A programme of studies including assessment of diagnostic accuracy of school hearing screening tests and a cost-effectiveness model of school entry hearing screening programmes

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Scientific summary

School hearing screening tests and SES programmes

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Scientific summary

Background

Identification of permanent hearing impairment at the earliest possible age is crucial to maximise the development of speech and language, and contribute to the best opportunities for educational achievement and quality of life. Approximately 1 in every 1000 children in the UK is born with a permanent bilateral hearing impairment > 40 dB (average across four frequencies: 0.5, 1, 2 and 4 kHz) and a further 0.6 per 1000 has a unilateral impairment. This equates to 800 children per year born with a permanent bilateral hearing impairment (moderate or greater) and 500 with a unilateral impairment. The introduction of the highly sensitive and specific Universal Newborn Hearing Screening (UNHS) programme has led to the identification of the vast majority of children born with a hearing impairment who undergo the screen. However, not all children who will ultimately have a hearing impairment are identifiable at birth.
The adjusted prevalence of permanent hearing impairment > 40 dB (average of 0.5, 1, 2 and 4 kHz) at age 3 years is reported as 1.07 per 1000 and the prevalence for children aged 9–15 years as 2.05 per 1000. Thus, because of acquisition, progression or late onset of hearing impairment and/or geographical movement of families, there remains a significant number of children to be identified with a permanent hearing impairment after the newborn period. The onset of hearing impairment in children can occur at any time, which means there is no optimum time for a further universal hearing screen. The universal distraction hearing test, established in the UK in the 1950s and undertaken by health visitors at around 8 months of age, was abandoned following the introduction of UNHS, based on a lack of robust implementation and a low yield of cases. Without formal screening between the newborn period and school entry, identification of hearing impairment in children is achieved through parental and professional awareness and a close follow-up of children who pass the neonatal screen but are considered to be at risk. A universal hearing screen when children start school, the school entry screening (SES) programme, was established in 1955 and remains in place in many parts of the UK. It is considered as a 'back-stop' screen to identify children as part of a 'captive population' at school entry.

Objectives

The overarching aims of this project were to evaluate the diagnostic accuracy of hearing screening tests and the cost-effectiveness of screening for hearing impairment at school entry in the UK.

The specific research objectives of this project were:

- to update the latest systematic review of diagnostic accuracy of tests used for SES, summarising the literature that has been published since the previous review and drawing together the evidence from the previous review and the updated review
- to estimate and compare the diagnostic accuracy of the pure-tone screen (PTS) (Amplivox, Eynsham, UK), and HearCheck (HC) screener (Siemens, Frimley, UK) tests for discriminating between children with a hearing impairment (of any type) and children with no hearing impairment, using pure-tone audiometry (PTA) results as the reference standard
- to investigate the impact of a potential false-negative result by reviewing the literature on the impact of false-negative results from screening tests and describing children with false-negative screening results in the diagnostic accuracy study

- to compare children referred for investigation of suspected hearing impairment in a geographical area that applies a routine SES programme (Nottingham) with those referred in an area with no routine SES programme (Cambridge) with respect to the number of referrals, the age at referral, the source of referral, the route through assessment to intervention, the number of children ultimately identified to have a hearing impairment (yield) and the nature of hearing impairment identified
- to determine the impact, both psychological and economic, for the child and the family of the child being referred for further assessment following SES (both true and false positives)
- to determine the time resource in implementing either of the two alternative screening methods (PTS and HC) in primary schools and to elicit the views of the school nurses implementing the screening tests
- to refine an existing SES economic model {from the 2007 Health Technology Assessment (HTA) report [Bamford J, Fortnum H, Bristow K, Smith J, Vamvakas G, Davies L, *et al.* Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen. *Health Technol Assess* 2007;**11**(32)]} and assess the cost-effectiveness of the SES programme
- to estimate the health-related quality of life, costs and utilities of the SES programme compared with no screening, and of the PTS compared with HC screener, with comparisons based on cost per quality-adjusted life-year (QALY) gained.

Methods

In order to explore and summarise the existing literature we updated the review of diagnostic test accuracy reported in a previous HTA report and reviewed the literature on false-negative rates in hearing screening.

For children with a known hearing impairment and for children assumed to have no hearing impairment we compared the diagnostic accuracy of two screening methods administered at or around the time children start school. These were the established and widely used PTS (which is applied using headphones) and HC screener (a hand-held PTS). We used PTA as a reference standard.

The yield, referral age and route through assessment to intervention for childhood hearing impairment were assessed for a paediatric audiology service that implements a routine universal SES programme (Nottingham) and one that does not (Cambridge) by collecting data prospectively for all children aged between 3 years and 6 years 364 days.

We surveyed parents of children referred from the SES programme in Nottingham via a postal questionnaire to assess the impact for the child and the family of a positive result from a screen (both true and false positives).

We determined the time spent in implementing either of the two screening tests in primary schools and explored the practical issues involved and the views of nurses conducting the screening tests.

The component data from each study were used to refine an existing SES economic model, providing robust estimates of key parameters beyond accuracy of SES to be assessed; in particular, the yield and nature of hearing impairment detected in a system with no SES progamme; the yield, consequences and costs of screen-positive individuals in an SES programme; and the costs of setting up a SES programme.

Results

The updated review of diagnostic accuracy studies confirms the conclusion from the 2007 HTA report that research to date demonstrates marked variability in the design, methodological quality and results. Robust conclusions about the performance of individual test types for use in SES cannot be drawn. It was found that:

- Parental questionnaires had the poorest diagnostic accuracy compared with all other tests.
- The findings from the new audiometry-based studies evaluating computer-based devices and the HC screener reported higher and more consistent specificity but lower and widely varying sensitivity estimates compared with the sweep PTA studies included in the original report.
- Studies evaluating transient-evoked otoacoustic emissions reported variable sensitivity with wide confidence intervals (CIs), whereas specificity estimates were relatively high and more consistent.
- The study evaluating the automated auditory brainstem response reported high sensitivity and specificity.

The review included studies from countries with and without an established UNHS system and with very different systems of health-care delivery. The generalisability of the findings to other situations, including the UK NHS system, is likely to be limited.

The findings of our diagnostic accuracy study indicate that the PTS and HC devices have a high level of sensitivity (PTS $\geq$ 89%, HC $\geq$ 83%) and specificity (PTS $\geq$ 78%, HC $\geq$ 83%) for identifying hearing impairment at the level of the ear. These conclusions appear robust, the child-level analyses indicating similar levels of sensitivity and specificity.

From our review of the existing literature and data from the diagnostic accuracy study, we are unable to quantify the effect of false-negative results for the PTS or HC screener, but were able to confirm that the rate was extremely low. Of the 16 ears in our diagnostic study (total $n = 630$) that passed one or both of the screening tests but were referred by the PTA measure, only four were confirmed to have a hearing impairment at diagnostic evaluation and all were mild.

There was strong evidence that the rate of referral for hearing problems is lower when a SES programme is present. The referral rate was 36% lower in Nottingham (SES) relative to Cambridge (no SES) (rate ratio 0.64, 95% CI 0.59 to 0.69; $p < 0.001$).

There was little evidence that the yield of confirmed cases differs between areas with and without a SES programme (rate ratio 0.82, 95% CI 0.63 to 1.06; $p = 0.12$); a higher proportion of referred children were subsequently confirmed to be hearing impaired in the area with a SES programme (17.0% in Nottingham vs. 10.6% in Cambridge).

The mean age of referral was nearly identical between areas with and without a SES programme when looking at all referrals, but for children who were subsequently confirmed as having a hearing impairment there was strong evidence that the children in the site with a screening programme are older at referral (mean age difference 0.47 years, 95% CI 0.24 to 0.70 years; $p < 0.001$).

We found from our survey of parents of children referred by the SES programme in Nottingham that the consequences of the referral process for parents and children, including false positives, are minor. The difference for parents whose child is referred by the SES programme is that they may have had no concerns prior to the screening test.

We demonstrated minimal differences between the PTS and HC screener in terms of time taken to conduct each examination and practical issues. Testing covered a range of schools throughout the school year and thus we suggest the findings might be generalisable beyond the Nottingham schools.

Our economic modelling showed that SES is unlikely to be cost-effective and, using base-case assumptions, is dominated by a no screening strategy. This is consistent with the observed results of the clinical studies, which suggest that cases of hearing impairment are identified in similar numbers but at a younger average age in the absence of SES.

Two situations where SES might be cost-effective were identified. In the first situation, a reduction in the number of referrals associated with SES or, conversely, an increase in referrals without SES, can give a cost-effectiveness ratio for the no screening option above the National Institute for Health and Care Excellence (NICE) £30,000 per QALY benchmark. This is supported by the observation from our clinical study that the referral rate (and by assumption, potential false positive rate) was lower in the site where SES had been in place for many years. However, in order for this to be the case, the reduction in referrals would need to be attributable to SES and there is considerable uncertainty about this. The second situation is subject to still greater uncertainty and requires referrals to happen more quickly with screening than is observed from our study comparing SES and non-SES sites.

Conclusions

In the context of the UK NHS, and similar health-care systems, SES using screening tests, such as the PTS and HC screener, is unlikely to be effective in increasing the number of cases of hearing impairment identified and lowering the average age at which these cases were identified. SES is also unlikely to be cost-effective when judged against the benchmarks normally used by NICE, relative to a system entirely reliant on ad-hoc referral when a suspicion of hearing impairment is raised.

Implications for practice

Although our finding of the lack of cost-effectiveness of SES may be considered as a reason to withdraw SES where it is currently being practised, we would highlight aspects of the results that suggest caution. First, we have shown that there are at least two scenarios in which it may be cost-effective. Second, our findings are very dependent on findings in the two specific areas (Nottingham and Cambridge) that were used here, and our conclusions from comparing areas with a SES programme and without a SES programme may not be generalisable to other areas. Third, the cost-effectiveness of SES depends on how effective (or ineffective) the 'no SES system' is. This in turn is highly dependent on the effectiveness of ad-hoc identification and referral for a diagnostic evaluation with an audiologist (DEA), which is not only largely unknown, but likely to be variable. It seems plausible that SES may have greater potential to be cost-effective where ad-hoc identification and referral is less well developed than in a system where it is well established. If withdrawal of the SES programme is to be considered it needs to be carefully managed to ensure that the ad-hoc referral system is working effectively. Health professionals, school and nursery staff, and parents who would then be responsible for referral of children about whom there were concerns in the school entry year may need to be reminded to be more vigilant for signs of hearing impairment.

Implications for research

Systematic reviews of the accuracy of devices, which might be used to measure hearing in children at around school entry age, should continue to be pursued.

Characterising and measuring the cost-effectiveness of different approaches to the ad-hoc referral system with a view to optimising it should be undertaken.

Examination of the process by which concern, or referral from SES, is converted into DEAs would be useful to inform further research on what determines programme specificity (as opposed to test specificity).

We should improve understanding of why the referral rate varies across different sites and determine if this is related to the presence of SES. Further observational studies similar to our comparison between Nottingham and Cambridge could be undertaken, albeit recognising the difficulty of matching the geographical areas.

Further research to better quantify the impact of referral, particularly with respect to anxiety, and whether or not all referrals are affected to the same degree as respondents in our study may be required, particularly if it appears that overall effectiveness and cost-effectiveness could be critically dependent on the costs and disutility experienced by false positives.

If withdrawal of SES is contemplated in particular settings, this could be used as an opportunity for further data collection; in particular where the pattern of referrals and cases was known over many years in the run up to withdrawal, any change in pattern of referrals/cases could be very useful evidence confirming the lack of effectiveness and cost-effectiveness of SES, or challenging it. More formally, if SES cessation is being contemplated in many areas, a randomised trial of withdrawal of SES services could be designed using referrals and hearing impairment cases identified as outcomes.

Trial registration

This trial is registered as ISRCTN61668996.

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For more information about the HTA programme please visit the website: http://www.nets.nihr.ac.uk/programmes/hta

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