A Cost-effectiveness Analysis of Newborn Hearing Screening Strategies

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Context: Congenital hearing loss affects between 1 and 3 out of every 1000 children. Screening of all neonates has been made possible by the development of portable automated devices. Universal screening is a 2-stage screening process using automated transient-evoked otoacoustic emissions, followed when indicated by automated auditory brain response testing. Targeted screening reserves the 2-stage screening process for those infants at risk for congenital hearing loss.

Objective: To compare the expected costs and benefits of targeted screening with universal screening for the detection of significant bilateral congenital hearing loss.

Design: Cost-effectiveness analysis from the health care system perspective, including costs directly related to screening and initial follow-up evaluation.

Main Outcome Measures: Number of cases identified, number of false positives, and cost per case.

Results: For every 100 000 newborns screened, universal screening detects 86 of 110 cases of congenital hearing loss, at a cost of $11 650 per case identified. Targeted screening identifies 51 of 110 cases, at $3120 per case identified. Universal screening produces 320 false-positive results, 304 more than targeted screening. Switching to universal screening from targeted screening would cost an additional $23 930 for each extra case detected.

Conclusions: Universal screening detects more cases of congenital hearing loss, at the expense of both greater cost and more false-positive screening results. Little is known about the negative impact of false-positive screening and about the benefits of early intervention for congenital hearing loss. Those who advocate adoption of universal screening should be aware not only of the direct costs of universal screening, but of the indirect costs and strategies to increase the benefits of screening.


CONGENITAL hearing loss affects between 1 and 3 out of every 1000 children born in the United States. With the development of new screening technologies, and since early intervention may improve outcome, a national goal has been set to reduce the mean age at diagnosis from the current 2.9 years to no more than 12 months. However, using universal newborn hearing screening to achieve this goal has generated significant controversy.

In 1993, a consensus development panel convened by the National Institutes of Health endorsed universal hearing screening for infants within the first 3 months of life, as did the American Academy of Pediatrics. More than 20 states have adopted legislation supporting universal newborn hearing screening and federal legislation (Newborn and Infant Hearing Screening and Intervention Act of 1999, HR1193 §956 [1999]) has been introduced to encourage universal screening.

However, the performance of the screening tests and the role of early intervention are unclear. The US Preventive Services Task Force has found that there is insufficient evidence to recommend for or against routine screening of asymptomatic infants.

Before analyzing the potential benefits and costs of newborn hearing screening, it is important to review the evidence regarding both the effectiveness of intervention and the performance of the screening strategies.

HEARING LOSS AND EFFECTIVENESS OF INTERVENTION

The goal of screening is to detect significant bilateral loss in the frequency region...
To evaluate newborn hearing screening, a comparison was made between the costs and benefits for both universal newborn hearing screening and targeted, risk-based screening. Cost analysis was conducted from the health care system perspective. The short-term costs, including the costs of screening and follow-up testing, were included. The long-term costs, including the costs of treatment and any potential savings from the early initiation of treatment, were not included, since these costs are unknown. Indirect costs, such as parental wages lost, transportation costs, and child-care costs incurred because of the screening process, were not included. Benefits for these screening programs were assessed by the number of cases properly identified.

DECISION ANALYSIS MODEL

This model of newborn hearing screening is a 2-stage process, similar to the one recommended by a 1993 National Institutes of Health consensus development panel (Figure). Universal screening assumes that all newborns will be screened with an automated TEOAE device. Infants with positive results would then be screened with an automated ABR device. Infants with positive results on this second screen would then be referred for diagnostic ABR testing. In targeted screening, only newborns at high risk for hearing impairment, defined as spending time in a neonatal intensive care unit, having a family history of impaired hearing, or the presence of a craniofacial abnormality, would receive the 2-stage screening process.

This analysis was based on the hypothetical experience of a cohort of newborn children in the United States. We assumed that all newborns would be screened and receive all necessary follow-up testing. This assumption would increase the number of cases detected and bias the model toward the effectiveness of screening.

All modeling was done with DATA 3.0 (TreeAge Software Inc., Williamstown, Mass). To test the robustness of the model and to identify important areas of uncertainty, univariate sensitivity analyses were performed on all variables, and 2-way sensitivity analyses were performed on selected variables. All calculated costs were rounded to the nearest $10.

MODEL PARAMETERS

We estimated probabilities (Table 1) and costs (Table 2) by reviewing the literature. We searched MEDLINE using the term hearing tests and restricted this set to articles written in English. The baseline estimate of the prevalence of congenital hearing loss was from a population-based surveillance program for serious hearing loss (>40 dB hearing loss bilaterally). The specificity of risk-based screening for congenital hearing loss was not available. We applied the Bayes theorem to calculate the specificity from the prevalence of hearing loss and the positive predictive value of risk assessment.

Cost data are difficult to obtain and standardize. For example, the cost of using an automatic screening device depends on the time over which the device will be depreciated and the estimate of the number of children who will be screened. We therefore evaluated costs over a large range. Baseline estimates of cost were based on reports in the literature and discussions with experts. The cost of maintaining a registry of high-risk patients to arrange for further screening has been reported to be between $10 and $15. With the development of more portable and easier-to-use screening equipment, we estimate that this cost has fallen substantially.

Important for speech recognition. Often, the definition of significant hearing loss is at least 30 to 40 dB in both ears. Screening programs attempt to detect congenital hearing loss (hearing losses acquired either prenatally or perinatally). Acquired hearing losses, such as progressive hearing impairment, late-onset impairment, or other postnatally acquired causes, such as meningitis, cannot be detected.

The importance of the detection of unilateral hearing loss is unclear. Unilateral hearing loss may affect language development and educational achievement. Since little is known of the natural history and the benefits of early intervention for unilateral hearing loss, screening programs focus on bilateral hearing loss.

There are no controlled trials evaluating the impact of early intervention in significant bilateral congenital hearing loss on long-term quality of life. Such a study would be difficult, because congenital hearing loss is rare and withholding treatment would not be appropriate. Reduced auditory stimulation for 3 to 6 months may delay the normal course of language acquisition. This is corroborated by animal studies showing that early auditory deprivation interferes with the development of neural structures necessary for hearing. There is evidence from case-control studies that early intervention improves linguistic skills. Decreasing the age of diagnosis may lead to improved treatment strategies.

Early detection has benefits beyond the development of new treatment strategies. For example, early detection gives families more time to investigate the various treatment options. Treatment options can include hearing aids, cochlear implants, and alternative communication methods. Each of these options has unique benefits and potential harms.

Hearing aids and cochlear implants improve hearing. However, both types of devices need special care to be used properly. Furthermore, cochlear implants need to be surgically placed and equipment failure can be as high as 11%. These risks must be weighed against the potential for early implantation to result in important improvements in prelingually deaf children.

American Sign Language (ASL), the language of the deaf community, has a grammar that is distinct from English. Early diagnosis may be helpful in allowing family members more time to learn ASL. Since the structure of ASL is unique, the child will have difficulty in communicating with people who do not know ASL if a bilingual-bicultural approach is not taken. Other oral methods are available that augment residual hearing or assist with lipreading. However, a child who learns these
oral methods and not ASL may be isolated from deaf culture.15,16

SCREENING STRATEGIES

Traditionally, hearing screening has been targeted to high-risk populations, such as those infants who have been in the neonatal intensive care unit, those with a family history of hearing impairment, or those with craniofacial abnormalities.9 These at-risk populations represent less than 10% of the birth cohort17 and can potentially identify between 50% and 59% of those with congenital hearing loss.6,9 In practice, however, not all at-risk children are identified.9

Until recently, universal newborn hearing screening has been limited by available screening technology.18 Two new strategies have become available. Transient evoked otoacoustic emission (TEOAE) measures sounds that are generated by the cochlea in response to acoustic stimulation.18 This reflects the integrity of the outer, middle, and inner ear.9 Auditory brainstem response (ABR) measures electroencephalographic waveforms in response to clicks.18 As with TEOAE, ABR assesses the lower auditory pathways.9

Both ABR and TEOAE have high reported sensitivity and specificity.16,17,18 However, since the prevalence of bilateral congenital hearing impairment is low, there are many false-positive screening results. False positives dampen enthusiasm for a screening program, increase costs, and may harm families because of unnecessary worry about the health of their child. To decrease the overreferral rate, some programs have rescreened children with either the same or a different screening test.6,20 However, this decreases the number of cases detected.

The baseline results of our analysis are listed in Table 3. Universal screening detects 86 cases of congenital deafness out of 110 for every 100 000 children screened, representing a 40% improvement over targeted screening. However, this improvement in case detection is associated with 320 false-positive referrals, a 19-fold increase over targeted screening. The positive predictive value is 21% for universal screening and 76% for targeted screening. Compared with targeted screening, universal screening is associated with a 530% increase in total costs and a 273% increase in the cost per case detected. Assuming that there are approximately 4 million births per year in the United States,25 targeted screening would cost $6.4 million compared with $40 million for universal screening.

### Table 1. Probability Estimates*

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Baseline Estimate (Range), %</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>Prevalence of hearing loss</td>
<td>0.11 (0.10-0.59)</td>
<td>1, 2, 9, 20</td>
</tr>
<tr>
<td>Risk screening</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sensitivity</td>
<td>59 (50-64)</td>
<td>5, 6, 9, 17, 18</td>
</tr>
<tr>
<td>Specificity</td>
<td>95 (91-99)</td>
<td>5, 6, 9, 17, 18</td>
</tr>
<tr>
<td>Automated TEOAE</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sensitivity</td>
<td>80 (66-100)</td>
<td>20, 21</td>
</tr>
<tr>
<td>Specificity</td>
<td>92 (91-93)</td>
<td>21</td>
</tr>
<tr>
<td>Automated ABR</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sensitivity</td>
<td>98 (80-100)</td>
<td>1, 19</td>
</tr>
<tr>
<td>Specificity</td>
<td>96 (86-98)</td>
<td>1, 19</td>
</tr>
</tbody>
</table>

*TEOAE indicates transient evoked otoacoustic emission; ABR, auditory brainstem response.

### Table 2. Cost Estimates*

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Baseline Estimate (Range), $</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>Risk screening</td>
<td>1.00 (0.50-15.00)</td>
<td>5, 18, AE</td>
</tr>
<tr>
<td>Automated TEOAE</td>
<td>7.42 (5.00-40.00)</td>
<td>22, AE</td>
</tr>
<tr>
<td>Automated ABR</td>
<td>25.00 (15.00-40.00)</td>
<td>23, 24</td>
</tr>
<tr>
<td>Diagnostic ABR</td>
<td>150.00 (100.00-200.00)</td>
<td>20, AE</td>
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*TEOAE indicates transient evoked otoacoustic emission; ABR, auditory brainstem response; and AE, authors’ estimate.

### Table 3. Baseline Results for 100 000 Newborns Screened*

<table>
<thead>
<tr>
<th>Strategy</th>
<th>No. of Cases Detected</th>
<th>No. of False Positives</th>
<th>Total Cost, $</th>
<th>Cost per Case Detected, $</th>
</tr>
</thead>
<tbody>
<tr>
<td>Targeted screening</td>
<td>51</td>
<td>16</td>
<td>158 860</td>
<td>3120</td>
</tr>
<tr>
<td>Universal screening</td>
<td>86</td>
<td>32</td>
<td>1 004 860</td>
<td>11 650</td>
</tr>
</tbody>
</table>

*All costs are rounded to the nearest $10.
ing. However, 1400 more cases of congenital hearing loss would be detected.

INCREMENTAL COST-EFFECTIVENESS

Moving from risk-based to universal screening would cost an additional $23,930 for each extra case detected.

SENSITIVITY ANALYSIS

Sensitivity analysis identifies important areas of uncertainty. Univariate sensitivity analyses on all variables and bivariate sensitivity analyses on sensitivity and specificity for each screening test were performed.

Probabilities

Over all ranges of probability estimates, universal hearing screening detects more cases than targeted screening at a greater cost.

Costs

Only the cost of risk-screening assessment affects the relationship between universal and targeted screening. If the cost of screening infants for high-risk factors were more than $5.34 per infant, the cost per case detected by universal screening would be less than by targeted screening. At the extreme limit of our analysis, at which the cost of risk screening is $15 per infant, the cost per case identified by targeted screening would be $18,990 more than the cost per case identified by universal screening.

This model suggests that universal newborn screening for congenital hearing loss would detect 40% more cases than targeted, risk-based screening. However, the probability that a newborn referred for further evaluation by a universal screening program has congenital hearing loss is 21%, compared with 76% for a newborn referred by a targeted screening program. Universal screening would cost the health care system nearly $24,000 extra in the short term for each additional case detected compared with targeted screening.

The results of our analysis are similar to reported experience with universal screening. Single-stage screening with automated ABR has been reported to cost $17,500 per case of significant bilateral hearing loss detected.10 In our model, the probability that a newborn would be referred for diagnostic testing was 4%.10 It is difficult to compare our results with those of other published studies, since the rate of follow-up for diagnostic examination is less than 70%.27,28

Cost-effectiveness analyses that take into account all costs to society and have standardized measures of benefit, such as the expected number of quality-adjusted life years, can help policy makers allocate scarce resources by allowing direct comparisons of different programs.29 This analysis only includes short-term costs to the health care system. The benefit of these hearing screening programs was measured by the number of cases identified.

Unfortunately, few cost data are available. Long-term costs, such as therapy for those with congenital hearing loss, were not included. These costs are difficult to estimate, since treatment plans can vary widely. Also not included were potential cost savings that might result from early intervention, such as improved productivity because of better language skills. Such savings might substantially reduce the overall cost of a screening program. Unfortunately, these benefits are not known.

Quantifying the long-term benefits of early detection of congenital hearing loss is problematic since the effects of early intervention are unclear. To use a measure of outcome such as the number of expected quality-adjusted life years, the possible health outcomes must be well defined and consistent.30 However, even if these outcomes were well established, it is unclear what population should be studied to evaluate the relative preferences for these health outcomes. Parents of newborns may have little personal experience with deafness but may have strong opinions about the value of being reassured by a true-negative test result or concerned about a false-positive result. Similarly, health care providers may have strong opinions about the harm of congenital deafness that is not detected early. Finally, adults with congenital deafness may feel quite differently than health care providers about the negative impact of deafness on their lives; furthermore, they may feel that the judgments of others on their quality of life are demeaning.30

Many states are now moving forward in adopting universal hearing screening, despite questions regarding the costs to society and the long-term benefits of early detection. This approach is not unreasonable, given the strong belief in the utility of early detection. In fact, this strategy has precedents. In the early 1960s, there was a drive to screen all newborns for phenylketonuria before the benefits of dietary therapy were conclusively established.31

Even without full knowledge of the costs and benefits of newborn hearing screening, there are strategies that can maximize the cost-effectiveness of a universal screening program. There is a learning curve for effective screening.32 A universal screening program should accurately test all infants and track all who test positive to ensure proper follow-up and, when needed, treatment.27,32 Those establishing new screening programs could learn from the experience of those who have already begun universal screening. Furthermore, a network of screening programs could be established to investigate different screening techniques and follow-up methods.

The benefits of early intervention need to be maximized. Since congenital hearing loss is rare, prospective analyses of treatment strategies are difficult. However, now that an increasing number of communities are adopting universal screening, more cases will be identified. These children should be enrolled in clinical trials to answer questions regarding the efficacy of treatment and to better define optimal treatment plans. Such a strategy has been successful in the evaluation of treatment for other rare diseases, such as childhood cancer.33 Parents are faced
with making difficult treatment decisions, including which method of alternative communication will be used, if needed. Collecting prospective data will allow families to be better informed.

Finally, the harm of false-positive screening needs to be minimized. Universal screening results in substantially more false positives than targeted screening. It has been suggested that the harm of a false-positive hearing screening result in a newborn is minimal. However, significant psychological distress has been reported with other false-positive screening results, such as maternal serum α-fetoprotein level for Down syndrome or mammography for breast cancer.

Little is known about parent preferences, including their feelings regarding false-positive screening results. Under universal screening, a positive test result is nearly 80% likely to be false. Counseling and information may not decrease the anxiety associated with false-positive test results. Little is known about the best time to discuss screening or what information is best to provide. Obtaining informed consent formally may ensure that parents understand the potential risks of screening. However, such a strategy could increase the costs and decrease the effectiveness of a screening program if the process is difficult to administer or causes undue parental concern, leading to many screening refusals. As with the evaluation of early intervention, the effects of providing counseling and information and the effects of requiring informed consent can and should be studied.

Universal newborn hearing screening has been a contentious issue. The benefits of early detection must be weighed against the negative impact of false-positive results and the costs of screening. Unfortunately, little is known about these factors. Many communities are now adopting universal newborn hearing screening programs. To maximize the cost-effectiveness of these programs, attempts should be made to improve the accuracy of screening, to improve the benefits of early intervention, and to decrease the harm of false-positive screening. These factors should be prospectively studied and continuously evaluated by those involved with these screening programs.

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