Cost and cost-effectiveness of universal screening for hearing loss in newborns
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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
Among the numerous protocols for screening for hearing losses in newborns, four distinct strategies were considered: a short version of auditory brainstem evoked response (S-ABR) performed only at birth (S-ABR/None); S-ABR repeated twice, at birth and at follow-up (S-ABR/S-ABR); otoacoustic emissions (OAE) performed at birth and at follow-up (OAE/OAE); and S-ABR at birth for newborns who failed OAE (OAE then S-ABR/None). These protocols were those most commonly practised in the USA. Together they represent over 90% of the screening programmes currently in place.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population included all newborns, given that screening for hearing losses is universally recommended at every hospital.

Setting
The setting was the community. The economic study was carried out in Seattle, WA, and Logan, UT, USA.

Dates to which data relate
Effectiveness and resource data were derived from studies published between 1990 and 1999. All cost data were gathered in 1999.

Source of effectiveness data
The effectiveness data were based on published studies.

Modelling
A decision-analytic model was constructed to represent the natural history of a cohort of infants screened at different stages through the four screening strategies. Three stages were considered in the model. In the first stage there was screening at birth, in the second stage a screening or “no screening” option was considered, and in the third stage diagnostic evaluation was performed. The inputs of the model were derived from the literature review. An important assumption of the model was that all infants had an initial screening test.

Outcomes assessed in the review
The outcomes included in the review, and used as inputs in the decision model, were the prevalence of hearing loss, the specificity, sensitivity and costs of each test, and the percentage of infants returning to the hospital for follow-up testing.

**Study designs and other criteria for inclusion in the review**
To reflect the real-world experience of screening programmes, only large, population-based studies and operation programmes were included in the review.

**Sources searched to identify primary studies**
Not stated.

**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**
Twelve primary studies were included.

**Methods of combining primary studies**
The method of combination of the primary studies was not reported. However, ranges of estimates were provided.

**Investigation of differences between primary studies**
Not reported.

**Results of the review**
The prevalence of hearing loss per 1000 infants was 3.5 (range: 2.3 - 4.6).

The sensitivity estimate was 0.95 (range: 0.90 - 0.98) for S-ABR at birth and follow-up, 0.95 (range: 0.90 - 0.98) for OAE at birth and follow-up, and 0.9025 (range: 0.81 - 0.96) for OAE then S-ABR.

The specificity estimates were 0.95 (range: 0.78 - 0.99) for S-ABR at birth, 0.925 (range: 0.83 - 0.97) for S-ABR at follow-up, 0.90 (range: 0.84 - 0.96) for OAE at birth, 0.87 (range: 0.78 - 0.92) for OAE at follow-up, and 0.95 (range: 0.93 - 0.98) for OAE then S-ABR.

Costs were $18.87 for S-ABR, $10.17 for OAE, $13.91 for OAE then S-ABR.

Diagnostic evaluation cost $150, and was associated with a sensitivity and a specificity both equal to 1.

The percentage of infants returning to the hospital for follow-up testing was 0.79 (range: 0.61 - 0.91).

**Measure of benefits used in the economic analysis**
The benefit measure was number of infants with hearing loss identified. This estimate was based on the sensitivity value of each protocol (obtained by pooling the sensitivity values of the screening test at each stage of the model).
Direct costs
The costs estimated in the study were the costs of equipment, personnel, overheads, and clerical work calculated for the performance of each category of screening and the costs of diagnostic evaluation (stage 3 of the decision tree). Discounting was not relevant and was not carried out given that costs were incurred over a period of less than 2 years. The estimation of the costs of the screening tests was based on data published by the US National Center for Hearing Assessment and Management for hospitals with 2,000 births per year. The costs of the diagnostic evaluation were derived from published cost-effectiveness studies. The overall cost of each screening protocol was obtained through the decision model. The quantity/cost boundary was that of the hospital. Costs were measured in 1999.

Statistical analysis of costs
Statistical analysis was not carried out.

Indirect Costs
Indirect costs were not included.

Currency
US dollars ($).

Sensitivity analysis
To assess the robustness of the base-case results, one-way sensitivity analyses were performed for all variables estimated in the review within the range of reported values. Two-way sensitivity analyses were also conducted by varying the sensitivity and specificity estimates of the screening tests. Finally, best and worst case scenarios were considered using 3 variables (costs, sensitivity and specificity).

Estimated benefits used in the economic analysis
The estimated benefit (protocol sensitivity) was 0.9025 for S-ABR/S-ABR, OAE/OAE, and OAE then S-ABR/None. The estimated benefit of S-ABR/None was 0.95.

Cost results
The total costs of the protocols were $20.48 for S-ABR/S-ABR, $25.17 for S-ABR/None, $12.91 for OAE/OAE, and $20.19 for OAE then S-ABR/None.

Synthesis of costs and benefits
Costs and benefits were combined by performing cost-effectiveness analysis. The cost per infant with hearing loss identified was $8,112 for S-ABR/S-ABR, $9,470 for S-ABR/None, $5,113 for OAE/OAE, and $7,996 for OAE then S-ABR/None. An incremental analysis was also performed, by computing the incremental cost-effectiveness of S-ABR/None with OAE/OAE, that was about $92,000. The OAE/OAE protocol was the most cost-effective strategy. Overall, sensitivity analyses confirmed the results of the base case and indicated that the cost of screening carried out at birth had the largest impact on the analysis.

Authors' conclusions
The otoacoustic emission testing protocol should be adopted in screening programmes concerned with cost and cost-effectiveness.

CRD COMMENTARY - Selection of comparators
The rationale for the choice of the health technologies compared was clear: they represent the current practice in USA.
Validity of estimate of measure of effectiveness
The effectiveness measures were based on a review, but the authors do not report the methods for identifying and assessing studies for inclusion in the reviews. Moreover, the authors only provided the range of effectiveness values, without mentioning the procedure used to combine the single estimates from the primary studies.

Validity of estimate of measure of benefit
The estimation of benefit was derived from a model, which seems appropriate to describing the course of the disease. However, resource use (and cost) was only correlated with the rate of true positives (infants with hearing loss detected). This does not account for the rate of false positives (infants without hearing loss classified as having hearing loss). In fact, it can be shown that increasing sensitivity in order to detect more true cases will be at the expense of decreasing specificity, which will increase the rate of false positives. Also it is not known what the value of detecting or not detecting these cases is in terms of health consequences or quality of life.

Validity of estimate of costs
The inclusion of indirect costs would have been useful given that the parents' productivity losses are likely to be relevant. Direct costs incurred not only from the hospital perspective should have been included in the analysis.

Other issues
The issue of generalisability to other settings, even though not specifically addressed, was likely to have been enhanced by the various sensitivity analyses performed, given that the authors used wide ranges of estimates derived from the literature. In addition, the authors reported comparisons with other studies in order to provide a rough assessment of the model's validity. The authors also reported some limitations of the analysis, such as the fact that data, especially those about the specificity and sensitivity of the tests, were quite limited. Finally, the authors’ conclusions were mainly based on average cost-effectiveness ratios: a more detailed incremental analysis might have been more appropriate.

Implications of the study
The authors suggested that hospitals should adopt the OAE strategy if the objective is the choice of the more cost-effective screening protocol. Further research is needed to assess the impact of distinguishing between unilateral and bilateral hearing losses on the effectiveness measures. Additional data about the sensitivity and specificity of the screening tests would also be useful.

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