Universal screening for infant hearing impairment: simple, beneficial, and presently justified

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Abstract

In a recent article, Bess and Paradise (Pediatrics 93 (1994) 330–334) rejected the recommendation of the National Institutes of Health that all infants be screened for hearing loss on being released from their birthing hospital. This article responds to their objections with data from the literature and operational newborn hearing screening programs. These data show that universal newborn hearing screening is practicable, effective, cost-efficient, and safe, and concludes that such programs should be implemented without further delay.

Keywords: Otoacoustic emissions; Newborn hearing screening; Hearing loss

1. Introduction

In a recent article in Pediatrics, Bess and Paradise [3] took issue with the March 1993 recommendation of a National Institutes of Health (NIH) Consensus Panel that all infants be screened for hearing impairment within the first 3 months of life. Although Bess and Paradise agreed 'with the desirability of identifying infants with congenital hearing impairment promptly' (p. 330), and they acknowledged that 'early childhood hearing loss can impose burdens on the affected child, on the child’s family, and on society' (p. 331), they nonetheless reached the conclusion that 'universal screening for infant hearing impairment... is not necessarily beneficial, and not presently justified' (p. 330). They went on to say, 'Universal screening is ill-
considered and at this time ill-advised, and that its implementation might result in more harm than good' (p. 330). The crux of their argument is that there is not sufficient evidence that the screening procedures recommended by the NIH Panel are 'valid, reasonably low in cost, and practicable' (p. 331).

On the surface, the arguments presented by Bess and Paradise sound quite logical. However, a review of the available evidence shows that their analysis did not include substantial relevant research data and practical experience. When all of the available data are considered, including some that have become available since their paper, it is clear that the conclusion of the NIH Panel is sensible.

2. Is the newborn hearing procedure recommended by NIH practicable, effective, reasonably low in cost, and harmless?

The NIH Consensus Statement was prepared by an independent 15-member panel representing audiology, otolaryngology, pediatrics, speech and hearing sciences, epidemiology, health care administration, various other child care areas, and the general public. The panel considered presentations and discussions at a 1.5-day Consensus Development Conference on the Early Intervention of Hearing Impairment in Infants and Young Children and subsequently issued a statement recommending that 'universal [hearing] screening be implemented for all infants within the first three months of life. This is most efficiently achieved by screening prior to discharge' [18]. The panel suggested a two-stage screen in which all infants would be screened first with evoked otoacoustic emissions (EOAEs). Those referred from the first stage would be tested with auditory brainstem response (ABR) at the second stage. Those failing the second stage ABR would be considered as screening fails.

In rejecting the NIH recommendation, Bess and Paradise [3] argued that 'the consensus panel's recommendation of universal infant screening falls short of being justified on grounds of practicability, effectiveness, cost, and harm-benefit ratio' (p. 330). Evidence regarding each of these issues is considered below.

2.1. Practicability

In discussing the practicability of the procedure recommended by NIH, Bess and Paradise stated that 'no data on actual use are available to our knowledge' (p. 332). Interestingly, a book edited by Bess [26] describes a newborn hearing screening program almost identical to that recommended by the NIH Panel. That program has been operational at Women and Infants Hospital of Rhode Island (WIHRI) since August 1990 [15,27,28]. The only difference is that the Rhode Island protocol calls for both EOAE and ABR at the second stage screen and uses a 30 dB nHL instead of a 40 dB nHL criterion for the second stage ABR. From July 1993 through June 1994, this universal newborn hearing screening program (the Rhode Island Hearing Assessment Program, or RIHAP) completed the first stage EOAE screen on more than 16 000 infants (approximately 99% of all births in the state), and more than 85% of the infants who failed the first stage screen returned for a second stage screen [14].

Because the initial implementation of RIHAP was supported with federal funds and was done at only one hospital, it is legitimate to ask whether it is reasonable
to expect similar results at other hospitals. After all, Rhode Island is an unusually small state and RIAP was a carefully controlled research project with staff hired specifically to do screening. Perhaps, according to this argument, the success in Rhode Island is attributable to higher levels of dedication and enthusiasm by the research staff than would be present at other hospitals. Although such questions are appropriate, it is clear that the success of universal newborn hearing screening in Rhode Island is not the result of such atypical conditions. First, beginning in July 1992, the universal screening program has been implemented in all other hospitals in the state of Rhode Island, with very similar results. All of this has been done without federal grant money, except for initial training costs. More importantly, there are now universal newborn hearing screening programs using variations of a two-stage TEOAE-based screening protocol very similar to that recommended by NIH in hospitals in at least twelve other states (Hawaii, Utah, Connecticut, Arizona, Arkansas, Texas, Oregon, Illinois, Georgia, Kentucky, North Carolina, and the District of Columbia) [17]. In each case, more than 95% of all live births are being screened and 90% or more of the program costs are from local and state sources.

Bess and Paradise envisioned ‘formidable problems of logistics and cost’ (p. 332) with hospitals who release most of their babies within 48 h and where many of the families live in rural/remote areas. The real life experiences of screening programs using procedures very similar to those recommended by NIH are quite different. Of course, some adjustments have had to be made to accommodate early releases and families that live in rural/remote areas. Nonetheless, even with those babies who are released early, or whose families live in rural/remote areas, the number of babies screened, the referral rates at each stage of the screening process, and the cost per baby screened are quite similar at these hospitals to those reported by RIHAP.

The best evidence concerning the practicability of any human services program is that in the current environment of limited fiscal resources, people are willing to implement and operate the program in the absence of any outside money or mandates. If the type of universal newborn hearing screening recommended by NIH were so difficult to implement and operate, there would not be so many hospitals implementing the procedure.

2.2. Effectiveness

With regard to effectiveness, the primary issue is whether the presence or absence of emissions can differentiate between infants with normal hearing and infants with hearing loss. In discussing whether the NIH-recommended newborn hearing screening program is an effective way to identify hearing loss, Bess and Paradise summarized why sensitivity, specificity, and positive predictive value are important, and then presented hypothetical data about what might happen if the NIH-recommended screening program were implemented. Unfortunately, they did not cite many of the independent studies which have evaluated the effectiveness using the presence or absence of emissions as a means of identifying hearing loss [4,9,19,21,22,25]. Furthermore, they did not cite the results of one newborn hearing screening program which is almost identical to that recommended by NIH [15,26–28]. These studies report consistently that EOAE identifies all of the infants with hearing loss who were
also identified by other currently available screening techniques, as well as some infants with hearing loss who are missed by these techniques.

The best example of a two-stage EOAE/ABR-based universal newborn hearing screening program similar to what was recommended by NIH is the RIHAP program in Rhode Island. RIHAP has now screened over 35,000 infants [15, 26-28]. The exact protocol for that program is reported in detail elsewhere [15, 27, 28]. The goal of RIHAP is to identify infants with unilateral or bilateral sensorineural hearing loss ≥ 30 dB HL. For many of these infants, there is extensive follow-up data (including infants who passed as well as those who failed the newborn screen), and no one has yet reported an infant screened with EOAE who had a congenital sensorineural hearing loss ≥ 30 dB HL and was missed by the EOAE screen.

Some have expressed concern about the high number of false-positives with EOAE screening [3, 18]. This concern was based on the fact that the first report of EOAE screening in Rhode Island reported a relatively high percentage of infants who failed the initial screen [28]. However, since that time, improvements in the EOAE screening software and testing procedures have dramatically reduced the initial fail rate. For example, of the 4,253 infants screened from July 1, 1993 to December 31, 1993 at Women and Infants Hospital of Rhode Island, only 7% failed the initial screen [14]. Other hospitals doing newborn hearing screening with EOAE report similar Stage I referral rates [17]. This is less than the number of infants who exhibit one or more of the high-risk factors listed by the Joint Committee on Infant Hearing (JCIH) [8, 10, 23]. Thus, the number of infants who would need any type of follow-up testing after the first stage of the NIH-recommended protocol is actually less than the number of infants who would need further testing in the protocol recommended by Bess and Paradise (i.e. screening with automated ABR of all infants who exhibit one or more of the JCIH risk factors).

In discussing the effectiveness of the NIH-recommended screening procedure, Bess and Paradise also noted that:

most studies of EOAE testing in newborns have been conducted under 'laboratory' conditions, i.e. by skilled professionals in sound-treated rooms. Use of the test by nonprofessionals in newborn nurseries, as would be required in the interest of practicability for mass screening, has not been evaluated but obviously risks being more problematic. (p. 332)

Although we have not done a tally of all previous studies of EOAE testing in newborns to determine the number which were conducted under 'laboratory' conditions, it is clear that there are some studies where this was not the case. For example, the chapter in Bess' book which describes the initial results of the Rhode Island Hearing Assessment Project [26] states that the screening was done by trained technicians in a relatively quiet room to which acoustic tiles and room dividers had been added.

In making the recommendation that a two-stage EOAE/ABR screening process be used for universal newborn hearing screening, the NIH Panel relied on an extensive body of research presented at the consensus conference. If someone wants to argue that the recommended procedure is not effective, it is reasonable to expect them to show why the research used by the NIH Panel is not trustworthy or cite different research that is more compelling. This has not been done.
2.3. Costs

Whether the newborn hearing screening procedure recommended by the NIH Panel is ‘reasonably low in cost’ is a question which can only be answered adequately by combining values with facts. Unfortunately, Bess and Paradise have based their answer on the wrong facts when they stated (without showing the basis for their calculations) that ‘the direct monetary costs alone of the proposed program, assuming ideal conditions, would approximate to $200 000 000 annually’ (p. 332).

Fortunately, a reasonable estimate of what the NIH-recommended screening program would cost can be made because over 35 000 infants have been screened in Rhode Island using a protocol very similar to that recommended by NIH. The actual expenditures for screening 4253 at Women and Infants Hospital of Rhode Island from July 1, 1993 to December 31, 1993 are shown in Table 1 [14].

These costs are based on hospital records for all expenditures associated with the program based on the hours worked, supplies and materials used, and actual equipment used (amortized over a 5-year period) to do the screening. All costs associated with doing an EOAE test at the first stage, EOAE and ABR at the second stage, scheduling the infants for all follow-up tests, communicating with the parents and pediatricians, managing the program, and keeping track of all the data are included in this summary. The costs of the diagnostic assessment for children referred from the second stage screen are not included, although the costs of keeping track of the family, referring the family to the diagnostic assessment, and following up to make sure they receive a diagnostic assessment, are included. The actual costs for this time period were $26.05 per infant screened. Obviously, these costs will fluctuate some from month to month and will vary somewhat for different localities.

Table 1
Actual costs* of operating a universal newborn hearing screening program for 4253 infants similar to the two-stage EOAE/ABR procedure recommended by NIH

<table>
<thead>
<tr>
<th>Cost</th>
<th>$60 654</th>
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<tbody>
<tr>
<td>Personnel</td>
<td></td>
</tr>
<tr>
<td>Screening technicians (average of 103 h/week)</td>
<td></td>
</tr>
<tr>
<td>Clerical (average of 60 h/week)</td>
<td></td>
</tr>
<tr>
<td>Audiologist (average of 18 h/week)</td>
<td></td>
</tr>
<tr>
<td>Coordinator (average of 20 h/week)</td>
<td></td>
</tr>
<tr>
<td>Fringe benefits (28% of salaries)</td>
<td>$16 983</td>
</tr>
<tr>
<td>Supplies, telephone, postage</td>
<td>$12 006</td>
</tr>
<tr>
<td>Equipment b</td>
<td>$5 575</td>
</tr>
<tr>
<td>Hospital overhead (24% of salaries)</td>
<td>$14 557</td>
</tr>
<tr>
<td>Total costs</td>
<td>$110 775</td>
</tr>
</tbody>
</table>

Cost per infant screened = $110 775 / 4253 = $26.05.

*These costs were calculated based on actual expenditures at Women and Infants Hospital of Rhode Island (WIHRI) between July 1 and December 31, 1993. Because WIHRI is the largest hospital in a statewide system of eight hospitals, staff at WIHRI were also responsible for training and technical assistance to staff at the other hospitals, doing the second stage rescreen and coordinating follow-up and referral for babies born at those hospitals.

Equipment includes 3 EOAE units, 1 ABR unit, 4 personal computers, 2 printers amortized over a 5-year period. Cost includes here are for 6 months only.
Table 2 provides an estimate of what it would cost to implement a nationwide universal newborn hearing screening program using the same protocol. In this calculation, the cost of doing the diagnostic assessment is also included so that we can estimate the actual cost of identifying a child with sensorineural hearing loss. A prevalence rate of 5.95 infants per 1000 is used in the estimate based on the number of infants with sensorineural loss actually identified in the Rhode Island screening program for the first 1850 infants screened [28]. This rate is similar to that reported by others when all children with sensorineural hearing loss are included instead of just those with bilateral severe/profound losses [23]. In the estimate, the rate at which children are referred from each stage of the screening protocol is the same as the rate for the 4253 infants screened at WIHRI from July 1, 1993 to December 31, 1993.

A cost estimate based on such facts, instead of some unknown hypothetical situation, results in an annual cost of nationwide implementation of $109 696 000. Such a nationwide program would identify 23 800 infants with sensorineural hearing impairment at a cost of $4609 per infant. Whether those costs are viewed as being high or low depends on how important you think it is to identify hearing loss at an early age. It is about 1/10th the cost per child identified that we currently pay to identify

Table 2
Estimated costs for nationwide universal newborn hearing screening using procedure similar to NIH-recommended two-stage EOAE/ABR (includes diagnostic testing for infants referred from screening)

<table>
<thead>
<tr>
<th>Assumptions</th>
<th>Costs</th>
</tr>
</thead>
<tbody>
<tr>
<td>• 4 000 000 infants born; all infants screened</td>
<td>$104 200 000</td>
</tr>
<tr>
<td>280 000 referred from 1st stage to 2nd stage screen</td>
<td></td>
</tr>
<tr>
<td>• 10.6 infants/1000 referred from 2 stage screen (42 400 infants)</td>
<td></td>
</tr>
<tr>
<td>2.8/1000 receive both ABR and BOA</td>
<td>2 688 000</td>
</tr>
<tr>
<td>7.8/1000 receive only BOA</td>
<td></td>
</tr>
<tr>
<td>• ABR testing costs $150/infant</td>
<td></td>
</tr>
<tr>
<td>• BOA costs $90/infant</td>
<td></td>
</tr>
<tr>
<td>• 2 stage screening procedures costs $26.05/infant</td>
<td></td>
</tr>
</tbody>
</table>

Total costs $109 696 000

Cost-effectiveness

• Assuming 5.95 infants per 1,000 identified with sensorineural hearing loss

23 800 infants identified
$4609 per infant identified
children with hypothyroidism, phenylketonuria, or sickle cell anemia [7]. Given that every state in the country is willing to pay the costs of screening for these diseases, we think the cost to identify a child with sensorineural hearing loss appears to be quite reasonable.

For the sake of comparison, it is interesting to note what the hearing screening programs currently recommended by the American Speech-Language-Hearing Association (ASHA) [2] would cost. The ASHA protocol is based on identifying infants with one or more of the JCIH risk factors and then testing these infants with ABR. There are reports of several statewide or hearing screening programs that have used the protocol recommended by ASHA [8,11] but detailed cost analyses for such programs have not been done to our knowledge. However, it is possible to use the costs for newborn hearing screening in Rhode Island for clerical/secretarial assistance, audiological support, and coordination to make such an estimate. We estimate it would cost at least $10–$15 per infant to collect the information about risk factors (because this usually requires interviewing parents and doing a medical records review), schedule the at-risk infants for follow-up testing, communicate with parents and pediatricians, manage the program and maintain the necessary data. The percentage of infants exhibiting one or more risk factors are based on the screening programs in Utah and Oregon, and the fail rate for the ABR testing is based on a recent large sample study which tested infants of about the same age as would be required here [6]. The fact that only about half of the infants with congenital sensorineural hearing loss will exhibit at least one of the JCIH risk factors is well documented [5,13,16,20,24].

As can be seen in Table 3, the cost per infant screened is about the same, or a little more expensive when the ASHA recommended protocol is used. However, only half as many infants are identified, which means that the cost per infant identified is substantially higher.

2.4. Harm–benefit ratio

Although they refer to a ‘harm–benefit ratio’ in concluding that the NIH Panel recommendation ‘falls short of being justified’ (p. 330), Bess and Paradise are never clear what they mean by a ‘harm–benefit ratio.’ From the context of their article, they are probably referring to:

the monetary costs of parents’ lost time from work, transportation to care facilities, otherwise unnecessary tests, and unnecessary treatments, and the human and probably more consequential costs of attendant parental anxiety, distraction, and potential misunderstanding, or disturbance of family function, and of unnecessary or harmful procedures or treatments carried out on children.

([3], p. 332)

This description makes the NIH-recommended newborn hearing screening sound like pretty risky business. Fortunately for thousands of infants now being screened with procedures similar to those recommended by the NIH Panel, but unfortunately for those born at hospitals whose administrators are hesitant to implement a newborn hearing screening program because of the alleged dangers involved, existing data from operational programs do not support the existence of such problems [17,28]. For example, even though over 35 000 infants have been screened in Rhode
Table 3
Estimated costs for nationwide hearing screening using high-risk register/ABR procedure currently recommended by ASHA (includes diagnostic testing for infants referred from screening)

**Assumptions**
- 4,000,000 infants born; 9% exhibit risk factors
- All infants with risk factors tested with ABR; 12.5% fail rate; cost = $150 per test
- All infants who fail ABR receive BOA @ $90 per test
- Cost of identifying infants with risk factors, scheduling for ABR and BOA, managing program estimated at $10 or $15 per infant born

<table>
<thead>
<tr>
<th>Costs</th>
<th>$10/Infant</th>
<th>$15/Infant</th>
</tr>
</thead>
<tbody>
<tr>
<td>Identification of infants with risk factors</td>
<td>$40,000,000</td>
<td>$60,000,000</td>
</tr>
<tr>
<td>ABR testing of 360,000 infants with risk factors</td>
<td>54,000,000</td>
<td>54,000,000</td>
</tr>
<tr>
<td>BOA testing of 45,000 infants who fail ABR</td>
<td>4,050,000</td>
<td>4,050,000</td>
</tr>
<tr>
<td>Total costs</td>
<td>98,050,000</td>
<td>118,050,000</td>
</tr>
<tr>
<td>Cost per infant screened</td>
<td>$24.51</td>
<td>$29.51</td>
</tr>
</tbody>
</table>

**Cost-effectiveness**
- Assuming 50% of infants with sensorineural hearing loss exhibit one or more risk factors, 2.725/1000 infants will be identified

If High-risk register costs:

<table>
<thead>
<tr>
<th></th>
<th>$10/Infant</th>
<th>$15/Infant</th>
</tr>
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<tbody>
<tr>
<td>Island</td>
<td></td>
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</table>

11,900 infants identified
$8239 to $9920 per infant identified

Island, there is no evidence of infants or families who have been harmed in the ways listed by Bess and Paradise. Of course, some parents of normal hearing children have been asked to return for a second stage screen or for a more detailed diagnostic assessment (currently, only 7% fail the first stage screen and 10.6 per 1000 are referred from the two-stage screening protocol for diagnostic assessment). Nevertheless, based on data collected from questionnaires completed by over 700 parents whose infants passed the screening protocol, regular contact with all pediatricians throughout the state, and interviews with parents who returned for rescreens, we can find no evidence of undue ‘anxiety, distraction, . . . disturbance of family function, . . . or harmful procedures or treatments carried out on children’ ([3], p. 332).

We are, however, painfully aware of dozens of instances of such problems that have occurred all over the country as a result of late identification of hearing impairment resulting from the absence of the universal newborn hearing screening [8,12,16,20,24]. Thus, the ‘harm–benefit ratio’ of not implementing a universal newborn hearing screening program is better documented than the alleged dangers of implementing such a program.

3. Conclusions

Hardly anyone, including Bess and Paradise, disagrees that it is important to iden-
tify congenital hearing loss as early as possible — preferably during the first year of life. The NIH-recommended procedure for universal newborn screening represents an effective, practicable, and cost-efficient way to accomplish that goal. The procedure is based on dozens of well-documented research studies and supported by actual implementation in an increasing number of hospitals around the country. In Rhode Island alone, more than 35,000 infants have been screened during the last 3 years using a protocol almost identical to that recommended by the NIH Panel. The results of that universal newborn hearing screening program have been clear. Specifically, dozens of babies with congenital hearing loss have been identified and helped during their first year of life, the procedure is practicable to integrate into a busy hospital routine, the costs of identifying each infant with confirmed sensorineural hearing loss are relatively low, and parents and primary care physicians who have participated in the program enthusiastically support universal newborn hearing screening using a protocol almost identical to that recommended by NIH.

Rather than delay the implementation of universal newborn hearing screening longer as we debate what might happen, or what the costs might be, or whether current intervention programs can possibly handle the children who will be identified, it is time to move forward. Data about the costs, practicability, and effectiveness of the NIH-recommended procedure are abundantly available, and hospitals who are already doing universal newborn hearing screening following protocols similar to what was recommended by NIH have demonstrated conclusively that it can be done. Now the challenge is to use the experience of existing programs and the results of ongoing research to improve the techniques and expand the availability and accessibility of universal newborn hearing screening so that every child born in this country with a hearing impairment will be identified in the first months of life.

References


[17] Names of each hospital with more detailed data about the number of births, the number of infants screened and the referral rates from the initial screen for each hospital in each of these states are available from the first author.


