A critical review of the role of neonatal hearing screening in the detection of congenital hearing impairment

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Authors' objectives
To assess the role of neonatal hearing screening in the detection of congenital hearing impairment. The authors also collected information on how to deliver a more uniform service, better outcomes and more cost effective screening, and identified areas for further research and development.

Searching
The authors searched the electronic databases of BIDS, EMBASE, ISSI and ISI, ERIC, PsycLIT, MEDLINE, CINAHL, the British Library catalogue and thesis listings (dates not specified) using a list of keywords developed by expert members of the review team. Searches of the internet and specialist bibliographies on paediatric audiology were also conducted. Additional, pre-publication studies were supplied by interested professionals.

Study selection
Study designs of evaluations included in the review
Screening studies and retrospective ascertainment studies. There were no randomised controlled trials in this field.

Specific interventions included in the review
The review included studies of the efficacy of universal and targeted neonatal hearing screening programmes, and the Health Visitor Distraction Test (HVDT).

Reference standard test against which the new test was compared
No inclusion criteria were specified in relation to any reference standard screening test.

Participants included in the review
Children with congenital hearing impairment defined as being a permanent bi-lateral hearing impairment on the better ear of greater than or equal to 40 dB over the frequencies 0.5, 1, 2 and 4 kHz. Participants had to be born between January 1, 1985 and December 31, 1993 and currently living in the area covered by the Trent Regional Health Authority.

Outcomes assessed in the review
Test sensitivity and specificity, screening sensitivity and specificity, coverage of screening method, median age of identification of hearing impairment (in months), and costs.

How were decisions on the relevance of primary studies made?
Two of the reviewers selected the papers for review. Papers were excluded only if both reviewers deemed them irrelevant. A senior member of the team made an additional scan of the discarded papers to check if relevant papers had been missed. The papers were graded on a scale of 1 to 3 for their relevance to the study (1 = essential reading; 2 = not essential but relevant; and 3 = do not read, not relevant).

Assessment of study quality
The authors did not report a formal method of assessing validity. However, they state that decisions on which papers to include were made on the basis of size, relevance, quality, and date of publication.

Data extraction
Reviewers read papers and extracted the data using a predefined checklist. A summary of each paper was made by the reviewer.
Methods of synthesis
How were the studies combined?
The studies were combined in a qualitative narrative review.

How were differences between studies investigated?
Studies were grouped into those relating to epidemiology and public health, early intervention and outcomes, surveys of current UK practice, costs associated with screening, and performance of screening. The latter three of these groups were further sub-divided by screening test.

Results of the review
Twenty-seven studies (designs not reported) with 143,825 participants (although one study's numbers of 1,367 counted ears screened not individual participants). Eighteen studies were peer-reviewed publications (52,260 participants); 8 studies were pre-prints (85,419 participants); 2 studies were general reviews for edited volumes (6,103 participants); and 1 study was an unpublished audit report (43 participants).

Neonatal:
All methods of neonatal hearing screens showed high screen specificity, generally well above 90% after a 'settling-in' period.

Evidence on screen sensitivity for moderate and greater cases of congenital PCHI is less available, but estimates range from 80% to 100%, except for the portable auditory response cradle (PARC) when used with at-risk babies, where sensitivity may fall to unacceptable levels. Generally, programme sensitivity (including cases which were not covered, and/or which are late-onset or progressive) may be estimated to be nearer 80% than 100%.

The two large cohort studies of UNS in the UK, including the controlled trial in Wessex, produce yields of the expected order, that is 1-1.3 per 1000, but decrease the subsequent incremental yields of the HVDT to very low levels.

At-risk neonatal screening has a potential yield of about 60% of all cases. In practice, this is likely to be much lower because of the difficulty of implementing full coverage for all indicators of at-risk cases; perhaps 45-50% at best.

The median age of identification of those screened neonatally is of the order of 2 months, depending on follow-up procedures and severity of impairment; this is earlier than for cases not screened neonatally.

Coverage of the HVDT screen probably falls in the range 80-95%, although there may be some urban areas where coverage falls to nearer 60%. There is some limited evidence suggesting that coverage and sensitivity is lower in the Asian population.

HVDT:
Sensitivity estimates of the HVDT vary widely, from 18% to 88% (all degrees of loss). Recent studies and more powerful studies are suggestive of poorer levels of sensitivity. Severity of impairment affects screen sensitivity substantially.

Screen-positive, that is, 'fail' rate is of the order of 5-10%. Many of these cases will have fluctuating non-permanent hearing loss, associated with otitis media with effusion (OME). This referral rate has considerable resource implications for services.

HVDT incremental yield may be at very best 40% but it falls off for the best districts to low levels (e.g. 25%) when at-risk neonatal screening is introduced. With UNS, the evidence indicates that the HVDT incremental yield falls to very low levels. (For those Districts who supplied data in the survey of current practice, the average HVDT yield was about 26-28% and at-risk neonatal screening made no difference).

Median age of identification via the HVDT varies from 12 to 20 months. Age of referral is severity-dependent.
Cost information
Cost comparisons within the different implementations of hearing screening in the first year of life are uniform, with systematic differences being observed between implementations such that UNS appears to have lower initial cost associated with it than the HVDT on a cost per child screened basis. The estimated cost per case is an order of magnitude lower with UNS.

Targeted neonatal screening programmes cost between about a quarter and one-third of the cost of UNS programmes. In terms of running costs (excluding start-up and equipment costs, but including employers full cost plus 40% overheads) in the nine districts surveyed, HVDT screening is costing about £24,500 per 1,000 live births, including follow-up of false-positives. This is reduced to about £20,600 when structured surveillance is used instead of the distraction test.

Authors’ conclusions
Neonatal screening, in the UK, has been successfully implemented for targeted and universal screening. In the UK, one research programme with a systematic prospective comparative study of UNS and HVDT has shown a substantial advantage for neonatal screening, with a greater yield and much younger ages of identification and aiding. These results for yield are corroborated by those from two further centres that UNS routinely. Furthermore, UNS has a lower marginal cost than the HVDT, and a much lower cost per child detected. UNS in the UK has shown that average can be in excess of 90%, with a specificity about 95%. It is too early to assess sensitivity yet but, judging by the yields, it should exceed that of the targeted neonatal screens and may be higher than 90%.

The authors state that the yield of targeted neonatal screening, as presently implemented, is somewhat lower than expected (about 35% rather than 50-60%) but its sensitivity for moderate or worse PCHI has been assessed as between 80% and 90%.

CRD commentary
Though broad, the research questions were clearly stated and limited inclusion and exclusion criteria were outlined. The literature search was thorough and some unpublished studies were included, although it was unclear how these were identified, (were unpublished data sort systematically by the authors or supplied on an add-hoc basis by contacts in the field?) No formal assessment of publication bias was reported.

The quality of the included studies was not formally assessed but the authors did discuss some of their quality-based reasoning for selection of the included studies. The authors reported on how the articles were selected and the process of data extraction. Included studies were summarised in tables and text and a narrative review was appropriate since there was no basis for a statistical pooling of the data.

Although no formal tests for homogeneity were reported, the authors discussed the lack of methodological quality of the studies and related issues, and studies were grouped appropriately within the narrative synthesis. The authors’ conclusions appear to follow from their stated findings but should be viewed with caution in the light of the stated limitations of the review.

Implications of the review for practice and research
Practice: The authors state a number of issues for practice.

1. The National Screening Committee should urgently consider whether there should be a national screening programme for congenital hearing impairment.

2. An information system strategy should be developed to facilitate the coordination of the services needed for screening and following up hearing-impaired children.

3. A model screening programme, with appropriate targets, is proposed around which the preferred option of universal neonatal screening might be based.
Research: The authors state a number of research and development needs.

1. To conduct RCTs to identify the most appropriate methods of management for children, with differing degrees of hearing impairment, identified by neonatal screening.

2. To identify optimum models for service coordination, including joint commissioning.

3. To further refine and evaluate screening techniques (both neonatal and infant) protocols and staff training programmes. To determine the most effective screening methods for those children born outside hospital or rapidly (within 12 hrs of birth) discharged.

4. To estimate prevalence and identify risk factors for late-onset and progressive permanent childhood hearing impairment, and to determine the most effective means of identifying such cases.

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