The economics of screening infants at risk of hearing impairment: an international analysis
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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.

CRD summary
The study estimated the cost-effectiveness of different screening strategies for detecting hearing impairment in infants. The authors concluded that as cost-effectiveness was dependent on the cost per patient of the intervention and the baseline risk, screening interventions were more likely to be cost-effective with higher baseline risk. The methodology was appropriate and the results were reported adequately. The methods were not well reported and so it is difficult to assess the authors’ conclusions.

Type of economic evaluation
Cost-effectiveness analysis

Study objective
To estimate the cost-effectiveness of different screening strategies for detecting hearing impairment in infants.

Interventions
The study made two unique comparisons. Universal screening with transient evoked otoacoustic emissions (TEOAE) or automated auditory brainstem response (AABR) was compared with selective screening for those with risk factors (family history of hearing impairment or infants with craniofacial anomalies). And a one-stage screening process (TEOAE only) was compared with a two-stage process (positive TEOAE followed by AABR).

Location/setting
UK and India/Primary care

Methods
Analytical approach:
Two decision tree models were used to combine published evidence to inform on the likely consequences of the alternative screening strategies on a hypothetical cohort of 100,000 newborn infants. The base case analysis took the perspective of the healthcare system. A secondary analysis took a societal perspective.

Effectiveness data:
Effectiveness data were derived from published studies. The main clinical effectiveness estimates were sensitivity and specificity of TEOAE and AABR. These estimates were derived from a previously published cost-effectiveness analysis.

Monetary benefit and utility valuations:
Not relevant.

Measure of benefit:
The primary measure of benefit was the number of detected true positive cases of hearing impairment.

Cost data:
The base-case analysis (healthcare system perspective) included direct costs associated with the TEOAE and AABR screening methods, resource use estimates associated with screening and a cost of false positive results. Costs of screening methods included costs of screening interventions and medical supplies. Resource costs included costs of a coordinator, screener, clerk and audiologist. Cost of a false positive result was assumed to be an additional cost of an outpatient audiologist visit. Cost estimates were obtained from studies in the published literature.
The secondary analysis (societal perspective) included travel time and lost productivity and an estimate of the lifetime societal costs in USA from a single published study.

Costs were provided in 2010 UK pounds sterling (£) in the base-case analysis; costs were converted into Indian Rupees (INR) where relevant.

Analysis of uncertainty:
Deterministic sensitivity analysis was performed to assess the impact of uncertainty around key parameters on results.

Results
For a cohort of 100,000 infants, universal screening was estimated to identify an additional 63 true positive cases in the UK at an incremental cost per case detected of £36,181 (US$58,497). Universal screening was estimated to identify an additional 376 true positive cases in India at an incremental cost per case detected of INR 157,084 (US$9,863).

Compared with two-stage screening, one-stage screening was estimated to cost an additional £861,925 in the UK and INR 21,845,669 in India. Compared with the two-stage strategy, one-stage screening was estimated to identify an additional seven true positive cases in the UK at an incremental cost per case detected of £120,972 (US$195,586) and an additional 24 true positive cases in India at an incremental cost per case detected of INR 926,675 (US$58,183).

The sensitivity analysis showed that the results were highly dependent on the baseline prevalence risk of hearing impairment and the cost per patient of the intervention.

Authors' conclusions
The authors concluded that as cost-effectiveness was dependent on the cost per patient of the intervention and the baseline risk, screening interventions were more likely to be cost-effective with higher baseline risk.

CRD commentary
Interventions:
The level of reporting of the interventions was good. The relevance of interventions to various settings should mean that the results were generalisable. It was unclear whether no screening was a relevant alternative in some international settings and may have been a relevant omitted comparator.

Effectiveness/benefits:
Much of the effectiveness data were taken from a previous cost-effectiveness analysis of newborn hearing strategies; this study was referenced but not described and so its quality could not be assessed. Readers would need to refer to this study to fully assess its quality. It was unclear how the included studies were identified. It was unclear whether a systematic review was undertaken and so whether the best available evidence was included in the study. The benefit measure appeared appropriate for this study, but it was quite disease specific and so may have reduced the comparability of this study's results with studies on other disease areas.

Costs:
The base-case analysis and secondary analysis perspectives were stated clearly. All relevant cost categories for the base-case analysis appeared to be included. The studies from which cost and resource use data were derived were referenced but not described so it was not possible to assess their quality and their appropriateness. Times associated with resource use were noted, but was not clear where these were from and whether they were appropriate for both UK and Indian settings. The source of costs applicable to the secondary analysis from the societal perspective were not provided and it was not clear how applicable a USA estimate of the lifetime societal costs of hearing impairment were to studies of UK and India.

It appeared that costs were adjusted appropriately for inflation. The time horizon of the study was not explicitly stated but seemed to be less than one year and so it was appropriate that costs were not discounted. The secondary analysis discussed lifetime costs. It appeared that these costs were not included in the analysis but were included in the discussion regarding the importance of hearing impairment strategies.

Analysis and results:
Use of an incremental approach was appropriate to estimate the relative cost-effectiveness of the alternative screening strategies. The analytical approach appeared appropriate and was described adequately. Graphical depictions were provided. The results were described adequately. The impact of parameter uncertainty was addressed to an extent by a series of one-way sensitivity analyses. The authors discussed the limitation of not including the health-related quality of life associated with avoiding hearing loss.

Concluding remarks:
The methodology was appropriate and the results were reported adequately. The methods were not well reported and so it was difficult to assess the authors' conclusions.

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