Referral Rates and Cost Efficiency in a Universal Newborn Hearing Screening Program Using Transient Evoked Otoacoustic Emissions

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Abstract
Recently, a National Institutes of Health Consensus Statement recommended that all infants be screened for hearing prior to leaving the birthing hospital using a two-stage screening process based on transient evoked otoacoustic emissions (TEOAEs). Although the value of identifying hearing loss before 1 year of age is widely recognized, the feasibility of universal newborn hearing screening using TEOAE is sometimes questioned because it is presumed that the technique has a high false positive rate and is not cost efficient. This paper presents new data for 4253 infants from an operational universal newborn hearing screening program using a TEOAE procedure that answers those arguments.

Key Words: Early identification of hearing loss, neonatal hearing screening, transient evoked otoacoustic emissions (TEOAEs), universal newborn hearing screening

In 1990, the US Department of Health and Human Services established a goal to lower the average age of identification of hearing loss to less than 12 months of age (US Department of Health and Human Services, 1990). Three years later, a 15-member independent panel representing audiology, otolaryngology, pediatrics, speech and language services, and health care administrators was convened by the National Institutes of Health (NIH) to consider and make recommendations on procedures for early identification of hearing impairment in infants and young children. This panel subsequently issued a consensus statement (NIH, 1993) recommending “that universal screening be implemented for all infants [both NICU and regular-nursery infants] within the first three months of life... [This] is most efficiently achieved by screening prior to discharge” (p. 9) using a two-stage screening procedure based on transient evoked otoacoustic emissions (TEOAEs) and auditory brainstem response (ABR).

Although many have supported the recommendation (Joint Committee on Infant Hearing, 1994; Meister, 1993), others have claimed that the available techniques for universal screening are too costly and result in too many false positives to be used universally (Bess and Paradise, 1994). The purpose of this paper is to report recent data from a universal newborn hearing screening program in Rhode Island that addresses these issues directly. The screening procedures used in Rhode Island are almost identical to those recommended by the 1993 NIH consensus statement.

BACKGROUND

In 1990, the Rhode Island Hearing Assessment Project (RIHAP) was developed to
employ the recently discovered TEOAEs to screen infants. RIHAP’s primary purpose, to determine the validity and cost effectiveness of TEOAE in a large-scale screening program, has been demonstrated and has been reported previously (Maxon et al, 1993; White et al, 1993). The results of that study replicated those observed in the use of TEOAE in neonatal screening with smaller samples (Bonfils et al, 1988a, b; Stevens et al, 1990). Specifically, TEOAE was shown to be effective in identifying infants with bilateral and unilateral sensorineural hearing loss (Maxon et al, 1993; White et al, 1993), thereby affording them significantly earlier access to diagnosis and management, including amplification when appropriate. The results of RIHAP also demonstrated a prevalence of sensorineural hearing loss equal to 5.9/1000 (White et al, 1993) when infants with hearing loss from mild to profound are included. This prevalence was similar to that (5/1000) reported by others (Alberti et al, 1983). To date, almost 30,000 infants have been screened through RIHAP, with the oldest children now over 3 years of age. No infant who passed the screen has subsequently been diagnosed with sensorineural hearing loss through state health department or preschool screening, by pediatricians or audiologists, or through a questionnaire completed by over 700 parents of infants passing the RIHAP screen. Based on all of the information from these sources, the validity of using TEOAE in a two-stage screen to identify hearing loss in infants is supported.

In spite of the increasing acceptance of TEOAE as a valid screening technique, there has been some reluctance to implement it as a universal newborn hearing screening procedure because of a relatively high failure rate (27%) reported for the initial screen (White et al, 1993). For example, even though the NIH consensus statement (1993) recommended TEOAE screening, they noted that “Newborn EOAE testing tends to have more false-positives...[and] over-referral may be a major problem” (p. 12). The concern about the number of false positives associated with this high failure rate was understandable, given the original data published from the RIHAP study. Many have argued that before TEOAE could be considered as a viable universal newborn hearing screening procedure, the failure rate for the initial screen reported by RIHAP (White et al, 1993) would have to be significantly reduced (Bess and Paradise, 1994). Such information is reported in this paper.

### METHOD

This study provides new information about the failure rate of a universal newborn hearing screening procedure that can be expected for infants prior to hospital discharge.

### Subjects

The data were obtained for a sample of 4253 infants (595 special care and 3658 regular nursery) screened at Women and Infants Hospital (the only tertiary care birthing hospital in Rhode Island) between July 1 and December 31, 1993. During that period, all newborn infants cared for in the regular and special care nurseries were screened at a rate of approximately 200 each week.

### Procedures

Figure 1 provides an outline of the two-stage screening protocol used at Women and Infants Hospital in these first 6 months of universal screening. This protocol has been very
successful at Women and Infants and other birthing hospitals in Rhode Island, while hospitals in other geographic regions have established two-stage screening programs with a modification of the RIHAP protocols. The initial TEOAE measure was obtained just prior to hospital discharge, with most infants from regular nurseries tested between 24 and 72 hours of life. Infants discharged before 24 hours of age also were screened just before leaving the hospital. NICU infants were tested when judged to be medically stable by their pediatricians. Infants were brought to a screening room (ambient noise = 60 dBA SPL) with one ABR and three TEOAE screening stations, where they were positioned in an enclosed isolette with an ambient noise level of 45 dBA SPL. TEOAE screenings were conducted by trained technicians using the ILO88 Otodynamic Analyzer (Kemp, 1988) coupled with a neonatal probe and disposable tip. Average actual TEOAE screen time was 3 minutes, 40 seconds per infant. Infants who failed the TEOAE measure returned to the screen site for a second-stage screen in 4 to 6 weeks. TEOAE screening and, for infants who failed the second-stage TEOAE, an ABR screening using a Biologic Traveler Express were conducted by the RIHAP screeners. All second-stage screen failures were referred for diagnostic audiologic evaluation. A summary of procedures can be found in the Appendix while a detailed description of all procedures can be found in Johnson et al (1993), Maxon et al (1993), and White et al (1993).

RESULTS AND DISCUSSION

Although information about the validity of this technique would be based on the hearing status of infants screened, such results are not available for this sample. The focus of this paper is the referral and cost efficiency of a universal newborn hearing screening program using TEOAE.

Referral Rates

Figure 2 displays the pass and referral rates for the two stages of the RIHAP screening protocol. At both stages, any infant who did not have a robust emission (see Appendix for definition) was referred for further screening or evaluation. The initial TEOAE pass rate of 93 percent resulted in the immediate discharge of 3945 infants from the program. The high pass rate (NICU = 91%, regular nursery = 94%) is a significant improvement from the early stages of RIHAP. The improvement can be attributed to modifications in the ILO88 hardware and software, modifications in probe fit techniques, and the experience of the screeners (see Appendix for more detailed information). Of the 308 infants who failed the first-stage screen, 249 (81%) returned at 4 weeks for the second-stage screen, and 82 percent of these passed the TEOAE measures. The 45 infants who failed the second-stage TEOAE screen were screened with ABR and were referred for either a behavioral audiologic evaluation if they failed the ABR at 30 dB nHL or a diagnostic ABR evaluation if they failed the ABR at greater than or equal to 60 dB nHL. Of the 4253 infants screened in the universal hearing screening program during this period, only 45 (1.06%) did not pass the two-stage screen procedure and were referred for a diagnostic evaluation.

Another way of expressing these data is the rate of referral for a diagnostic audiologic evaluation, which was 10.6/1000, with only 2.8/1000 of these referred for the typically more expensive ABR procedure. Since the youngest of these infants are still less than 5 months old at the time of this writing, complete audiologic diagnostic evaluation data are not yet available. However, if Women and Infants Hospital's previously established prevalence of sensorineural hearing loss of 5.9 infants per 1000 (see White et al, 1993) is true for this sample, we can estimate that of the 10.6 infants per 1000 referred,
5.9 per 1000 would have a hearing loss and only 4.7 per 1000 would be false positives. In other words, only less than one half of 1 percent of all of the infants screened would have to return for what some have termed “unnecessary” audiological evaluations. This is a small price to pay for the increased efficiency of this universal newborn hearing screening program, particularly when compared to the previously recommended use of a high-risk registry that missed at least half of all infants with congenital hearing loss (Elssman et al, 1987; Mauk et al, 1991).

Cost Effectiveness

As already mentioned, another frequently voiced objection to universal newborn hearing screening using TEOAE is that it will cost too much. Such objections are often accompanied by hypothetical estimates of the costs of operating such a program, the number of infants who would fail the screen, and the number of hearing-impaired infants to be identified. Instead of using hypothetical data, it is instructive to consider the actual costs of operating the universal newborn hearing screening program in Rhode Island during the period described above. In RIHAP, the costs are not separated for the regular and special care nurseries since it is important to determine the cost efficiency of a program that screens all infants. Since different hospitals will have special care and regular nursery populations that differ from those at Women and Infants (14% NICU, 86% regular nursery), these costs will have to be adjusted on an individual hospital basis. The actual costs of screening 4253 infants at Women and Infants Hospital amounted to $110,775, as shown in Table 1. This cost includes the screening of all babies who were cared for in either the NICU or regular care nursery during the 6-month period. These costs included personnel and fringe benefits for the initial and second stages of the screen, including the TEOAE and ABR measures, scheduling, tracking, and referral for the diagnostic audiological evaluation. As indicated earlier, RIHAP’s responsibilities extend to include referral for diagnostic evaluation and tracking to ensure that such an evaluation has taken place. Personnel covered by this total sum include the screeners, clerical staff, secretarial staff, half-time coordinator, and half-time audiologist.

The costs of the equipment shown in Table 1 are those for the initial outlay when infants are screened at a rate of 10,000 per year. The computers are used to run the TEOAE equipment, generate the paperwork (e.g., printouts, letters) and track testing and results. The initial outlay of $65,750 associated with all equipment is amortized (at 20%) over 5 years and is figured into the annual costs as $13,150. Including supplies and hospital overhead, the actual cost of running the program during this 6-month period was $110,775, resulting in a cost per infant screened of $26.05. Based on a prevalence rate of approximately 5.9 infants per 1000 (as demonstrated by White et al [1993] and Alberti et al [1983]), the cost of actually identifying an infant was therefore equal to $4378. Although specific costs will vary according to geographic location, these figures reflect the actual costs of operating the Women and Infants' universal newborn hearing screening program using a protocol very similar to that recommended in the NIH consensus statement.

A related opposition to universal screening is that the cost of audiological evaluations for infants referred from the screening program will be too high. The information presented above (also see Table 1) indicates that for every 1000 infants screened, 2.8 will be referred for a diagnostic ABR evaluation. At an average cost of $150.00 for each evaluation, the cost of diagnostic ABR evaluations for a program that screens 10,000 infants per year is only $4200.

Table 1  Actual Costs of Operating a Universal Newborn Hearing Screening Program at Women and Infants Hospital of Rhode Island*

<table>
<thead>
<tr>
<th>Personnel</th>
<th>$60,654</th>
</tr>
</thead>
<tbody>
<tr>
<td>Screeners (average 103 hr/wk)</td>
<td></td>
</tr>
<tr>
<td>Clerical (average 60 hr/wk)</td>
<td></td>
</tr>
<tr>
<td>Audiologist (average 18 hr/wk)</td>
<td></td>
</tr>
<tr>
<td>Coordinator (average 20 hr/wk)</td>
<td></td>
</tr>
<tr>
<td>Fringe benefits (@ 28% of salaries)</td>
<td>$16,983</td>
</tr>
<tr>
<td>Supplies</td>
<td>$12,006</td>
</tr>
<tr>
<td>Equipment (three TEOAE units, one ABR unit, four computers, two printers, amortized over 5 y)</td>
<td>$6,575</td>
</tr>
<tr>
<td>Overhead (29% of salaries)</td>
<td>$14,557</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td><strong>$110,775</strong></td>
</tr>
</tbody>
</table>

*Time period is July 1, 1993 to December 31, 1993.

Note: 4253 infants screened at total cost of $110,775 = $26.05 per infant screened, assuming prevalence of 5.9/1000 = $4378 per infant identified.

1 Although not specific to this sample, the prevalence rate is based on a sample of over 3000 infants whose hearing status is established. There is no reason to believe that the present sample is significantly different from that previously studied.
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CONCLUSIONS

Even though everyone agrees that it is important to identify hearing impairment as early as possible, hypothetical data are frequently used to argue that a two-stage universal newborn hearing screening program using TEOAE and ABR (such as that recommended by the NIH consensus statement) is too expensive and results in too many false positives that will result in increased parental anxiety, inconvenience, and unnecessary audiologic evaluations. The actual data from a large-scale operational universal newborn hearing screening program should put those arguments to rest. In this program, only about 1 percent of infants screened in a two-stage method fail and are referred for audiologic evaluations. The cost of such a two-stage newborn hearing screen is only about $26.00 per infant screened, which means that it costs approximately $4378 to identify a child with sensorineural hearing loss.

Universal newborn hearing screening is not only feasible, but the relatively low associated costs make it very practical. We believe it is now time to move forward in implementing universal newborn hearing screening.

REFERENCES


APPENDIX

Screening Protocols

TEOAE

Measurements were made by the RIHAP screeners (trained technicians) using the Otodynamics ILO88. The screeners strictly adhered to the TEOAE screening protocol given below to ensure that an adequate test had been completed.

Infants were screened using the Quickscreen option (see description given below), which employs a broad-band click stimulus (80 microsecond, 80 dB pe SPL-40 dB spectrum level, 500-5000 Hz, 80/sec). The peak stimulus level was considered acceptable when it was between 71 and 83 dB pe SPL. When necessary, screeners adjusted the gain to achieve an appropriate level. Probe stability was maintained (at least 80%) during data collection by monitoring the light in the stimulus box.

At least 50 low-noise samples were collected for each ear. If the signal-to-noise ratio was not adequate (see below for description), the screener continued to collect for up to 125 samples.

ABR

Measurements were made by the RIHAP screeners using the Biologic Traveler Express in the screen mode. The screeners strictly adhered to the ABR screening protocol given below to ensure that an adequate test had been completed.

Disposable electrodes (active, reference, ground) were placed after skin preparation. After achieving the appropriate impedance, testing was initiated in either ear at 30 dB nHL using a click stimulus (100- to 3000-Hz filter) with a repetition rate of 20 clicks/second. A minimum of 1024 trial averages was collected twice for each level. Higher screen levels (60 dB and 85 dB) were used when a clear response was not measured at 30 dB.

Scoring Protocols

1. Scorable TEOAE results were classified as pass or refer according to the following scoring protocol:
   Pass: An emission is present, that is, a signal-to-noise ratio of +6 dB or greater over half of each test frequency band (1000–2000 Hz, 2000–3000 Hz, 3000–4000 Hz), when the test conditions were met.
   Refer: An emission is present in one or two of the test frequency bands, but not all three when the test conditions were met; or no emission is present in any of the test frequency bands when all of the test conditions were met.

2. Scorable ABR results were classified as pass or refer according to the following protocol:
   Pass: Wave V is present at 30 dB nHL within 1 standard deviation of the mean latency that is appropriate for an infant's gestational age (as provided by the equipment manufacturer).
   Refer: Wave V is absent or has an abnormally long latency for an infant's gestational age (as provided by the equipment manufacturer) at any test intensity.

Referral Protocols

1. First-stage screen. Weekly, the results of the TEOAE initial screens were judged for adequacy of test and scored by the RIHAP audiologist, using the protocols shown above. Infants who passed the screen in both ears were discharged from the program. Those who did not were scheduled to return in 4 to 6 weeks for the second-stage screen.

2. Second-stage screen. Weekly, the results of the TEOAE and any ABR second-stage screens were judged for adequacy of test and scored by the RIHAP audiologist. Infants who passed the TEOAE in both ears were discharged from the program. Those who failed TEOAE only or TEOAE and ABR at 30 dB nHL (but passed at higher intensity levels) were referred for a diagnostic behavioral audiologic evaluation conducted at 6-months post-initial screen by a certified clinical audiologist at another site.

   Infants who failed the ABR >_ 60 dB nHL were referred for a diagnostic ABR conducted within 1 month of the second-stage screen by a certified clinical audiologist at another site.

3. All infants who demonstrated abnormal diagnostic ABRs were referred for a behavioral audiologic evaluation to obtain frequency-specific, individual ear information as quickly as possible. These referral procedures were developed in an attempt to ensure the earliest possible diagnosis and management of the hearing loss.
Factors that Maintain a Low Referral Rate

In RIHAP, the referral rate went from an early high of 27 percent to the present 7 percent rate. A primary factor in attaining and maintaining a low first-stage-screen referral rate is the experience of screeners. When screeners are familiar with the equipment (hardware and software) and the protocols, they are readily able to implement procedures that help facilitate reducing the false positive rate. The following is a summary of the procedural modifications that were developed in RIHAP since its inception and are employed by the screeners:

1. Reducing internal and external noise.
   a. When an infant is noisy (crying, hungry, sucking), it may be difficult to extract the response (emission) from the noise. Therefore, the screeners bring infants to the initial screen when it is likely to achieve an appropriate state (i.e., they are fed and ready for sleep). Then infants can be swaddled and readily settled after transfer to the test isoket (Vohr et al, 1993).
   b. The fit of the probe in the ear canal can affect the interference from external noise. Screeners use the probe tip that gives the tightest fit. They monitor the stimulus and can quickly make adjustments (e.g., change the gain) to compensate for infant movement.
   c. All infants are screened using the Quickscreen option on the ILO88. In Quickscreen, the response window is 12.5 msec and the stimuli are presented at a more rapid rate. The result of reducing the time window is the reduction of low-frequency information in the TEOAE response, including the noise. Therefore, when an emission is present, it is more easily viewed in Quickscreen than in the original ILO88 test that uses a 20-msec window.
   d. Infants who are noisy may also be tested using the low-cut filter option. This will further reduce the low-frequency information in the response and make an existing emission more visible.

2. Good condition of the external ear canal.
   a. With neonates, debris (vernix caseosa, cerumen) in the external auditory meatus can directly block the probe (housing the microphone and signal transmitter), making accurate measurement impossible. Blocking the input stimulus results in a reduced or absent emission and a false positive outcome. Removal of debris from the ear canal is more likely (91%) to yield a normal TEOAE response than no cleaning (76%) (Chang et al, 1993). A simple procedure for removing debris from the external ear canal is removing the probe, cleaning the tip, and refitting it. RIHAP screeners routinely carry out this procedure when an abnormal emission is obtained.
   b. In the early stages of RIHAP, infants were discharged from the hospital between 48 and 72 hours postdelivery unless there was a complication or a delivery by C-section. The current trend is toward earlier discharge, which increases the likelihood that birthing debris will be present in the external ear canal. Therefore, the screeners employ the probe refit technique regularly. They also screen the infant as close to discharge as possible. For infants who are screened very early in life, a second test is conducted at discharge.