity increased to 99.9% from 98% as predicted. Notably, false positives were reduced from 19,940 to 399 by simply adding a second screen. Of course, this method will not improve the chance of identifying a false negative from the first screen. After the second screen, an additional 60 children pass the screening that have the disorder, which may constitute an unacceptable tradeoff.

The screening order will effect the efficiency of the final screening result. Ideally, when a sequential test strategy is used, the first screen should have higher specificity and the second should have higher sensitivity. Figure 3 illustrates this point. The exact number of true positives was identified using either sequence (i.e., 2,910). However, using the tool with the lower specificity rate first nearly doubled the number of children that had to be screened a second time. If both tools have a similar cost associated with them, and they are equally invasive, then it is clear which test should be used first - the one that will provide the lowest number of false positives. Although, if there is some risk associated with one technique and not the other, or one is significantly more expensive, then the less risky or less costly tool might be more appropriate for the initial screen.

For maximum benefit, the two tests should be relatively independent. The second should not simply duplicate known information. It is known that the two physiologic measures used in UNHS, EOAEs and ABR are highly correlated with the status of peripheral hearing sensitivity. EOAEs are generated by the outer hair cells of the cochlea, and the presence of EOAEs does not ensure the integrity of the neural auditory pathway. The ABR, however, does reflect activity of the cochlear, auditory nerve, and the auditory brainstem pathways. The ABR is sensitive to auditory nerve and brainstem abnormalities. Other differences include the susceptibility to middle ear disorder. EOAEs are much more sensitive to middle ear disorder than the ABR. The differences and the similarity of these two techniques suggest they are effective tools for a sequential screening strategy. Of course, in 1993 the NIH Consensus Group recommended a sequential testing strategy with EOAE screening followed by ABR screening.

A fourth method to reduce the high false positive outcomes is to screen only a subgroup at risk for a specific condition. It remains true that screening programs based solely on risk indicators will miss as many as 50% of infants with hearing loss (Finitzo, Albright, & O'Neal, 1998; Mauk, White, Mortensen, & Behrens, 1991). Despite the lack of significant

Figure 3. The effect of test order in a sequential screening strategy. For Test X, specificity = 95% and for Test Y, specificity = 90%. For both tests, sensitivity = 99%. Panel A shows the effect of a higher specificity rate for the initial test. Panel B shows the effect of higher specificity for the second test.



correlation with congenital hearing loss, many risk indicators can be successful in targeting children who develop later onset of hearing loss (Feiner, Pardue, Raffin, & Matz, 1996). In fact, the JCIH (2000) continues to address risk indicators, but focuses the role of these primarily for the identification of late-onset or progressive hearing loss. Risk indicators certainly have a place in identifying who should be monitored for hearing loss or hearing difficulties throughout the early developmental years (Dahle et al., 2000; Lutman, Davis, Fortnum, & Wood, 1997). Therefore, it is important for UNHS program directors to determine methods to provide for the continued surveillance for late-developing sensorineural hearing loss in an effective, efficient, and cost-effective manner.

With the exception of improved technology, these methods to reduce false positive outcomes require a trade-off. As the number of false positive decreases, the number of false negative increases, albeit by a much smaller percentage of the population screened. Directors of UNHS programs should consider these performance characteristics of screening tools carefully. Then, these various options to improve the screening program's performance should be weighed against the costs of the false positives to the program and to the families.

Optimizing Performance

Selection of Technology

In 1997, the National Institute on Deafness and Communication Disorders (1997) Working Group on Early Identification of Hearing Impairment suggested that no more than 5% of neonates with no risk indicators and 8% of those at risk should be referred. Later, in 1999, the Task Force on Newborn and Infant Hearing for the American Academy of Pediatrics (1999) published guidelines that recommended the referral rate not exceed 4%. The new 2000 JCIH Guidelines include this 4% criterion as a benchmark for quality UNHS programs. Program directors should consider these recommended guidelines when selecting appropriate technology.

In the previous section, an example of how programs using serial screening strategies improve false positive rates was included. Interestingly, UNHS programs using serial testing with two technologies (e.g., EOAEs and AABR/CABR) or AABR alone often result in superior failure rates over serial testing with TEOAEs alone (Barsky-Firkser & Sun, 1997; Clemens, Davis, & Bailey, 2000; Finitzo et al., 1998; Gravel et al., 2000; Huynh, Pollack, & Cunningham, 1996; Mason & Herrmann, 1998; Vohr, Carty, Moore, & Letourneau, 1998). Typically, for established UNHS programs using AABR alone or a two-technology protocol, the reported failure rates are well below the recommended level of 4%. Conversely, those UNHS programs using a one-technology protocol of TEOAEs often report levels above this criterion. Finitzo and colleagues (1998) reported failure rates across nine hospitals using a serial screening strategy. Two hospitals used an AABR and three used EOAE technology. All newborns failing an initial screen were rescreened with the same technology before hospital discharge. The remaining four hospitals used a two-technology (i.e., TEOAEs then AABR) serial screening strategy. The annual failure rates reported across the hospitals screening with EOAEs alone, AABR alone, and the two-technology protocols were averaged. It was found that the average failure rates for these three groups were 12.3, 1.4, and 3.0%, respectively. Mason and Herrmann reported a false positive rate of 0.2% when using a serial screening strategy with AABR alone. Gravel and colleagues reported average failure rates for well-baby nursery (WBN) screening across three years in eight New York hospitals. Six hospitals used a two-technology protocol of TEOAEs for the initial screen and ABR, either conventional or automated, for the second screen and had an average failure rate of 2.4%. Two hospitals used EOAEs only and had an average failure rate of 8.5%. They also reported that after three years experience, the lowest failure rates were observed for a two-technology protocol rather than the sole technology of TEOAEs (Gravel et al.).

It is known that the prevalence rate for congenital hearing loss in the neonatal intensive care units (NICU) is higher than in the WBN. Spivak and colleagues (2000) found that a higher percentage of infants from NICU failed than from the WBN for six of eight New York hospitals. The failure rate for infants in the NICU was 6.2% and in the WBN it was 3.9% (Prieve et al., 2000). Mason and Herrmann (1998) found a similar relationship; failure rate for the NICU infants was 4.8% and for the infants in the WBN the failure rate was 3.9%. Considering the larger population in the WBN, these small differences in percentages would translate to significantly more infants failing the hearing screening in that setting. Interestingly, the higher failure rates for the NICU population versus the WBN nursery population occurred irrespective of the various technologies employed (e.g., TEOAE only, TEOAE/AABR, TEOAE/CABR). However, the average failure rate for TEOAEs alone was higher at 8.7% compared to 5.1% for the two-technology protocol (Gravel et al., 2000). Meyer and colleagues (1999) compared EOAE outcomes for 464 infants with risk indicators for hearing loss assuming the AABR as the "gold standard". EOAEs agreed with the AABR outcomes 74% of the time. In addition, seven false negatives and 120 false positives resulted from the comparison revealing a sen-

sitivity of 71% and specificity of 73% for EOAEs in this population. The overall data suggest the EOAEs are less effective in correctly identifying congenital hearing loss in an NICU population.

In either the WBN or the NICU, technology is available to meet the guidelines for optimal failure rates. Keeping failure rates low is a critical issue for screening programs. Choosing the optimal technology or technologies is an important decision for maintaining an efficient and effective UNHS program.

Infants Missed During the Inpatient Screening

What should one do with infants who miss the newborn hearing screening as inpatients? How these infants are screened as outpatients and how they are recorded in program statistics are both important issues. Of course, these infants should be referred for outpatient screening. A protocol similar to those used for rescreening inpatient failures should be applied to this population. In addition, for data management, the number of children in both of these groups (missed infants and screening failures) should be added to determine the overall failure rate. Every attempt should be made to keep the number of missed children low through careful and thorough protocol development or else the screening program cannot reach an acceptable failure rate under 4%.

Often it is thought that the newborn in the WBN will be available for screening for a limited time, but that the newborn in the NICU is "captive" for days, weeks or even months. However, this is not always the case. Newborns requiring NICU care are often quickly transferred to other hospitals based on medical needs or are transferred to other intensive care nurseries closer to home after their health condition has stabilized. Spivak and colleagues (2000) found that this was the case for NICU newborns in the New York State Universal Newborn Hearing Screening Demonstration Project. Although newborns often have a lengthy stay in the NICU, they found that the number of newborns missed there far exceeded the number missed in the WBN. Surveillance of all components of the UNHS program will identify these situations so that protocols can be continually refined and improved.

Cost Issues Associated with UNHS

Screening programs are expensive and constitute a substantial portion of today's healthcare practices. The billions of dollars expended for just three of the widely recognized screening programs in the United States (i.e., cervical cancer, prostate cancer, and high blood levels of cholesterol) are sufficient to fund the basic healthcare system for all of the poor and uninsured (Russell, 1994)! For some disorders, it is more expensive to implement screening programs than it is to treat the actual disease (Russell). Factors that contribute to the high costs of screening programs include expensive screening tests, tests with low sensitivity and specificity, low prevalence of the disorder even if the specificity is high, high referral rates, unnecessary follow-up tests, poor compliance, and salaries of audiologists.

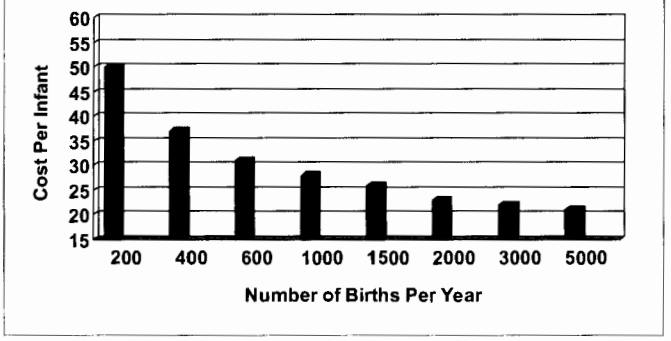
Much of the controversy surrounding UNHS has centred on the issue of cost. Pundits of UNHS have argued that a more evidence-based, cost effective approach to early identification of hearing loss is to screen all high risk and intensive care unit babies because such an approach will detect most children with congenital hearing impairment (Bess & Paradise, 1994a,b; Kileny & Jacobson, 2000a, 2000b; Paradise, 1999). A substantial portion of the audiology community, however, argues that UNHS is well worth the costs when one considers the benefits and savings derived from early detection and subsequent intervention (Hayes & Downs, 2000; Prieve et al., 2000; Robinette & White, 1998). Given that a goal of health promotion is cost containment, it is important for program directors to consider both sides of these arguments and to possess a good understanding of the direct and indirect costs associated with newborn screening *before* a program is implemented.

Direct costs associated with screening programs typically include costs of equipment, supplies, and personnel. Other direct expenses not usually considered in a cost analysis include organization/administration expenses, data collection, data analysis and interpretation, record-keeping communication, training of test administrators/interpreters, and program evaluation. Estimates of the direct monetary costs associated with UNHS range between 144 million and 200 million dollars. The variation in estimated costs is due, in part, to the differences in prevalence rates used in the cost calculations. For example, the cost per identified child is $50,000 if the prevalence is 1/1000 and $12,500 per identified child if the prevalence is 4/1000 (Robinette & White, 1998).

Barker (1995) calculated the hospital costs for the NIH protocol using a sensitivity analysis. The analysis assumed an average of 1,184 babies born in the hospital each year, an EOAE specificity of 85%, an ABR specificity of 90%, a prevalence of 1/1000, and a salary of $39,250 for an audiologist to administer the program. Barker found that the average annual hospital cost of the NIH protocol was $31,314.17, or $26.45 per infant. Interestingly, Barker noted that approximately half of the total annual expenses went toward follow-up costs for

infants who failed the EOAE screen; of this figure, 99.1% of the costs were for the follow-up of infants with false positive screening tests. Importantly, the total hospital cost, cost per infant, and cost per child identified will vary based on the number of babies born per year. Figure 4 illustrates the effect

Figure 4. Effect of hospital size on cost per infant (from Barker, 1995).



of hospital size on cost per infant. It is seen that the smaller the size of the hospital, the higher the screening costs per infant; an important consideration for smaller hospitals contemplating the development of a newborn screening program.

Far less is known about the indirect costs associated with UNHS. Indirect costs of a screening program can include transportation and parking costs, unnecessary tests and treatment, lost wages during follow-up testing, and the more human costs of distraction, stress, frustration, and parental anxiety (Bess & Paradise, 1994a). Of special concern is the potential deleterious effect of large numbers of false positive identifications. How many infants will receive how many unnecessary test procedures? How many infants will be unfavorably labeled? And, what are the effects of false positive identification on the parent and the parent-child bonding? In developmental pediatrics, there is mounting evidence to show that if a child is identified as disordered in the newborn period, even if incorrectly, and even for only a short period of time, there can be long-lasting effects on the parent, the parent-child relationship, and subsequently, the child's psychosocial development (Bess & Paradise, 1994a,b). In fact, parents can become unduly protective or attached to their child as a consequence of the screening outcome and, subsequently, over-utilize the healthcare system. Such a phenomenon has been referred to as the "vulnerable child syndrome". These indirect costs, seldom, if ever receive the same consideration that is given to the benefits of screening babies for hearing loss. Moreover, there has been limited research in hearing screening programs directed toward assessing the impact of false positives on parents; no studies have explored the issue of parent-child bonding. Abdala de Uzcategui and Yoshinaga-Itano (1997) surveyed a small number of parents whose infants had failed the initial screen and were referred for additional testing. Some of the results of the survey revealed that 22% experienced anger, 42% were confused, 52% were afraid, 37% were depressed, 31% were frustrated and 19% expressed feelings of guilt. In another study, Luterman and Kurtzer-White (1999) reported that 17% of the parents surveyed did not want to know at birth that their baby was deaf. Finally, Stuart, Moretz, and Yang (2000) examined stress levels in a small group of mothers whose infants had failed a newborn hearing screen and compared these findings to the stress levels of 20 mothers whose children had passed the hearing screen. Stuart and coworkers reported essentially no differences in stress levels between the groups. Although these studies represent a good first start, more systematic, carefully controlled research is clearly needed before one can determine whether false positive identifications impact negatively on parents and on the parent-child relationship.

Legal Issues in UNHS

More than 32 states in the United States have now developed legislation mandating UNHS. Interestingly, when comparing state regulatory laws, programs vary significantly with regard to type of screening recommended, program participation and informed parental permission, fees for the screening service, time limitations, and screening and follow-up responsibilities (Clayton & Tharpe, 1998). It is important to recognize that once a healthcare practice is regulated by law it becomes the accepted standard of care and the potential for litigation is ever present. Failure to perform newborn hearing screening according to the law exposes the audiologist, medical staff, and the hospital to liability. For example, caution must be taken by the program leaders to insure that names are not released to outside parties without the written consent of the parent or guardian (Clayton & Tharpe). Confidentiality is a given right and provisions for confidentiality should be built into any screening program. The potential for litigation also exists if a child with hearing loss is missed or fails to receive follow-up care. Other types of screening programs offer insight into the potential problems that can occur as a consequence of false negatives. Holtzman, Slazyk, Cordero, and Hannan (1985) found that 20% of the missed cases in PKU screening occurred during the follow-up stage of the program. Moreover, Holtzman, et al. found that 29% of the missed cases resulted in litigation. It is almost superfluous to note that the costs associated with these lawsuits were substantial.

In order for a lawsuit to take place, the child with a missed hearing loss (plaintiff) needs to demonstrate that the defendants breached the standard of care and that the breach caused damages that are compensable under the law (Clayton & Tharpe). As noted earlier, the standard of care is defined as the practice behaviour of other practitioners within the community and the state. If a hospital fails to perform newborn hearing screening when most others within the community and the state do, then, a breach of standard is evident. The risk of litigation is even greater in states that have mandated hearing screening. Failure to comply with state requirements may be sufficient cause to prove breach of the standard of care irrespective of what other providers do within the community.

Finally, the child who is missed cannot successfully sue a state program because such programs are typically immune from liability (Clayton & Tharpe, 1998). However, the likelihood is high that the child's representatives will sue the audiologist, hospital staff, and the hospital in their search for compensation. For example, the child's parents could sue for compensation for the pain and suffering experienced by the child or for their own emotional and economic loss (Clayton & Tharpe). Such issues are important to consider in the development of newborn hearing screening programs and in the development of any state regulatory laws.

Follow-up Issues in UNHS

Perhaps the most important component of any newborn screening program is the follow-up of those children who have failed the screening test. In order for the follow-up to be successful, there must be suitable facilities that are readily available and accessible, there must be adequate compliance, and for those who do comply, there must be a timely and an appropriate diagnosis and intervention. It is well recognized that 25% of all births in this country occur in rural or remote areas, many of which lack qualified audiologic professionals and sophisticated audiometric equipment; under such circumstances, the screening and follow-up of infants who have failed the test could pose formidable problems of logistics and costs. Indeed, it is inappropriate to screen for any disorder without certainty that facilities for suitable follow-up care of individuals who fail the screen are readily available. Moreover, families in such areas often lack health insurance or are underinsured so that even if suitable professional services were available, they might not be affordable for such families until such time as a national health insurance program had come into being. One possible way to minimize the problems associated with screening infants located in remote and rural regions is to create regional centres of excellence (Bess, 2000). That is, to establish referral centres that meet specified criteria for serving young children with hearing impairment.

The issue of compliance is also a concern for newborn hearing screening programs. On average, 20 to 30 percent of those infants who fail the newborn screening are lost to follow-up despite aggressive recruiting efforts and cost-saving incentives to the parents (Dalzell et al., 2000; Diefendorf & Weber, 1994; Kileny & Jacobson, 2000b; Shimizu, et al., 1990). Noncompliance rates are usually highest in the beginning years of a program. Recently, the New York State Screening Project reported that 15 to 27 percent of the babies who failed the screening did not return for follow-up (Prieve et al., 2000) - Henry Ford Hospital (Detroit, MI) reports a failure rate of 25 percent. Screening is of limited value if large numbers of infants fail to comply with the recommendation of the screen.

Several other important issues in the follow-up process represent potential areas of concern. The increased numbers of newborn hearing screening programs has resulted in large numbers of young infants being referred to audiologic facilities for appropriate diagnosis and intervention. Unfortunately, one continues to find that a substantial lag time exists between the initial confirmation of hearing loss and the fitting of amplification (Bess, 2000). Almost half of the screening sites of The Marion Downs National Center for Infant Hearing reported that the average initial diagnostic evaluation occurred within the first six months of life; 33% reported that the average age of confirmation of hearing loss was within the first six months of age. In more than half of these sites, however, children did not receive amplification until they were more than 12 months of age.

It is also troubling to note that many clinicians do not use the available technology and evidence-based practices for the fitting of appropriate amplification to very young infants referred from UNHS programs. Hedley-Williams, Tharpe, and Bess (1996) demonstrated from a large survey of pediatric audiologists that no systematic procedure exists for determining and fitting hearing aids for young children. The survey showed that many of the prescriptive procedures available for selecting hearing aids were not being used, probe-tube measurements with children are typically not included as part of the verification strategy, and programmable hearing aids are seldom, if ever, selected for young infants. Tharpe (2000) reported similar findings.

Finally, there is evidence to suggest that a needs exists for audiologists to develop better counseling skills - skills that are so important for communicating screening test results to

young parents (Luterman & Kurtzer-White, 1999).

Training Issues in UNHS

One way in which to minimize the potential problems of universal newborn hearing screening is to improve the educational/professional training of audiologists. It is well recognized that many universities offer only one course in pediatrics—the course typically offers limited exposure to pediatric assessment (i.e., < 6 months), hearing aid selection and fitting, and counseling. In a recent survey it was found that only 18% of the respondents had taken a course in pediatric amplification in graduate training programs (Tharpe, 2000). The increased numbers of babies that are being referred to audiologists from UNHS programs prompts a need for audiologists to receive better, more comprehensive training in the areas of pediatric assessment, pediatric amplification fitting, counseling, and early identification.

The areas of pediatric assessment and hearing aid fitting are especially important to the success of any UNHS programs. The very fact that some pediatric audiologists wait until a child is more than 12 months of age for a hearing aid fitting following identification is clearly unacceptable. One must fit children with amplification shortly after they have been identified if one expects to offer them the best possible services. The technology and the know-how for the assessment and hearing aid fitting of infants below six months of age are currently available and should be used. Comprehensive information is available on recommended protocols for pediatric assessment and hearing aid fitting of very young infants (Bess, Gravel, & Tharpe, 1996; Seewald, 2000).

Unfortunately, one finds problems with the preparation of audiologists in the area of counseling. Several studies have demonstrated that students receive minimal exposure to family counseling (Crandell, 1996; 1997). In the words of Luterman and Kurtzer-White (1999), "with the number of hearing screening programs rapidly expanding, highly trained professionals with parent counseling skills are at a premium, as are specialists in working with infants with hearing loss and their families".

Audiologists must also develop skills in the area of early intervention. In general, university programs emphasize training of audiologists for populations above the toddler age and seldom give any preparation on effective techniques for working with very young children. Even deaf education programs fail to prepare adequately educators to work with younger age groups. According to Roush and co-workers (1992), less than half of deaf education programs offer any kind of preparation in the area of early intervention.

Finally, one cannot over-emphasize the importance of educating primary care physicians, healthcare personnel, and parents to recognize the potential signs of hearing loss in young infants and to make prompt referrals for audiologic assessment and management.

Summary

Forty years have now passed since the initial publication of reports describing programs to screen hearing in infants. Even less time has passed since the first reports of universal screening were published. UNHS is still in its early development - less than a decade old. Recognizing this, one can be fairly certain that circumstances naturally will arise that will suggest modifications or alterations in current methods and protocols. New technological advancements are likely and these will influence protocol. As published reports appear in the literature describing experiences within existing UNHS programs, further policy and procedural changes may be warranted. Therefore, vigilant monitoring of existing programs, on local, state, and national levels should be a natural component of all programs. By selecting the most successful aspects of programs nationwide, one can establish a high-quality standard that will provide momentum for UNHS growth.

To those audiologists who are considering the development of a UNHS program careful planning, reliance on evidence-based data, and caution are urged. Premature implementation of a newborn screening could well work against the ultimate success of the program— good intentions do not always ensure the success of UNHS. To be sure, the most successful UNHS programs will be those that are dynamic, entrepreneurial, evidence-based, and willing to change practices even when the status quo is more comfortable.

Author Note

Please address all correspondence to Fred H. Bess, PhD, Professor and Chair, Department of Hearing and Speech Sciences, Vanderbilt Bill Wilkerson Center for Otolaryngology and Communication Sciences, Vanderbilt University Medical Center, Nashville, Tennessee 37232. Email: fred.h.bess@vanderbilt.edu.

References

Abdala de Uzcategui, C., & Yoshinaga-Itano, C. (1997). Parents' reactions to newborn hearing screening. *Audiology Today, 9*(1), 24, 26.

American Academy of Pediatrics. (1999). Task Force on Newborn and Infant Hearing. Newborn and infant hearing loss: Detection and intervention. *Pediatrics, 103,* 527-530.

Barker, A. M. (1995). *Cost analysis of universal newborn hearing*

screening. Unpublished master's thesis, Vanderbilt University, Nashville, TN.

Barsky-Firkser, L., & Sun, S. (1997). Universal newborn hearing screenings: A three-year experience. *Pediatrics, 99,* 862. Retrieved December 1, 2000, from the World Wide Web: http://www.pediatrics.org/cgi/content/full/99/6/e4.

Berg, A. (1999). Comment on: Universal newborn hearing screening: Should we leap before we look? *Pediatrics, 104,* 351-352.

Bess, F. H. (2000). Early amplification for children: Implementing change. In R. C. Seewald (Ed.), *A sound foundation through early amplification* (pp. 247-251). Staefa, Switzerland: Phonak AG.

Bess, F. H., Gravel, J. S., & Tharpe, A. M. (Eds.). (1996). Amplification for children with auditory deficits. Nashville, TN: Bill Wilkerson Center Press.

Bess, F. H., & Paradise, J. L. (1994a). Universal screening for infant hearing impairment: Not simple, not risk-free, not necessarily beneficial, and not presently justified. *Pediatrics, 93,* 330-334.

Bess, F. H., & Paradise, J. L. (1994b). Universal screening for infant hearing impairment: a reply. *Pediatrics, 94,* 959-963.

Clayton, E. W., & Tharpe, A. M. (1998). Ethical and legal issues associated with newborn hearing screening. In F. H. Bess (Ed.), *Children with hearing impairment: Contemporary trends* (pp. 33-44). Nashville, TN: Bill Wilkerson Center Press.

Clemens, C. J., Davis, S. A., & Bailey, A. R. (2000). The false-positive in universal newborn hearing screening. *Pediatrics, 106,* 128-129. Retrieved December 1, 2000, from the World Wide Web: http://www/pediatrics.org/cgi/content/full/106/1/e7.

Crandell, C. C. (1997). An update on counseling instruction within audiology programs. *The Journal of the Academy of Rehabilitative Audiology, 30,* 77-86.

Crandell, C. C., McDermott, D. J., & Pugh, K. (1996). A survey on amplification and counseling skills in audiology. *The Hearing Review, 3*(9), 26-29, 48.

Dahle, A. J., Fowler, R. B., Wright, J. D., Boppana, S. B., Britt, W. J., & Pass, R. F. (2000). Longitudinal investigation of hearing disorders in children with congenital cytomegalovirus. *Journal of the American Academy of Audiology, 11,* 283-290.

Dalzell, L., Orlando, M., MacDonald, M., Berg, A., Bradley, M., Cacace, A., Campbell, D., DeCristofaro, J., Gravel, J., Greenberg, E., Gross, S., Pinheiro, J., Regan, J., Spivak, L., Stevens, F., & Prieve, B. (2000). The New York State Universal Hearing Screening Demonstration Project: Ages of hearing loss identification, hearing aid fitting, and enrollment in early intervention. *Ear and Hearing, 21,* 118-130.

Diefendorf, A. O., & Weber, B. A. (1994). Identification of hearing loss: Programmatic and procedural considerations. In J. Roush & N. Matkin (Eds.), *Infants and toddlers with hearing loss: family-centered identification, assessment and intervention* (pp. 43-64). Baltimore, MD: York Press.

Downs, M. P., & Sterritt, G. M. (1964). Identification audiometry for neonates: A preliminary report. *Journal of Audiology Research, 4,* 69-80.

Feiner, M. V., Pardue, K. M., Raffin, M. J. M., & Matz, G. J. (1996). Infant hearing screening program: High-risk factors for hearing loss. *Seminars in Hearing, 17,* 165-170.

Finitzo, T., Albright, K., & O'Neal, J. (1998). The newborn with hearing loss: Detection in the nursery. *Pediatrics, 102,* 1452-1460.

Gravel, J., Berg, A., Bradley, M., Cacace, A., Campbell, D., Dalzell, L., DeCristofaro, J., Greenberg, E., Gross, S., Orlando, M., Pinheiro, J., Regan, J., Spivak, L., Stevens, F., & Prieve, B. (2000). New York State Universal Newborn Hearing Screening Demonstration Project: Effects of screening protocol on inpatient outcome measures. *Ear and Hearing, 21,* 131-140.

Hayes, D., & Downs, M. R. (2000). Value of UNHS is priceless. *The Hearing Journal, 53*(11), 61-66.

Hayes, D., & Northern, J. L. (1997). The quest for early identification of hearing loss. In D. Hayes & J. Northern (Eds.), *Infants and hearing* (pp. 3-27). San Diego, CA: Singular.

Hedley-Williams, A., Tharpe, A. M., & Bess, F. H. (1996). Fitting hearing aids in the pediatric population: A survey of practice procedures. In F. H. Bess, J. S. Gravel, & A. M. Tharpe (Eds.), *Amplification for children with auditory deficits* (pp. 107-122). Nashville, TN: Bill Wilkerson Center Press.

Holtzman, C., Slazyk, W. E., Cordero, J. F., & Hannan, W. H. (1985). Descriptive epidemiology of missed cases of phenylketurmia and congenital hypothyroidism. In L. B. Andrews (Ed.), *Legal liability and quality assurance in newborn screening* (pp. 17-20). Chicago, IL: American Bar Foundation.

Huynh, M. T., Pollack, R. A., & Cunningham, R. A. J. (1996). Universal newborn hearing screening: Feasibility in a community hospital. *Journal of Family Practice, 42,* 487-490.

Hyde, M. L., Riko, K., & Malizia, K. (1990). Audiometric accuracy of the click ABR in infants at risk for hearing loss. *Journal of the American Academy of Audiology, 1,* 59-66.

Joint Committee on Infant Hearing. (1994). Joint Committee on Infant Hearing 1994 Position Statement. *Asha, 36*(12), 38-41.

Joint Committee on Infant Hearing. (2000). Year 2000 Position Statement: Principles and guidelines for early hearing detection and intervention programs. *American Journal of Audiology, 9,* 9-29.

Kemper, A. R., & Downs, S. M. (2000). A cost-effectiveness analysis of newborn hearing screening strategies. *Archives of Pediatric and Adolescent Medicine, 154,* 484-488.

Kileny, P. R., & Jacobson, G. P. (2000a). Is UNHS Worth the cost? *The Hearing Journal, 53*(11), 61-67.

Kileny, P. R., & Jacobson, G. P. (2000b). Comment: The New York State Project. *Ear and Hearing, 21,* 640-641.

Luterman, D., & Kurtzer-White, E. (1999). Identifying hearing loss: Parents' needs. *American Journal of Audiology, 8,* 13-18.

Lutman, M. E., Davis, A. C., Fortnum, H. M., & Wood, S. (1997). Field sensitivity of targeted neonatal hearing screening by transient-evoked otoacoustic emissions. *Ear and Hearing, 18,* 265-276.

Mason, J. A., & Herrmann, K. R. (1998). Universal infant hearing screening by automated auditory brainstem response measurement. *Pediatrics, 101,* 221-228.

Mauk, G. W., White, K. R., Mortensen, L. B., & Behrens, T. R. (1991). The effectiveness of screening programs based on high-risk characteristics in early identification of hearing impairment. *Ear and Hearing, 12,* 312-319.

Meyer, C., Witte, J., Hildmann, A., Hennecke, K-H., Schunck, K-U., Maul, K., Franke, U., Fahnenstich, H., Rabe, H., Rossi, R., Hartmann, S., & Gortner, L. (1999). Neonatal screening for hearing disorders in infants at risk: Incidence, risk factors, and follow-up. *Pediatrics, 104,* 900-904.

National Institutes of Health. (1993). *Early identification of hearing impairment in infants and children. NIH Consensus Statement* (Vol. 11). Bethesda, MD: Author.

National Institute of Deafness and Other Communication Disorders. (1997). *Recommendations of the NIDCD Working Group on Early Identification of Hearing Impairment on acceptable protocols for use*

in state-wide universal newborn hearing screening programs. Bethesda, MD: Author.

Newman, L., (1998). Newborn hearing tests: Critics drowning in the tidal wave. *Medical Outcomes & Guidelines Alert, 6,* 4-7.

Northern, J. L., & Hayes, D. (1994). Universal screening for infant hearing impairment: Necessary, beneficial and justifiable. *Audiology Today, 6*(3), 10-13.

Paradise, J. L. (1999). Universal newborn hearing screening: Should we leap before we look? *Pediatrics, 103,* 670-672.

Prieve, B., Dalzell, L., Berg, A., Bradley, M., Cacace, A., Campbell, D., DeCristofaro, J., Gravel, J., Greenberg, E., Gross, S., Orlando, M., Pinheiro, J., Regan, J., Spivak, L., & Stevens, F. (2000). The New York State Universal Newborn Hearing Screening Demonstration Project: Out patient outcome measures. *Ear and Hearing, 21,* 104-117.

Robinette, M. S., & White, K. R. (1998). The state of newborn hearing screening. In F. H. Bess (Ed.), *Children with hearing impairment: Contemporary trends* (pp. 45-67). Nashville, TN: Bill Wilkerson Center Press.

Roush, J., Harrison, M., Palsha, S., Davidson, D. (1992). A national survey of educational preparation programs for early intervention specialists. *American Annals of the Deaf, 137,* 425-430.

Russell, L. B. (1994). *Educated guess: Making policy about medical screening tests.* Berkley, CA: University of California Press.

Seewald, R. C. (Ed.) (2000). *A sound foundation through early amplification.* Staefa, Switzerland: Phonak AG.

Shimizu, H., Walters, R. J., Proctor, L. R., Kennedy, D. W., Allen, M. C., & Markowitz, R. K. (1990). Identification of hearing impairment in the neonatal intensive care unit population: Outcome of a five-year project at the Johns Hopkins Hospital. *Seminars in Hearing, 11,* 150-160.

Spivak, L., Dalzell, L., Berg, A., Bradley, M., Cacace, A., Campbell, D., DeCristofaro, J., Gravel, J., Greenberg, E., Gross, S., Orlando, M., Pinheiro, J., Regan, J., Stevens, F., & Prieve, B. (2000). New York State Universal Newborn Hearing Screening Demonstration Project: Inpatient outcome measures. *Ear and Hearing, 21,* 92-103.

Stein, L. K. (1999). Factors influencing the efficacy of universal newborn hearing screening. *Pediatric Clinics of North America, 46,* 95-105.

Stuart, A., Moretz, M., & Yang, E. Y. (2000). An investigation of maternal stress after neonatal hearing screening. *American Journal of Audiology, 9,* 135-141.

Tharpe, A. M. (2000). Service delivery for children with multiple impairments: How are we doing? In R. C. Seewald (Ed.), *A sound foundation through early amplification* (pp. 175-187). Staefa, Switzerland: Phonak AG.

Tharpe, A. M., & Clayton, E. W., (1997). Newborn hearing screening: Issues in legal liability and quality assurance. *American Journal of Audiology, 6*(2), 5-12.

Truman, B. I., & Teutsch, S. M. (1998). Screening in the community. In R. C. Brownson & D. B. Petitti (Eds.), *Applied epidemiology* (pp. 213-247). Oxford University Press: New York.

Van Riper, L. A., & Kileny, P. R. (1999). ABR hearing screening for high-risk infants. *American Journal of Otology, 20,* 516-521.

Vohr, B. R., Carty, L. M., Moore, P. E., & Letourneau, K. (1998). The Rhode Island Hearing Assessment Program: Experience with statewide hearing screening (1993-1996). *Journal of Pediatrics, 133,* 353-357.

White, K. R., & Maxon, A. B. (1995). Universal screening for infant hearing impairment: Simple, beneficial, and presently justified. *International Journal of Pediatric Otorhinolaryngology, 32,* 201-211.