Projected cost-effectiveness of statewide universal newborn hearing screening
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Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

Health technology
The use of universal newborn hearing screening (UNHS; i.e. the screening of all newborn babies) to identify deaf infants. Deaf infants were defined as those with moderate to profound bilateral hearing loss (i.e. at least 40 decibels). A 2-step screening procedure was used. First, an automated transient evoked otoacoustic emissions (TEOAE) test, followed by an automated auditory brainstem response (AABR) test for infants who did not pass the TEOAE.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population comprised newborn babies suspected of having bilateral moderate to profound deafness (i.e. at least 40 decibels).

Setting
The setting appears to have been a hospital. The economic study was performed in the USA.

Dates to which data relate
The effectiveness data were collected from studies published between 1996 and 2000, and from one expert panel for which the date was not reported. The dates to which the cost data related were not given. The price year was 2001.

Source of effectiveness data
The estimates of effectiveness were derived from a review of published studies and estimates made by an expert panel.

Modelling
A decision tree was used in the study to estimate the effectiveness and costs of the proposed interventions. A hypothetical cohort of 80,000 infants was used to model the effectiveness results and costs. It was assumed that there would be proportions of children who would have false-negative test results, would not be screened under SS, and who would fail the hearing screening but would not have immediate follow-up.

Two outcomes were modelled. First, the probability that an infant with moderate to profound deafness would be identified by 6 months of age and received early intervention by 12 months of age (the authors stated that these are the age targets in most published studies about UNHS). Second, the probability that a non-deaf infant would reach audiologic evaluation and be incorrectly diagnosed and treated as deaf.
**Outcomes assessed in the review**

The outcomes assessed in the review were:

- the proportion of newborns at high risk (HR);
- the proportion of HR and low-risk (LR) deaf newborns;
- TEOAE sensitivity and specificity;
- AABR sensitivity and specificity;
- the rate of follow-up with diagnostic audiologic evaluation after a positive screen;
- the proportion of infants whose deafness was diagnosed by 6 months, the proportion of them who were subject to intervention by 12 months, and the proportion of non-deaf infants falsely labelled as deaf with newborn hearing screening;
- the proportion of HR and LR infants whose deafness was diagnosed by 6 months, and the proportion of them subject to intervention at 12 months without newborn hearing screening.

**Study designs and other criteria for inclusion in the review**

Not reported, although 2 controlled trials were identified from the references.

**Sources searched to identify primary studies**

Not reported.

**Criteria used to ensure the validity of primary studies**

Not reported.

**Methods used to judge relevance and validity, and for extracting data**

Not reported.

**Number of primary studies included**

At least 11 published studies were included in the review as sources of the effectiveness evidence.

**Methods of combining primary studies**

Not reported.

**Investigation of differences between primary studies**

Not reported.

**Results of the review**

The proportion (range tested in sensitivity analysis) of newborns at HR was 0.13 (0.08 - 0.15).

The proportion of HR deaf newborns was 0.0083 (0.001 - 0.05) and the proportion of LR deaf newborns was 0.0006 (0.00005 - 0.0013).

The sensitivity of TEOAE was 0.95 (0.9 - 1) and the specificity was 0.85 (0.8 - 1).
The sensitivity of AABR was 0.95 (0.9 - 1) and the specificity was 0.90 (0.8 - 1).

The rate of follow-up with diagnostic audiologic evaluation after positive screen was 0.77 (0.5 - 1).

When newborn hearing screening was performed, the proportion of infants whose deafness was diagnosed by 6 months was 0.80. Of these, the proportion subject to intervention by 12 months was 0.70. The proportion of non-deaf infants falsely labelled as deaf was 0.00008.

When newborn hearing screening was not performed, the proportions of infants whose deafness was diagnosed by 6 months were 0.25 (0.1 - 0.25) for HR infants and 0.20 (0.1 - 0.2) for LR infants. The proportion of them that were subject to intervention by 12 months was 0.30.

These effectiveness estimators were included in the decision tree as key parameters in order to model the health benefits.

Methods used to derive estimates of effectiveness
A panel of 4 experts on hearing screening and language development in the deaf, and a modified Delphi process, were used to derive estimates of effectiveness. In particular, the proportion of LR and HR infants with a normal language quotient (i.e. language quotient greater than 80) after early (before 12 months) and late (after 12 months) interventions.

Estimates of effectiveness and key assumptions
The proportions of LR infants with a normal language quotient were 0.70 (0.4 - 1) after early intervention and 0.40 (0 - 0.7) after late intervention. The proportions of HR infants with a normal language quotient were 0.50 (0.28 - 1) after early intervention and 0.28 (0 - 0.5) after late intervention.

Measure of benefits used in the economic analysis
The summary measures of benefit used were:

the number of infants, in each one of the strategies, whose deafness would be diagnosed by 6 months;

the number of deaf infants who would be subject to intervention by 12 months;

the number of non-deaf infants with unnecessary interventions; and

the number of deaf children with normal language outcomes at entry to grade school.

These measures of benefits were obtained from modelling, assuming a cohort of 80,000 infants.

Direct costs
The direct costs considered in the economic analysis were those of the health service. These included fixed costs (machinery), variable costs, diagnostic evaluation costs, costs of special education, vocational rehabilitation, assistive devices and medical costs. The variable costs were for supplies, probe tips and probes for TEOAE, electrode and machine calibration for AABR, wages (programme coordinator, screener, clerk and audiologist) and the tracking software costs.

To estimate the UNHS costs it was assumed that one TEOAE machine and one AABR machine were needed for every 2,000 newborns. To estimate the SS costs it was assumed that only one AABR machine was needed, and the costs of the screening procedure to identify patients at HR were also included. Most of the resource quantities were reported separately from the costs. The sources of the costs were the National Center for Hearing Assessment and Management, and the audiology department of the Children's Hospital (Boston). The costs were estimated from actual data and some assumptions made by the authors. The total costs were derived using the decision tree. Both the average and incremental costs were reported. Discounting was performed using a rate of 3%. This was appropriate since the time
horizon considered at analysis was lifetime. The price year was 2001. Adjustments were made using the Consumer Price Index for medical costs and other goods, and the employment cost index for wages.

**Statistical analysis of costs**
No statistical analysis of the costs was reported.

**Indirect Costs**
The indirect costs included in the economic analysis were the productivity losses due to delayed language. The resource quantities and the costs were not reported separately. These costs appear to have been based on a published study and, therefore, actual data. On the basis of this study, the authors assumed that improve language outcomes resulting from early intervention would result in a 75% decrease in lost productivity. Discounting was performed using a 3% discount rate. This was appropriate since the period of follow-up was a lifetime temporal horizon. The price year was 2001.

**Currency**
US dollars ($).

**Sensitivity analysis**
Sensitivity analyses were performed on both the effectiveness and costs estimates in order to assess the robustness of the results. The area of uncertainty investigated was variability in the data. All the effectiveness estimates were varied within ranges that seem to have been sufficiently wide (see the 'Effectiveness Results' section). Moreover, the authors performed sensitivity analyses on the cost assumptions they had made to estimate the costs. Generally, one-way sensitivity analyses appear to have been conducted. The exception was a two-way sensitivity analysis, which was performed by varying the proportion of LR deaf infants with normal language abilities after early intervention against the percentage increase in lifetime productivity due to normal language abilities.

**Estimated benefits used in the economic analysis**
Considering a cohort of 80,000 infants, 86 would be HR and 42 would be LR.

The number of HR infants (out of 86) whose deafness would be diagnosed by 6 months was 22 with NS, 58 with SS and 67 with UNHS. The corresponding numbers of LR infants (out of 42) were 8 (NS), 8 (SS) and 32 (UNHS).

The incremental number of deaf infants who were diagnosed by 6 months with SS when compared to NS was 36, while 33 additional deaf infants were diagnosed by 6 months with UNHS when compared to SS.

The number of HR deaf infants who would be subject to intervention by 12 months would be 35 with NS, 59 with SS and 66 with UNHS. The corresponding numbers of LR deaf infants would be 15 (NS), 15 (SS) and 32 (UNHS).

The number of HR non-deaf infants with unnecessary interventions was 0 with NS, 1 with SS and 1 with UNHS. The corresponding numbers of LR non-deaf infants were 0 (NS), 0 (SS) and 7 (UNHS).

The number of HR deaf children with normal language outcomes at entry to grade school was 32 with NS, 37 with SS and 39 with UNHS. The corresponding numbers of LR deaf children were 21 (NS), 21 (SS) and 26 (UNHS). Therefore, SS resulted in 6 additional deaf children with normal language abilities when compared to NS, while UNHS resulted in 6 additional deaf children with normal language abilities when compared to SS.

These benefits were estimated according to a lifetime temporal horizon.

**Cost results**
The total cost of screening was $1,555,100 ($19 per infant) for UNHS and $614,700 ($59 per infant) for SS.
The diagnostic evaluation costs per infant were the same for both the UNHS and SS strategies, $540.

The lifetime costs per deaf child (including lost productivity, special education, vocational rehabilitation, assistive devices and medical costs) were $1,126,300 per deaf child with delayed language and $697,500 per deaf child with normal language. These costs appeared to be the same, independent of the strategy adopted.

The lifetime costs for the whole cohort were $116,980,800 for NS, $115,520,600 for SS and $114,648,300 for UNHS. Therefore, the incremental cost-savings were $1,460,200 for SS when compared with NS and $872,300 for UNHS when compared with SS.

**Synthesis of costs and benefits**
The estimated benefits and costs were combined in cost-effectiveness ratios. These calculated the total cost of detecting one case of deafness in the cohort, the cost per infant whose deafness was diagnosed by 6 months, and the cost per deaf child with normal language outcomes.

The total cost per deaf infant detected in the cohort was $69,200 for NS, $671,200 for SS and $2,122,700 for UNHS.

The cost per deaf infant diagnosed by 6 months was $2,300 for NS, $10,100 for SS and $21,400 for UNHS.

The cost per deaf child with normal language outcomes was $2,215,500 for NS, $1,978,100 for SS and $1,769,300 for UNHS.

Incremental cost-effectiveness ratios were also calculated, as the incremental cost per infant whose deafness was diagnosed by 6 months for SS compared to NS and for UNHS compared to SS.

The incremental cost-effective ratio per deaf infant diagnosed by 6 months was $16,400 for SS compared to NS and $44,300 for UNHS compared to SS.

The authors reported that the model was moderately sensitive to changes in the screening success rate for SS and the rate of follow-up with diagnostic evaluation after positive screening. Moreover, the cost-effectiveness of UNHS was very sensitive to changes in the estimates of the proportion of LR deaf infants with normal language abilities after early intervention, and the percentage increase in lifetime productivity given normal language abilities. The sensitivity was such that UNHS was only cost-saving for a limited range of probabilities for these parameters.

**Authors’ conclusions**
The results of the analysis show that universal newborn hearing screening (UNHS) will be cost-saving in the long run when the proportion of deaf infants achieving normal language outcomes and the increased productivity under early intervention are high. The incremental cost-effectiveness ratios for both UNHS and selective screening (SS) are comparable to those of screening programmes for other newborn conditions, such as congenital hypothyroidism and phenylketonuria.

**CRD COMMENTARY - Selection of comparators**
A justification was given for the comparators chosen. SS was chosen because it was the alternative used prior to the implementation of UNHS in the authors’ setting. Also, it was the current strategy used in many other settings related to the authors. NS was not justified, but it is a reasonable comparator for the health technology under study. You should decide whether these alternatives are widely used in your own setting.

**Validity of estimate of measure of effectiveness**
The authors did not state that a systematic review of the literature had been undertaken. The effectiveness estimates were reported using narrative methods. The authors considered the impact of the differences between the primary studies by means of sensitivity analyses, which were performed for all the effectiveness estimates included in the analysis.
The proportion of infants with HR status may have been underestimated in the base-case analysis, because it only considered those children who were admitted to a neonatal intensive care unit. However, this issue was accounted for in the sensitivity analyses. Sensitivity analyses were performed on all the effectiveness estimates. The ranges reported appear to have been realistic according to the results of the review. A Delphi panel of 4 experts was assembled to estimate the impact of early interventions on language outcomes and subsequent increases in productivity. However, as the authors reported, the true impact of these estimates is not known. Moreover, they stated that the estimates of the proportion of deaf infants diagnosed by 6 months, those receiving intervention by 12 months and those reaching normal language abilities, may have been too optimistic. Therefore, caution should be taken when interpreting these results. The reliability of the results is uncertain because the model was sensitive to some of the parameters.

**Validity of estimate of measure of benefit**
The estimation of benefits was modelled using a decision tree. The measures used appear to have been appropriate for the analysis.

**Validity of estimate of costs**
All the categories of costs relevant to the adopted perspective appear to have been considered. The price year was reported and most of the resource quantities were reported separately from the costs. These factors may assist reflation exercises and generalisability to other settings. However, the resource quantities related to special education, vocational rehabilitation, assistive devices, medical costs of treating deaf children, and productivity losses were not reported separately from the costs. The authors made assumptions to estimate the costs, some of which were based on evidence, and performed sensitivity analyses to evaluate how robust the results were when these assumptions were modified. Discounting was performed using a 3% discount rate. This was appropriate given that the study was performed in the USA and the period of follow-up was a lifetime temporal horizon.

**Other issues**
The authors made some comparisons of their findings with those from other studies although, as they stated, there are no studies about the long-term impact of early intervention and normal language abilities on the lifetime costs for deaf infants. The issue of generalisability of the results to other settings was not specifically addressed, other than through the sensitivity analyses. The screening strategies under study attempted to identify infants with bilateral moderate to profound deafness, and this was reflected in the authors’ conclusions. The authors highlighted the fact that this study analysed one specific protocol for screening, but different protocols can be applied depending on the setting considered at analysis.

**Implications of the study**
The authors highlighted the fact that the cost-effectiveness of a screening programme does not depend only on the test properties of the screening machines and protocols used, but also on the ability to ensure follow-up of infants who do not pass screening tests. The authors recommend further research to measure the utilities of deaf children and adults given early or late identification and normal or delayed language abilities, as well as parental utilities concerning hearing screening test results. There is also a need to obtain better evidence about how early intervention affects the language, educational and vocational costs, and lifetime productivity of deaf individuals.

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**Other publications of related interest**


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