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 technology studied was universal screening of newborns for the detection of significant bilateral congenital hearing loss. All newborns would be screened with an automated transient-evoked otoacoustic emissions (TEOAE) device. Infants with positive results would then be screened with an automated auditory brain response (ABR) device. Infants with positive results on this second screen would then be referred for diagnostic ABR testing.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population comprised a hypothetical cohort of newborn children in the United States.

Setting
The study setting was hospital. The economic analysis was carried out in the United States.

Dates to which data relate
Effectiveness and resource use data were collected from studies published between 1993 and 1997. Cost data were collected from studies published between 1995 and 1997. The price year was not reported.

Source of effectiveness data
Effectiveness data were derived from a review of the literature and authors' assumptions.

Modelling
A decision analytic model employing a decision tree was used to determine the cost-effectiveness of the two screening strategies.

Outcomes assessed in the review
The review assessed the prevalence of hearing loss, and the sensitivity and specificity of risk screening, automated TEOAE and automated ABR.

Study designs and other criteria for inclusion in the review
Not stated.
Sources searched to identify primary studies
The authors searched MEDLINE using the term 'hearing tests' and restricted the review to articles written in English.

Criteria used to ensure the validity of primary studies
Not stated.

Methods used to judge relevance and validity, and for extracting data
Not stated.

Number of primary studies included
Ten primary studies were included in the review.

Methods of combining primary studies
A narrative method was used to combine primary studies.

Investigation of differences between primary studies
Not stated.

Results of the review
The results of the review were as follows:

prevalence of hearing loss was 0.11% (range: 0.10% - 0.59%);
sensitivity and specificity of risk screening were 59% (range: 50% - 64%) and 95% (range: 91% - 99%), respectively;
sensitivity and specificity of automated TEOAE were 80% (range: 66% - 100%) and 92% (range: 91% - 93%), respectively; and
sensitivity and specificity of automated ABR were 98% (range: 80% - 100%) and 96% (range: 86% - 98%), respectively.

Measure of benefits used in the economic analysis
The number of cases identified was used as the measure of health benefits.

Direct costs
Direct costs were not discounted given the short time frame of the study (less than one year). Quantities and unit costs per test were reported separately. Direct costs related to the costs of screening and follow-up testing. The quantity/cost boundary adopted was that of the hospital. Cost data were taken from the published literature and discussions with experts. The price year was not reported.

Statistical analysis of costs
No statistical analyses of costs were reported.

Indirect Costs
Indirect costs were not included.

**Currency**  
US dollars ($).

**Sensitivity analysis**  
Univariate sensitivity analyses were performed on all variables and two-way sensitivity analyses were conducted to test sensitivity and specificity.

**Estimated benefits used in the economic analysis**  
At baseline, prevalence was 110/100,000 newborns. The numbers of cases detected with targeted screening was 51 and with universal screening, 86.

The number of false positives with targeted screening was 16 and with universal screening, 320.

The positive predictive value was 76% with targeted and 21% with universal screening.

**Cost results**  
Total costs were $158,860 with targeted screening and $1,004,860 with universal screening.

**Synthesis of costs and benefits**  
The cost per case detected was $3,120 with targeted screening and $11,650 with universal screening.

The incremental cost per case detected of universal over targeted screening was $23,930.

Only if the cost of screening infants for high-risk factors was more than $5.34 per infant would the cost per case detected by universal screening be less than by targeted screening. The authors also stated that "over all ranges of probability estimates, universal hearing screening detects more cases than targeted screening at a greater cost."

**Authors' conclusions**  
"Universal screening detects more cases of congenital hearing loss, at the expense of both greater cost and more false-positive results. Little is known about the negative impact of false positives 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."

**CRD COMMENTARY - Selection of comparators**  
A justification was given for the comparator used, namely that it represented a commonly used strategy. You, as a user of the database, should decide if these health technologies are relevant to your setting.

**Validity of estimate of measure of effectiveness**  
The authors did not state that a systematic review of the literature had been undertaken. More information about the design of the review and the method of pooling primary effectiveness estimates could have been reported. As the authors noted, the benefits of a screening programme accrue through the accuracy of the strategy, which is measured using sensitivity and specificity, (for which appropriate evidence was given), and the subsequent consequences in terms of quality of life, for which they state evidence was not available.
Validity of estimate of measure of benefit
Benefit was measured only in terms of cases detected, which the authors admit is flawed, in that, a strategy which detects more cases, as with universal screening, detects false positives, which might be harmful. However, the harm is not likely to be nearly as great as in their comparisons with mammography or for Down's syndrome. The authors cited lack of data as justification for not using a standardised measure of benefits, such as quality-adjusted life years, for comparison of the results with those of other health technologies.

Validity of estimate of costs
Good features of the cost analysis were that sensitivity analyses were conducted on cost estimates, and quantities and unit costs were reported separately, which enhances the generalisability of the results. However, the price year was not reported (which would make reflation exercises in other settings difficult), as they acknowledged the authors only included short-term costs and did not consider long-term costs, such as the costs of treatment and any potential savings from the early initiation of treatment, and indirect costs, such as parental wages lost, transportation costs, and child-care costs incurred because of the screening process, were not included. Also, as the results of the sensitivity analysis were not given, it is not possible to observe any possible thresholds in terms of willingness to pay for the increase in case detected.

Other issues
The authors did make appropriate comparisons of their findings with those from other studies but the issue of generalisability to other settings was not addressed, except generally through the sensitivity analysis. The authors did not seem to present their results selectively. The study considered newborn children and this was reflected in the authors' conclusions.

Implications of the study
"Universal screening detects more cases of congenital hearing loss, at the expense of both greater cost and more false-positive results. To maximise the cost-effectiveness of screening 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."

It appears that the study does not provide any new evidence; it should have been possible to predict that universal screening would detect more cases at increased cost. Furthermore, the study does not give a useful idea of cost per unit benefit, since, as the authors acknowledge, these data were not included in the model. If the literature search was sufficiently extensive, it might highlight the need for the further research recommended by the authors. However, a decision-maker should consider seeking confirmation of this before commissioning such research.

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