Screening strategies for neonatal hearing loss: which test is best?

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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
Three screening strategies for neonatal hearing loss: automated auditory brainstem response (AABR), distortion-product otoacoustic emissions (DPOAEs), and click-evoked otoacoustic emissions (TEOAEs). All screening was carried out with commercially available equipment and all used a computer external unit and sound delivery system (either insert earphones or probe) to deliver the auditory stimulus.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population consisted of neonates from the well baby (WBN) and special care (SCN) nurseries at the Foothills Hospital in Calgary, Canada. The sex distribution was roughly equal although girls outnumbered boys in the SCN population. The majority of babies (92/105) came from the WBN. The mean age of the SCN babies was significantly older than the WBN babies (480 versus 31 hours) due to the fact that SCN babies were typically sicker and had a longer hospital stay.

Setting
The setting was tertiary care. The economic study was carried out in Canada.

Dates to which data relate
No effectiveness or resource use dates were reported. The price year was not reported.

Source of effectiveness data
Effectiveness data were derived from a single study.

Link between effectiveness and cost data
Costing was prospectively carried out on the same sample of patients as that used in the effectiveness study.

Study sample
Power calculations were not reported. The sample was selected by asking for parents' consent in the nurseries of the hospital. The authors did not investigate whether there were significant differences between the study sample obtained through consenting parents and the study population which comprised all neonates. The overall number of subjects was 105; the breakdown by type of screening intervention was not reported. The percentage of parents who refused
participation for their babies was not reported. There were no other inclusion or exclusion criteria for the study.

**Study design**
The study design was a randomised controlled trial conducted in a single centre. The test sequence was randomly assigned using a random numbers table to determine the test order. There was no follow-up of babies beyond the initial screening (although babies who failed hearing screening were referred for further testing). Partial data were obtained on some infants due to the nature of testing newborns but the losses to follow-up were not reported. Blinding was not conducted for the assessment of outcomes, but it was not stated whether blinding would have been possible for this type of intervention.

**Analysis of effectiveness**
The basis for the analysis of effectiveness (intention to treat or treatment completers only) was not stated. The primary health outcomes used in the analysis were the sensitivity and specificity of the DPOAEs and TEOAEs tests, using the AABR as the gold standard. No statistical analyses of patient characteristics were conducted at baseline and no investigation of potential confounding factors was conducted.

**Effectiveness results**
The sensitivity and specificity of TEOAEs were 85.7% and 49.1% respectively. The sensitivity and specificity of DPOAEs were 71.4% and 61.4% respectively.

**Clinical conclusions**
AABR was the most accurate test in screening for hearing loss in neonates.

**Measure of benefits used in the economic analysis**
No summary measure of benefits was used in the economic analysis, the clinical outcomes being left disaggregated; as such, a cost-consequences analysis was performed. Please refer to the effectiveness results reported earlier.

**Direct costs**
Discounting was not carried out, as the duration of the study was less than 2 years. Some quantities (time to conduct test) were reported separately from costs. The quantities/costs measured were equipment, and disposables and the staffing time needed to conduct the test. The quantity/cost boundary adopted was that of the hospital. The estimation of test duration was based on actual data; the method used for the estimation of other quantities and costs was not reported. With the exception of the test duration, the sources of the quantity and cost data were not reported. The price year was not reported. It was assumed that testing equipment would need to be replaced every five years as a result of wear and tear and the availability of better technology.

**Statistical analysis of costs**
No statistical analyses of costs were carried out.

**Indirect Costs**
Indirect costs were not included in the analysis.

**Currency**
Canadian dollars (Can$).
Sensitivity analysis  
No sensitivity analyses were carried out.

Estimated benefits used in the economic analysis  
The reader is referred to the effectiveness results reported previously. There was no follow-up of patients beyond the initial testing and referral for babies who had tested positive for hearing loss.

Cost results  
The cost results were as follows:

Based on 1,000 infants, the total cost per infant using the AABR test was $25.55.

The total per infant costs using the DPOAEs was $12.89 and the TEOAEs was $15.70.

The screening cost for the Calgary region for the first year, assuming 11,158 newborns at three testing sites was $150,000 for AABR, $81,000 for DPOAEs, and $84,000 for TEOAEs.

The cost per infant in the first year was $13.44 for AABR, $7.24 for DPOAEs, and $7.53 for TEOAEs.

The cost per infant in subsequent years was $8.96 for AABR, $5.20 for DPOAEs, and $4.50 for TEOAEs.

Costs of further testing, referrals and rehabilitation of infants with hearing loss were not included in the analysis.

Synthesis of costs and benefits  
A synthesis of costs and benefits was not performed.

Authors' conclusions  
Hearing screening in a hospital based newborn population is both feasible and cost-effective. Although AABR was more expensive, its better accuracy must be considered. As technology improves, the cost of all three tests will diminish. More robust conclusions cannot be made based on this small patient population.

CRD COMMENTARY - Selection of comparators  
A justification was given for the comparator used; namely that AABR was the conventional testing method for hearing loss. You, as a user of the database, should decide if this is a widely used health technology in your own setting.

Validity of estimate of measure of effectiveness  
The study was based on a randomised controlled trial, which was appropriate for the study question. The study sample was representative of the study population, namely neonates. There were some weaknesses in the analysis of effectiveness viz.: potential confounding factors due to the non random agreement to participate in the study were not accounted for in the analysis; patient groups were not shown to be comparable at analysis; statistical analyses were not undertaken in the analysis of effectiveness; the breakdown by intervention category was not reported; and the dates of the study were not reported.

Validity of estimate of measure of benefit  
The authors did not derive a summary measure of health benefit, and, as such, a cost-consequences analysis was conducted.
Validity of estimate of costs
The analysis of the cost of testing was well conducted and included all relevant categories of cost, with some quantities (duration of testing) reported separately from costs. However, the main weakness with the costing was the exclusion of costs incurred when the testing showed loss of hearing, such as referrals and rehabilitation as well as the cost of not detecting hearing loss in neonates. The omission of these costs is likely to have affected the authors’ conclusions. Other problems with the analysis of costs included the omission of the source of the cost data (for example the labour costs, costs of equipment and disposables) as well as the source of some of the resource use data (equipment and disposables). Furthermore, no sensitivity analyses or statistical analyses were conducted. Finally, the dates for which data were collected as well as the price year were not reported.

Other issues
The authors made appropriate comparisons of their results with findings from other studies but did not address the issue of generalisability to other settings. The authors did not present their results selectively. The study enrolled neonates and this was reflected in the authors’ conclusions. The authors reported that the small size of the study was likely to affect the robustness of the results.

Implications of the study
The authors stated that they could not draw final conclusions about the utility of otoacoustic emissions in a hearing screening programme in their centre based on the results of this study, which was preliminary. These conclusions await the completion of further study.

Source of funding
Support from the Alberta Heritage Foundation for Medical Research and the Campbell McLaurin Foundation for Hearing Deficiencies.

Bibliographic details

PubMedID
11003070

Indexing Status
Subject indexing assigned by NLM

MeSH
Canada /epidemiology; Catchment Area (Health); Cochlea /physiology; Cohort Studies; Cost-Benefit Analysis; Evoked Potentials, Auditory, Brain Stem /physiology; Feasibility Studies; Female; Health Care Costs; Hearing Disorders /diagnosis /economics /epidemiology; Humans; Infant, Newborn; Male; Neonatal Screening /economics /standards; Otoacoustic Emissions, Spontaneous /physiology; Prospective Studies; Sensitivity and Specificity; Time Factors

AccessionNumber
22000001505

Date bibliographic record published
30/11/2001

Date abstract record published
30/11/2001