

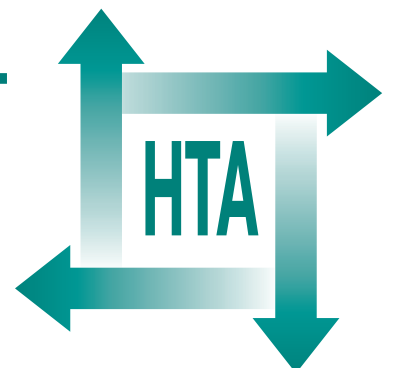
Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen

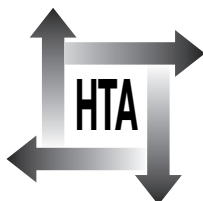
J Bamford, H Fortnum, K Bristow, J Smith, G Vamvakas, L Davies, R Taylor, P Watkin, S Fonseca, A Davis and S Hind



August 2007

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Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen

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The research reported in this monograph was commissioned by the HTA Programme as project number 03/05/01. The contractual start date was in October 2004. The draft report began editorial review in August 2006 and was accepted for publication in February 2007. As the funder, by devising a commissioning brief, the HTA Programme specified the research question and study design. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HTA editors and publisher have tried to ensure the accuracy of the authors' report and would like to thank the referees for their constructive comments on the draft document. However, they do not accept liability for damages or losses arising from material published in this report.

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Abstract

Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen

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Objectives: To describe and analyse in detail current practice of school entry hearing screening (SES) in the UK.

Data sources: Main electronic databases were searched up to May 2005.

Review methods: A national postal questionnaire survey was addressed to all leads for SES in the UK, considering current practice in terms of implementation, protocols, target population and performance data. Primary data from cohort studies in one area of London were examined. A systematic review of alternative SES tests, test performance and impact on outcomes was carried out. Finally, a review of published studies on costs, plus economic modelling of current and alternative programmes was prepared.

Results: The survey suggested that SES is used in most of England, Wales and Scotland; just over 10% of respondents have abandoned the screen; others are awaiting national guidance. Coverage of SES is variable, but is often over 90% for children in state schools. Referral rates are variable, with a median of about 8%. The test used for the screen is the pure tone sweep test but with wide variation in implementation, with differing frequencies, pass criteria and retest protocols; written examples of protocols were often poor and ambiguous. There is no national approach to data collection, audit and quality assurance, and there are variable approaches at local level. The screen is performed in less than ideal test conditions and resources are often limited, which has an impact on the

quality of the screen. The primary cohort studies show that the prevalence of permanent childhood hearing loss continues to increase through infancy. Of the 3.47 in 1000 children with a permanent hearing loss at school screen age, 1.89 in 1000 required identification after the newborn screen. Newborn hearing screening is likely to reduce significantly the yield of SES for permanent bilateral and unilateral hearing impairments; yield had fallen from about 1.11 in 1000 before newborn screening to about 0.34 in 1000 for cohorts that had had newborn screening, of which only 0.07 in 1000 were unilateral impairments. Just under 20% of permanent moderate or greater bilateral, mild bilateral and unilateral impairments, known to services as 6-year-olds or older, remained to be identified around the time of school entry. No good-quality published comparative trials of alternative screens or tests for SES were identified and studies concerned with the relative accuracy of alternative tests are difficult to compare and often flawed by differing referral criteria and case definitions; with full pure tone audiometry as the reference test, the pure tone sweep test appears to have high sensitivity and high specificity for minimal, mild and greater hearing impairments, better than alternative tests for which evidence was identified. There is insufficient evidence regarding possible harm of the screen. There were no published studies identified that examined the possible effects of SES on longer term outcomes. No good-quality published economic evaluations of SES were identified and a

universal SES based on pure tone sweep tests was associated with higher costs and slightly higher quality-adjusted life-years (QALYs) compared with no screen and other screen alternatives; the incremental cost-effectiveness ratio for such a screen is around £2500 per QALY gained; the range of expected costs, QALYs and net benefits was broad, indicating a considerable degree of uncertainty. Targeted screening could be more cost-effective than universal school entry screening; however, the lack of primary data and the wide limits for variables in the modelling mean that any conclusions must be considered indicative and exploratory only. A national screening programme for permanent hearing impairment at school entry meets

all but three of the criteria for a screening programme, but at least six criteria are not met for screening for temporary hearing impairment.

Conclusions: The lack of good-quality evidence in this area remains a serious problem. Services should improve quality and audit screen performance for identification of previously unknown permanent hearing impairment, pending evidence-based policy decisions based on the research recommendations. Further research is needed into a number of important areas including the evaluation of an agreed national protocol for services delivering SES to make future studies and audits of screen performance more directly comparable.



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List of abbreviations

AABR	automated auditory brainstem response	MMR	measles, mumps and rubella
ABR	auditory brainstem response	MRC	Medical Research Council
AOAE	automated otoacoustic emissions	NA	not applicable
ASHA	American Speech and Language Hearing Association	NHSP	Newborn Hearing Screening Programme
BACDA	British Association of Community Doctors in Audiology	NICE	National Institute for Health and Clinical Excellence
BHE	better hearing ear	NNHS	no neonatal hearing screen
CASP	Critical Skills Appraisal Programme	NPV	negative predictive value
CEAC	cost-effectiveness acceptability curve	NR	not reported
CI	confidence interval	NZHTA	New Zealand Health Technology Assessment
CRD	Centre for Reviews and Dissemination	OME	otitis media with effusion
df	degrees of freedom	PCHI	permanent childhood hearing impairment
DNA	did not attend	PCT	primary care trust
DPOAE	distortion product otoacoustic emissions	PPV	positive predictive value
ENT	ear, nose and throat	PSA	probabilistic sensitivity analysis
HB	health board	PT	pure tone
Hib	<i>Haemophilus influenzae</i> type b	PTA	pure tone audiometry
HL	hearing level	QALY	quality-adjusted life-year
ICER	incremental cost-effectiveness ratio	QUADAS	Quality Assessment of Studies of Diagnostic Accuracy
IDT	Infant Distraction Test	R&D	research and development
IQR	interquartile range	RCT	randomised controlled trial
LHSCG	local health and social care group	RICHHS	Regional Interactive Child Health System

continued

List of abbreviations continued

ROC	receiver operating characteristic	STARD	Standards for Reporting Studies of Diagnostic Accuracy
SD	standard deviation	SVEP	Sweep Visual Evoked Potential
SEM	school entry medical examination	TEOAE	transient evoked otoacoustic emission
SES	school entry hearing screen	TNHS	targeted neonatal hearing screen
SES-C	composite school entry screening	UNHS	universal newborn hearing screen
SES-PQ	SES using parental questionnaire	VASC	verbal audiometric screening for children
SES-PTS	SES using pure tone sweep testing	WHE	worse hearing ear
SES-SW	SES using spoken word tests	YNHI	year with no hearing impairment
SES-T	SES using tympanometry		
SHA	strategic health authority		

All abbreviations that have been used in this report are listed here unless the abbreviation is well known (e.g. NHS), or it has been used only once, or it is a non-standard abbreviation used only in figures/tables/appendices in which case the abbreviation is defined in the figure legend or at the end of the table.



Executive summary

Background

The ability to hear is important, particularly during children's formal education. Hearing impairment is amenable to intervention and hence a screening programme when children begin their school careers has potential value. School entry hearing screening (SES) has been implemented throughout the UK since the 1950s. There is evidence of mixed practice and uncertainty about the value of the screen. In addition, recent changes in childhood hearing screening policy (abandonment of a screen at 8 months and introduction of universal newborn screening) have implications for identification of children with hearing impairment at school entry.

Objectives

This report aimed to determine answers to the following three questions:

- What is current practice for the SES in the UK?
- What is known about the accuracy of alternative screening tests and the effectiveness of interventions?
- What is known about costs, and what is the likely cost-effectiveness of the SES?

Methods

A national postal questionnaire survey was addressed to all leads for the SES in the UK, considering current practice in terms of implementation, protocols, target population and performance data. Primary data from cohort studies in one area of London were examined. A systematic review of alternative SES tests, test performance and impact on outcomes was carried out. Finally, a review of published studies on costs, plus economic modelling of current and alternative programmes was prepared.

Results

The evidence from the national survey of current practice is that:

- the SES is in place in most areas of England, Wales and Scotland; just over 10% of respondents have abandoned the screen; others are awaiting guidance in the light of the national implementation of newborn hearing screening
- coverage of the SES is variable, but is often over 90% for children in state schools
- referral rates are variable, with a median of about 8%
- the test used for the screen is the pure tone sweep test but with wide variation in implementation, with differing frequencies, pass criteria and retest protocols; written examples of protocols were often poor and ambiguous
- there is no national approach to data collection, audit and quality assurance, and there are variable approaches at local level
- the screen is performed in less than ideal test conditions
- resources are often limited and this has an impact on the quality of the screen.

The evidence from the primary cohort studies is that:

- the prevalence of permanent childhood hearing impairment continues to increase through infancy
- of the 3.47 in 1000 children with a permanent hearing impairment at school screen age, 1.89 in 1000 required identification after the newborn screen
- the introduction of newborn hearing screening is likely to reduce significantly the yield of SES for permanent bilateral and unilateral hearing impairments; yield had fallen from about 1.11 in 1000 before newborn screening to about 0.34 in 1000 for cohorts that had had newborn screening, of which only 0.07 in 1000 were unilateral impairments
- just under 20% of permanent moderate or greater bilateral, mild bilateral and unilateral impairments, known to services as 6-year-olds or older, remained to be identified around the time of school entry.

The evidence from the systematic review of the alternative tests and of the effectiveness of interventions is that:

- no good-quality published comparative trials of alternative screens or tests for school entry hearing screening were identified
- studies concerned with the relative accuracy of alternative tests are difficult to compare and often flawed by differing referral criteria and case definitions; with full pure tone audiometry as the reference test, the pure tone sweep test appears to have high sensitivity and high specificity for minimal, mild and greater hearing impairments, better than alternative tests for which evidence was identified
- there is insufficient evidence to draw any conclusions about possible harm of the screen
- there were no published studies identified that examined the possible effects of SES on longer term outcomes.

The evidence from the cost-effectiveness study is that:

- no good-quality published economic evaluations of SES were identified
- a universal SES based on pure tone sweep tests was associated with higher costs and slightly higher quality-adjusted life-years (QALYs) compared with no screen and other screen alternatives; the incremental cost-effectiveness ratio for such a screen is around £2500 per QALY gained; the range of expected costs, QALYs and net benefits was broad, indicating a considerable degree of uncertainty
- targeted screening could be more cost-effective than universal SES
- lack of primary data and the wide limits for variables in the modelling mean that any conclusions must be considered indicative and exploratory only.

A national screening programme for permanent hearing impairment at school entry meets all but three of the criteria for a screening programme, but at least six criteria are not met for screening for temporary hearing impairment.

Conclusions

The lack of good-quality evidence in this area remains a serious problem. Services should improve quality and audit screen performance for identification of previously unknown permanent hearing impairment, pending evidence-based policy decisions based on the research recommendations.

Recommendations for research

Further research is highlighted in the following areas:

- evaluation of an agreed national protocol for services delivering the SES to make future studies and audits of screen performance more directly comparable
- development and evaluation of systems for data monitoring so that robust data on screen performance are available
- determination with greater certainty of the prevalence of congenital unilateral hearing impairment, and permanent mild and minimal hearing impairment at school entry, that could be identified by a suitable quality-assured screen protocol
- a comparison of the effectiveness, efficacy and efficiency of alternative approaches (reactive services, formal surveillance, targeted screening and universal screening at school entry age) to the identification of permanent hearing impairment postnewborn screen
- controlled studies of the effectiveness of hearing screening and subsequent interventions for later outcomes in children with permanent mild, minimal and unilateral hearing impairment identified at school entry
- determination of the distribution of detection thresholds for pure tones in the population at school entry.

Chapter I

Background and main questions

Historical background

There is a long history in the UK of screening for hearing impairment in childhood. By the 1930s hearing screening by various methods was being implemented at school entry, which at that time represented the most obvious point at which the child population was available for mass screening. In 1955, as simple screening audiometers became available, it was recommended that all children undergo school entry hearing screening using the pure tone 'sweep' test.¹ This test requires the child to indicate that he or she has detected each of a number of tones of different frequencies (itches) presented to each ear separately at an intensity level indicative of normal hearing. The screen quickly became established across the UK, organised and managed through local authority school health services. In 1976 the Court Report recommended that hearing screens be carried out at least twice in school.² However, there was no nationally agreed protocol for the screen, and implementation thus varied in small but possibly important details across services.

In the 1974 reorganisation of local government the school health service was brought into the NHS, and the school entry hearing screen (SES) has remained the responsibility of the NHS ever since, undertaken in the main by school nurses and community paediatricians. Evidence about the costs and effectiveness of the screen has remained elusive, as has clarity about its aims, despite a number of reviews.³⁻⁶ There are anecdotal reports that some services have supplemented the pure tone sweep test with other tests, while others are said to have abandoned the screen in the light of lack of national guidance and unwanted variability from a variety of sources, including screen protocols, test environment (schools can be noisy places), tester competence and equipment calibration. Despite this, there has until recently been a widespread if implicit consensus that the pure tone sweep test has value educationally and provides a safety net to catch any deficiencies of the earlier screening system in the overall public health provision,⁴ a position broadly endorsed by Hall.⁷

Haggard's⁴ comment on the deficiencies of the earlier screening system refers to the Infant

Distraction Test (IDT) screen. From the mid-1950s a hearing screen was performed on all infants in the UK aged 8 months using the IDT.⁸ This is a behavioural test in which sounds are presented to the infant under controlled conditions and the child's responses, if any, noted. However, this apparently simple test did not perform well, and there were credible reports of high referral rates, with high false-positive and false-negative rates. During the 1990s, developments in technology made it possible to test the auditory function of newborn babies using otoacoustic emissions and/or auditory evoked responses. A review in 1997⁵ led to a policy decision in England (with Wales, Scotland and Northern Ireland following suit) to phase out the IDT screen and to replace it with a national programme of newborn hearing screening. The newborn screen in England is a contingent two-test screen involving an automated otoacoustic emissions (AOAE) test followed, if either ear fails to show a clear response, by an automated auditory brainstem response (AABR) test, again requiring a clear response on both ears for a pass decision. The Newborn Hearing Screening Programme (NHSP) was fully implemented in England by March 2006; the evidence is that it is highly effective, reducing the age of identification of permanent congenital bilateral hearing impairment of moderate or greater degree from some 80 weeks to 10 weeks of age.^{9,10} The extent of the beneficial effects of this early identification for children with permanent hearing impairment upon developmental outcomes in general, and communication in particular, has been demonstrated,¹¹ although much detail remains to be added.

The nature of childhood hearing impairment

Hearing impairment in childhood can be permanent or temporary. Permanent childhood hearing impairment of a moderate degree or greater [i.e. detection thresholds >40 dB hearing level (HL) averaged across 0.5, 1, 2 and 4 kHz] is present at birth at a rate of about 1.6 per 1000 live births, of which approximately 1.0 in 1000 are bilateral impairments and 0.6 in 1000 are unilateral impairments.^{5,10} In terms of incidence,

this means that in the UK about 800 children per year will be born with permanent bilateral hearing impairment of a moderate or greater degree; and about 500 per year will be born with unilateral hearing impairment (i.e. hearing within normal limits in one ear, but hearing impairment of moderate or greater degree in the other ear). There is good evidence that the prevalence of permanent bilateral moderate or greater hearing impairment increases through the first decade of childhood.^{12,13} The reasons for this are not entirely clear, but include meningitis, measles and other causes of acquired impairment; progression of unilateral to bilateral impairments; and late-onset/progressive impairments linked to prenatal or perinatal infection or to hereditary factors. It is possible that the prevalence of bilateral moderate or greater impairment reaches 2 in 1000 by the age of about 9 years.

The evidence on permanent unilateral hearing impairment is more limited. Although it appears from NHSP data that the prevalence at birth is about 0.6 in 1000, it is not known whether there are significant numbers of later onset cases, whether some of the impairments are progressive, and whether there is a tendency for congenital or postnatal unilateral hearing impairment to progress to bilateral impairment. Unilateral hearing impairment would be expected to affect auditory perception in various predictable ways, such as poor localisation of sound sources, and difficulty in noisy or reverberant environments such as schools, and there is some evidence of detrimental effects on academic progress.¹⁴ Unlike bilateral permanent childhood hearing impairment however, management of unilateral impairment remains uncertain, and it is not known whether early family advice and support, a hearing aid in the affected ear or other approaches would be helpful.

The significance of the increase in prevalence in permanent hearing impairment in the first decade of life is that a newborn hearing screening programme would fail to detect these additional cases and other processes are required, whether based on professional responsiveness to parental observations, structured surveillance, or later screening. In addition, the newborn screen as presently conceived will not identify mild and minimal permanent hearing impairment, whereas a later screen could.

In children, temporary hearing impairment is much more common than permanent hearing impairment. It is linked in the main to colds and

upper respiratory tract infections that lead to otitis media with effusion (OME) – the presence of fluid in the middle ear. There is a huge literature on OME (see Haggard and Hughes 1991¹⁵ for an early but comprehensive review), which addresses prevalence, pathology, assessment, management options, the time-course, and short-, medium- and long-term effects of the condition. The point prevalence of OME is of the order of 15–25% in the 0–6-year-old age group, with peaks in the first year of life and at school entry.^{15–17} The period prevalence across that age range may be as high as 80%. Most cases resolve within 2–3 months. Some recur, and some persist for much longer. In those that do, there can be significant short- and medium-term effects not only on hearing, but also on behaviour, socialisation, speech and academic progress. The difficulty for services is to be able to identify those cases, perhaps around 3 or 4%, that are likely to have the condition recurrently, and/or with a persistence and severity likely to cause concern (i.e. to affect significantly development, whether attention, communication, behaviour or other domains). Although case finding is done through hearing impairment, intervention options may be directed at other effects, and include advice for parents and teachers, speech and language therapy, and/or surgery to remove the fluid and decrease the chance of recurrence [myringotomy, ventilation tubes (grommets), adenoidectomy].

Main questions and overall design of the study

Despite several attempts, some of which are recent,^{5,6} to investigate the value and effectiveness of the SES in the UK, there remains a pressing need to understand the pattern of current practice (which has developed ‘bottom-up’) and to evaluate the likely accuracy of alternative tests and costs and effectiveness of the screen to guide policy decisions. Furthermore, the introduction of newborn hearing screening to replace the underperforming 8-month hearing screen has significantly changed the landscape of hearing screening in childhood. Whereas the 8-month IDT screen was very unlikely to identify permanent unilateral hearing impairment, the newborn screening protocol will identify unilateral as well as bilateral permanent hearing impairment; this potentially reduces one of the justifications for the SES. However, the SES might continue to yield significant numbers of mild, high-frequency or late-onset/progressive impairments that would otherwise be missed (or missed until later

concerns led to identification), and might identify significant numbers of children with persistent middle ear disorders not otherwise known to services at a time when good hearing is of particular importance educationally.

These uncertainties lie behind the current research study. The authors were commissioned in 2004 by the HTA Programme of the NHS Research and Development initiative to

- carry out a national survey of current SES practice
- conduct a systematic review of the accuracy of alternative screening tests and the effectiveness of subsequent interventions
- assess the costs of the screen and to model cost-effectiveness.

The study therefore has three strands:

- Strand 1 (reported in Chapter 2) is a national questionnaire survey of current practice. A survey instrument was designed and piloted, and the final version completed by lead clinicians/managers for the SES across the UK. The questionnaire was designed to collect information on whether the screen was still being implemented, what tests and protocols were being used, what the target population was, who performed the screen and where, with what training and what equipment, and whether any screen performance data were collected and available.
- Strand 2 (reported in Chapter 4) is a systematic review of the existing research literature which aims to evaluate the accuracy of alternative approaches to a school entry hearing screen, and to summarise the evidence on screen performance (i.e. screen uptake and yield for different screen options and different case definitions) and the impact of the screen on children's outcomes (language, communication, social and educational).
- Strand 3 (reported in Chapter 5) is an assessment of the costs, outcomes and associated levels of uncertainty of alternative models of school entry screening using economic modelling techniques.

Throughout the three strands, cognisance is taken of three distinct possible case definitions: first, children with moderate, severe or profound bilateral permanent hearing impairment, for whom the evidence on the consequences of not identifying and intervening appropriately is strong; secondly, children with permanent mild,

minimal or unilateral hearing impairment, or hearing impairment affecting only some frequencies, about which the evidence on consequences, intervention (and prevalence) is less clear; and thirdly, children with temporary hearing impairment associated with persistent and/or severe OME, the treatment for and sequelae of which give rise to considerable controversy.¹⁸ One reason for this is that while permanent hearing impairment has a variety of causes, the treatment and management are in the main directed at the hearing impairment itself; for children with OME, however, although there is a coherent set of disease processes at its core about which something is known, hearing impairment is by no means the whole story, and interventions aim to treat more than just the hearing impairment.

Since the evidence base for the characteristics and yield of screening for the latter two categories is so weak, the study also considers a case definition in terms of a disability measure for which there is some evidence, hearing in noise, or specifically the minimum signal to noise ratio required to score at a given criterion level on a speech perception task.

When gathering evidence on the effectiveness and the efficiency of the SES for each of these case definitions, it is important in the cost-effectiveness modelling (strand 3) to take account of the likely incremental yield of the screen: for this one needs to know, or to be able to estimate, the number of cases that remain to be identified by the SES after the identification of these cases by the newborn hearing screen, standard surveillance, parental concern and professional responsiveness. As well as referring to the published evidence on prevalence of cases and yields of screens and systems before school entry, the researchers examined primary data from cohort studies to which they have direct access through authorship (Chapter 3). One of these (from Watkin) was a series of studies undertaken in Waltham Forest where universal newborn screening was introduced some 10 years ago (so the outcomes from recent SES cohorts would be expected to reflect the effect of newborn screening on cases left to be found), and the other has been the use of a large database of children with persistent OME from the Medical Research Council (MRC) Otitis Media Study Group to answer some specific questions thrown up by the discussions and to address the issue of the effectiveness of subsequent interventions for OME.

The authors decided at an early stage not to review the literature on the effectiveness of

treatment of OME fully or in a concentrated way, for two main reasons. First, it is very large and heterogeneous, with much of it of poor quality, giving some information but not of the type that can be easily extracted from aggregating the results of the studies of best quality. Secondly, as noted above, hearing is neither the only relevant nor necessarily the ultimate outcome of such treatment. Nevertheless, since there is a high continuing risk of two recent trials particularly relevant to screened caseloads continuing to be misinterpreted in relation to other evidence (arising from the generally poor understanding of the importance of the characteristics of populations selected and the economic pressures

in differing health systems), the interpretation and implications of these particular studies are discussed.

Finally, Chapter 6 outlines the strengths and weaknesses of the study, summarises the findings from each strand, draws together the evidence on screening at school entry in the UK from all three strands into a series of conclusions and makes recommendations. The conclusions are used to examine the justification for the SES as a route to identifying children with permanent and temporary hearing impairment in the light of the National Screening Committee's criteria for screening programmes (Appendix 1).

Chapter 2

National survey of current screening practice

Introduction

Scientific background

A national survey of paediatricians responsible for the SES in the UK⁶ confirmed clinical impressions that services had evolved such that there was considerable variation between services in terms of programme organisation, pass/fail criteria, case management and screen performance, despite recommendations to retain and standardise the SES.¹⁹ At the time of that survey of 96 services, four services had discontinued their SES programme as a result of local audit.²⁰ In programmes where the SES was not carried out by dedicated screeners, competing programmes, such as immunisation, took priority over the SES, affecting its performance. There was also an awareness that information may have been incomplete as there were no available data from services that may have been provided by non-medical service leads.

In recent years consideration has been given to the impact on individual children of unrecognised mild, unilateral or temporary hearing impairment^{21,22} and the possible need to identify and manage these children. There is also growing debate about the likely impact of newborn hearing screening on the yield of new cases from the SES and about cases that will not be detected by the NHSP.¹² Evidence is emerging that services will undergo further revision on an ad hoc basis as a result of the changing pattern in the yield from the SES following the introduction of newborn hearing screening and local variation in the epidemiology of hearing impairment (see Chapter 3).

This chapter provides an up-to-date account of current practice and performance of the SES in the UK.

Aims

The aims of this study were to describe and analyse in detail current practice of the SES throughout the UK, in order to:

- quantify variability in screening practice nationally
- evaluate current screen performance as reported by service leads in terms of screen coverage, referral rates and yield
- record the views of SES leads regarding the value of the screen together with their ideas for improvements or alternatives.

Methods

Ethics and NHS Research and Development Approval

The study met the criteria for a multicentre study with no local investigators. Application for full ethical approval for the UK was submitted to the Central Manchester Local Research Ethics Committee. Local research and development (R&D) approval was applied for in all primary care trusts (PCTs) in England, NHS acute trusts which employed an SES lead clinician, the primary care arm of each health board (HB) in Scotland, the NHS trusts in Wales and local health and social care groups (LHSCGs) in Northern Ireland. Some R&D departments for the PCTs are grouped into consortia with administrative responsibility for a number of PCTs, varying from two to 15. Applications for approval were made to 124 departments.

Identification of service leads for SES provision

Service delivery of the SES varies across the UK in terms of the organisations responsible for coordinating the programme and employing staff who undertake it. It was therefore necessary to use several lines of enquiry to identify and recruit the service leads.

- Letters were sent to all members of the British Association of Community Doctors in Audiology (BACDA) asking them to contact the research team if they were responsible for the SES in their area or, if they were not, to return the name of the responsible person if they knew it.
- The Directory of Community Nursing 2004/2005²³ was used to identify school nursing departments.
- Advertisements were placed in the BACDA newsletter and British Society of Audiology newsletter.
- Oral and poster presentations were made to the 28th Annual Children's Hearing Screening Conference.
- 'Cold calls' were made to NHS trusts.

Development of a postal questionnaire/survey instrument

A postal questionnaire (Appendix 2) was developed to establish:

- the target population of children who are routinely entered in the SES programme
- locations and conditions under which the screen is performed
- test methods used
- pass/fail criteria
- who carries out screening tests and equipment used for screening
- data management systems used
- coverage, referral rates and yield of the screen
- views of the SES leads regarding the screen.

The questionnaire was reviewed by the project's advisory committee and piloted by seven audiology professionals closely related to the running of the SES in their area. These professionals were identified either via BACDA or through contacts known to members of the research group. Changes were made following the pilot and the questionnaire was finalised.

Data collection

Questionnaires were posted to all identified service leads between September and November 2005 with a covering letter giving further details about the study and a reply-paid envelope. If the service lead failed to return the completed survey within 4 weeks a reminder letter and an additional copy of the questionnaire were posted to them. Those leads failing to respond within an additional 2 weeks were contacted by telephone. A final telephone reminder was made after a further 2 weeks and if, at this stage, no reply was received non-response was assumed.

Data were entered into an Access database by one researcher (KB) and 10% of questionnaires were entered independently by another (HF) to check for errors in data entry. Data were converted to Excel for analysis and possible completion errors checked by identifying outlier values.

Results

Ethics and NHS research and development approval

The Central Manchester Local Research Ethics Committee granted final approval for the study in May 2005.

In England, Scotland and Wales 124 R&D departments were approached for approval, covering a total of 304 NHS trusts. Owing to difficulties in identifying the relevant departments, R&D applications could be made to only two out of 15 LHSCGs within Northern Ireland. Six R&D departments covering seven NHS trusts in England did not give approval for the following reasons:

- One required a separate consent sheet with the questionnaire and information letter.
- Two required full Criminal Records Bureau and locally administered occupational health checks for the principal investigator to secure an honorary contract.
- Three approvals were still pending at the time of writing.

Questionnaires were not posted to any staff employed by these seven trusts.

Response

In the UK, 244 services responsible for the SES were identified. This does not match the number of primary care organisations because it was common for services to cover a geographical area encompassing more than one primary care organisation; that is, the PCT/LSHCGs/Welsh and Scottish HB boundaries did not match the SES service boundaries.

Questionnaires were sent to 229 service leads and 195 (85.2%) responded (*Table 1*).

The numbers of PCTs that were covered by a returned questionnaire within each of the 28 strategic health authorities (SHAs) in England are shown in *Table 2*.

In only two of the 28 SHAs the survey failed to achieve 70% coverage of PCTs. Overall, 86.5% of PCTs in England are represented in the survey.

Within Scotland questionnaires were returned from ten SES services covering ten out of 15 (66.7%) HBs. In Wales eight SES services returned a questionnaire, covering 16 out of 22 (72.7%) local health boards. In Northern Ireland, one SES service covering one out of 15 LHSCGs returned a questionnaire.

Questionnaire findings

The descriptive results for each question in the questionnaire are detailed. The responses from each of the countries have been grouped together to represent the response for the UK as a whole.

TABLE 1 Number of SES services identified in each country, number sent a questionnaire and number responding

	Services				
	No. identified	No. sent a questionnaire (contacted)	Response		
			<i>n</i>	% of identified	% of contacted
England	208	201	176	84.6	87.6
Scotland	15	15	10	66.7	66.7
Wales	11	11	8	72.7	72.7
Northern Ireland	10	2	1	10.0	50.0
Total	244	229	195	79.9	85.2

TABLE 2 Geographical coverage by PCTs in England

SHA	PCTs in each SHA	
	No. within the SHA	No. (%) covered by a returned questionnaire
Avon, Gloucestershire & Wiltshire	12	12 (100)
Bedford and Hertfordshire	11	6 (54.5)
Birmingham and the Black Country	12	9 (75)
Cheshire & Merseyside	15	13 (86.7)
County Durham & Tees Valley	10	10 (100)
Coventry, Warwickshire, Herefordshire & Worcestershire	8	8 (100)
Cumbria & Lancashire	13	12 (92.3)
Dorset & Somerset	9	8 (88.9)
Essex	13	12 (92.3)
Greater Manchester	14	13 (92.9)
Hampshire and Isle of Wight	10	9 (90)
Kent and Medway	9	8 (88.9)
Leicestershire, Northamptonshire & Rutland	9	8 (88.9)
London North Central	5	5 (100)
London North East	7	5 (71.4)
London North West	8	8 (100)
London South East	6	5 (83.3)
London South West	5	4 (80)
Norfolk, Suffolk and Cambridgeshire	17	14 (82.3)
North and East Yorkshire and Northern Lincolnshire	10	6 (60)
Northumberland, Tyne and Wear	6	6 (100)
Shropshire and Staffordshire	10	9 (90)
South West Peninsula	11	10 (90.9)
South Yorkshire	9	8 (88.9)
Surrey and Sussex	15	12 (80)
Thames Valley	15	14 (93.3)
Trent	19	14 (73.7)
West Yorkshire	15	14 (93.3)
Total	303	262 (86.5)

In all cases 'n' refers to the numbers of service leads indicating that response. Unless otherwise indicated, percentages are of the total number (*n*) of services responding to that question. Missing data are tabulated separately as a percentage of 195 responses. When relevant, the missing values include the ten services who reported that they no longer run an SES (see next subsection).

Population entered into the SES programme

Twenty-four services (12.2%) no longer run a universal school entry hearing screen; 11 run no screen and 13 implement a targeted screen. Ten services gave reasons for not running a screen at all, including resource limitations (five) and low yield (six). Only two services running a targeted screen gave reasons, both resource limitations. All

TABLE 3 The extent to which children are routinely entered into the school entry hearing screen (question 1)

	All	Some	None	Total responses	Missing <i>n</i> (% of 195 responses)
Children in state schools	170 (87.6)	13 (6.7)	11 (5.7)	194 (100)	1 (0.5)
Children in private schools	37 (20.4)	52 (28.7)	92 (50.8)	181 (100)	14 (7.2)
Children who are home educated	10 (5.7)	39 (22.4)	125 (71.8)	174 (100)	21 (10.7)
Children in special schools with known physical or sensory disability	85 (47.2)	44 (24.4)	51 (28.3)	180 (100)	15 (7.7)
Children in special schools with known mental disability (excluding those with hearing loss)	79 (44.4)	46 (25.8)	53 (29.8)	178 (100)	17 (8.7)
Children known to have hearing loss	37 (21.3)	60 (34.5)	77 (44.3)	174 (100)	21 (10.8)
Other	2	7	1	10	

Data are shown as *n* (%).

but two of the remaining 172 services screen all children in state schools. *Table 3* indicates the extent to which the SES is offered to children in different educational environments.

Only 20.4% (37/181) of services screen all children in private schools. An additional 28.7% (52/181) screen some such children usually when requested to do so. Services that do not routinely screen children in private schools commented that they did perform screening when the school requested it (*n* = 31), but others said that resources prevented them giving full coverage (*n* = 10). Several said that private schools have their own arrangements for screening. The proportion of children in the UK attending private schools is up to 7%.²⁴ One service that does not run a universal SES at all does carry out screening in two private schools on a fee-for-service basis. This was reported to be because the private schools insisted on having a screen and the service therefore provided it to make sure that it was conducted properly.

It was found that 55.5% (97/174) of services screen all (21.3%, 37/174) or some (34.5%, 60/174) children who are already known to have a hearing loss. (Note that the term 'hearing loss' was used throughout the questionnaire and has therefore been used in this chapter when reporting results.) One reason given for screening children known to be hearing impaired was that doing so was beneficial so as not to exclude children in mainstream education from a whole class activity. Eighteen services also stated that they would not be aware of the child's hearing status prior to screening unless the child wore a hearing aid.

Most comments supporting not screening children with a known hearing loss referred to the fact that such children all had full audiology cover. Only two services specifically stated that screening a child with known hearing loss would have no value.

Children who are educated at home are not screened by 71.8% (125/174) of services. Many services (22/63 commenting) were unaware of home-educated children, but a further 28 claimed to screen such children if they were requested to do so. One service commented that they were "not responsible for children educated at home through parental choice".

The most common reason given for not screening children attending special schools was that such children all receive full audiological testing routinely, or are looked after by specialist services.

Additional comments recognised that some children not in state schools would be missed by the service provided, but that responsive services were available. Some services were actively addressing this issue.

Table 4 details the arrangements made to screen children who did not attend the first scheduled screen.

Children miss the screening opportunity offered in schools for a variety of reasons, including illness, periods of time spent out of the country, travelling families, lack of parental consent and transfer into the school after the screen. The majority of

TABLE 4 The arrangements in place to screen children for whom consent is obtained but who did not attend the screen for any reason (e.g. through school absence, had a cold) (question 2)

	All of the time	Most of the time	Some of the time	Rarely	Never	Total responses	Missing n (% of 195 responses)
Revisit to the school	135 (76.7)	26 (14.8)	13 (7.4)	1 (0.6)	1 (0.6)	176 (100)	19 (9.7)
Appointment arranged at school health clinic	8 (6.0)	4 (3.0)	34 (25.6)	25 (18.8)	62 (46.6)	133 (100)	62 (31.8)
Appointment arranged at audiology clinic	6 (4.3)	5 (3.6)	28 (20.3)	46 (33.3)	53 (38.4)	138 (100)	57 (29.2)
No arrangement made	–	–	2 (1.7)	14 (12.1)	100 (86.2)	116 (100)	79 (40.5)
Other ^a	2	–	1	2	–	5	–

Data are shown as n (%).

^a Retest in school next year (2), next term (1); direct referral to doctor if concern (1); referral to school nurse audiology clinic (1).

TABLE 5 The school year in which the school entry hearing screen is routinely performed (question 3)

	n (% of 181 responses ^b)
Preschool	1 (0.6)
Reception/primary 1 ^a	160 (88.4)
Year 1/primary 2 ^a	48 (26.5)
Year 2/primary 3 ^a	6 (3.3)
Other	19 (10.5)

^a Primary 1, 2 and 3 refer to the system in Scotland and are equivalent to the year numbers in the English system.

^b Forty-three responses indicated multiple (two or three) answers.

services (91.5%, 161/176) make arrangements to screen non-attendees during a revisit to the school and 13.8% (16/116) of services say they rarely or sometimes make no arrangement to screen.

Respondents indicated that they would screen children in later school years if necessary and that the procedures in place for screening such children usually involved recall to the community clinic.

The majority of screening (88.4%, 160/181) is performed in the first year of primary school (Table 5). Screening occurring at other times included screening a child in any school year if they were new to the school and had no evidence of undergoing a previous hearing screen; 'responsive screening' at any time if concerns were raised about a child's hearing; and annual screening of special cases such as children with visual impairment or Down's syndrome.

Commenting on the timing of the screen, respondents referred to the conflict that needs to be considered between screening early (in the reception year) to ensure early identification of any problems versus screening later (in year 1) to maximise the child's maturity and ability to perform the tests and thereby reduce the referral rate.

Screening procedure

An important aspect of any screening programme is the information provided to, and consent received from, those covered by the screen. Table 6 lists the documentation available: 123 services were able to provide documentation.

Four services used an opt-out system for consent and several others used a global consent for the wider school health check to include consent for hearing screening.

Although all respondents reported screening within the school most or some of the time (Table 7) it is notable that comments made highlighted that the conditions under which the screen was frequently performed are very variable (Table 8) and can be problematic, and that suitable conditions are sometimes difficult to identify. Seven respondents said that they would refuse to screen in unsuitable conditions. No services routinely use a sound-treated booth or room (one did use a sound-treated van), but most services operate the screen in 'quiet' areas of the school.

Test methods

Services were fairly equally divided in whether they always implemented the SES as a stand-alone screen or incorporated it into a wider health check

TABLE 6 The extent to which written documentation concerning the SES is available (question 4)

	Yes	No	Total responses	Missing <i>n</i> (% of 195 responses)
Parent/guardian agreement for the screen	151 (85.3)	26 (14.7)	177 (100)	18 (9.2)
Information provided to the parent/guardian prior to screening	124 (71.3)	50 (28.7)	174 (100)	21 (10.8)
Information provided to the parent/guardian prior to referral	142 (83.5)	28 (16.5)	170 (100)	25 (12.8)
Test protocol	137 (85.1)	24 (14.9)	161 (100)	34 (17.4)
Retest protocol	129 (80.1)	32 (19.9)	161 (100)	34 (17.4)
Referral protocol	133 (84.2)	25 (15.8)	158 (100)	37 (19.0)

Data are shown as *n* (%).

TABLE 7 The location of the first test within the school entry hearing screen (question 5)

	All of the time	Most of the time	Some of the time	Rarely	Never	Total responses	Missing <i>n</i> (% of 195 responses)
School	156 (86.2)	25 (13.8)	–	–	–	181 (100)	14 (7.2)
Community clinic	–	4 (3.1)	19 (15.0)	55 (43.3)	49 (38.6)	127 (100)	68 (34.9)
Home	–	–	3 (2.4)	39 (31.5)	82 (66.1)	124 (100)	71 (36.4)
GP clinic	–	–	2 (1.6)	9 (7.4)	111 (91.0)	122 (100)	73 (37.4)
Other	–	–	2	1	1	4	

Data are shown as *n* (%).

TABLE 8 The conditions under which the school entry hearing screen is performed (question 6)

	All of the time	Most of the time	Some of the time	Rarely	Never	Total responses	Missing <i>n</i> (% of 195 responses)
Soundproof booth	–	–	–	1 (0.7)	140 (99.3)	141 (100)	54 (27.7)
Sound-treated room	–	–	3 (2.1)	14 (9.9)	125 (88.0)	142 (100)	55 (28.2)
Quiet office	18 (10.8)	87 (52.4)	54 (32.5)	4 (2.4)	3 (1.8)	166 (100)	29 (14.9)
Noisy office	–	4 (2.8)	49 (34.0)	36 (25.0)	55 (38.2)	144 (100)	51 (26.2)
Quiet classroom/area	16 (9.6)	48 (28.9)	65 (39.2)	19 (11.4)	18 (10.8)	166 (100)	29 (14.9)
Noisy classroom/area	–	6 (4.1)	31 (21.2)	27 (18.5)	82 (56.2)	146 (100)	49 (25.1)
Other ^a	2	3	17	2	–	24	171 (87.7)

Data are shown as *n* (%).

^a Twenty-four services mentioned 14 different areas of the school: medical room (15), staff room (10), library (4), corridor (3), stationery cupboard/store room/broom cupboard (4), main hall (2), sound-treated van, kitchen, hall, toilet, entrance area, head's office (1 each).

(Table 9), but if those who answered 'most of the time' are included, 72.8% (115/158) incorporate screening as part of a wider health check, compared with 52.7% (70/133) who screen for hearing loss on a separate occasion.

The estimates of children that could feasibly be screened in 1 day were very variable (Table 10) and were said to depend on many practical factors, including:

- child:
 - child's understanding and cooperation
 - ease of testing
 - attendance rates
- administration:
 - mistake-free administration
 - ability to manage workload
- staff:
 - numbers
 - skills

TABLE 9 The extent to which children at school entry are screened for hearing loss only or for hearing loss as part of a wider health check (question 9)

	All of the time	Most of the time	Some of the time	Rarely	Never	Total responses	Missing n (% of 195 responses)
Screen for hearing loss only	65 (48.9)	5 (3.8)	19 (14.3)	24 (18.0)	20 (15.0)	133 (100)	62 (31.8)
Screen for hearing loss as part of a wider health check	95 (60.1)	20 (12.7)	5 (3.2)	5 (3.2)	33 (20.9)	158 (100)	37 (19.0)

Data are shown as n (%).

TABLE 10 Estimates of the numbers of children (minimum, average and maximum) that could be screened under normal circumstances during the course of a one-day visit to a school (question 10)

Number of children	No. of responses	Lowest response ^a	Highest response	Median response
(a) When screening for hearing loss only				
Minimum	94	1	45	10
Average	92	2	60	33
Maximum	93	5	110	40
(b) When screening for hearing loss as part of a wider health check				
Minimum	96	1	20	5
Average	99	2	55	14
Maximum	100	3	90	20

^a One service reported only testing one child per day.

- school:
 - size and location (several small rural schools entailing greater time spent on travelling)
 - support provided by schools
 - level of disturbance
 - other activities for children
 - experience of school nurses
- available time:
 - length of session
 - travelling time
 - number of schools visited.

Not all services screen children for a whole day. Some run the screening in the morning and do the administration and paperwork in school in the afternoon; others attend schools only for half a day at a time. Where services stated the time taken to screen a child, estimates varied from nine per hour to 20 minutes per child.

The majority of services (71.7%, 124/173) implement a two-test screen, with only 16.8% (29/173) referring after a single test (Table 11).

TABLE 11 The number of tests routinely performed within the screening programme before onward referral (question 7)

No. of tests before referral	n (%)
1	29 (16.8)
2	124 (71.7)
3	18 (10.4)
4	2 (1.2)
Total	173 (100)
Missing responses	22 (11.3)

Pure tone (PT) sweep testing is used by 97.1% (170/175) of services as the first test. Thirty-one services (18.1%) add in pure tone audiometry (PTA) and/or tympanometry and/or otoscopy at the second test (Table 12).

When a two-test screen is carried out, the time between tests varies from doing both tests on the same day up to an interval between tests of more than 12 weeks (Table 13).

The majority of referrals are made to 'audiology', but details of whether this was a second tier

TABLE 12 The test methods used within the screening programme (question 7)

Test no.	PTA	PT sweep	Tympanometry	Otoscopy	Total responses	Missing n (% of 195 responses)
1	4 (2.3)	170 (97.1)	1 (0.6)	1 (0.6)	175 (100)	22 (11.3)
2	29 (20.3)	112 (78.3)	1 (0.7)	1 (0.7)	143 (100)	52 (26.7)
3	16 (72.7)	5 (22.7)	1 (4.5)	–	22 (100)	173 (88.7)

Data are shown as n (%).

TABLE 13 The time between tests for services implementing more than one test (question 7)

Between tests	Same day	2 to <6 weeks	6–12 weeks	>12 weeks	Total responses	Missing n (% of 195 responses)
1 and 2	11 (8.3)	56 (42.4)	59 (44.7)	6 (4.5)	132 (100)	63 (32.3)
2 and 3	5 (41.7)	3 (25.0)	4 (33.3)		12 (100)	183 (93.8)

Data are shown as n (%).

TABLE 14 Service/professional to which onward referral is made on failure of the screen (question 7)

Referral to	n (%)
Audiology/hearing clinic	127 (75.1)
GP	15 (8.8)
Children's hearing assessment clinic	7 (4.1)
Community paediatrician/medical officer	6 (3.6)
ENT	3 (1.8)
Community/hearing/school nurse clinic	11 (6.5)
Total responses	169 (100)
Missing responses	26 (13.3)

ENT, ear, nose and throat.

community clinic or a tertiary audiology clinic were not given. Fifteen services (8.8%) refer the child to their GP (Table 14).

Pass/fail criteria

The test levels at which services decide a child should be retested or referred varied widely with many combinations of levels and frequencies. The details for the first test are shown in Table 15. One-hundred and forty of the 195 services consistently screened at 20 or 25 dB for 1, 2 and 4 kHz, with a variety of other levels and frequencies added to that base.

One-hundred and thirty-five services gave levels at which the child would be said not to have passed the test at test 2. Of these, only 30 were different from the levels at test 1. Seven added a

frequency, 12 increased the level, one decreased the level, nine tested to threshold, and one included fails at tympanometry and otoscopy (unspecified).

Twenty-one services gave levels at which the child would be said not to have passed the test at test 3. Two added a fail at tympanometry, six tested to threshold, one decreased the level, five increased the level and one added observation of behaviour.

Screen personnel and equipment

Staff from different professional backgrounds are involved in carrying out the SES in different parts of the country (Table 16). Of the respondents, the majority of screeners are school nurses (66.3% of services) or their assistants (18.5% of services).

Many services emphasised the importance of training for staff undertaking the SES. A general theme emerged of staff less qualified in audiology undertaking the initial screen(s) followed by referral when necessary to audiological qualified staff. Resource issues were cited as the reason for using 'cheaper' staff, including one instance of 'mumsy' ladies who know the schools and children. In contrast, the benefit of employing highly qualified staff, which meant fewer unnecessary referrals, was also mentioned.

The majority of services (94.3%) use screening audiometers (Table 17).

TABLE 15 Pass/fail criteria (levels and frequencies) (question 8)

Screening level and frequencies	No. of responses (%)
20 dB at 1, 2 and 4 kHz	51 (29.3)
No other frequencies	6
+ 20 dB at 500 Hz only	16
+ 25 dB at 500 Hz and/or 250 Hz	13
+ 30 dB at 500 Hz only	7
+ 20 dB at 500 Hz and/or at 8 kHz and/or at 6 kHz and/or at 250 Hz	4
+ 30 dB at 250 and 500 Hz and 8 kHz	2
All other frequencies (not specified) at 20 dB	3
25 dB at 1, 2 and 4 kHz	89 (51.1)
No other frequencies	10
+ 25 dB at 500 Hz only	37
+ 30 dB at 500 Hz and/or 250 Hz and/or 6 kHz	29
+ 25 dB at 500 Hz and 8 kHz and/or 250 Hz and/or 6 kHz	9
All other frequencies (not specified) at 25 dB	4
30dB at 1, 2 and 4 kHz	15 (8.6)
No other frequencies	1
+ 30 dB at 500 Hz and 8 kHz and/or 250 Hz	8
+ 35 dB at 500 Hz only	2
All other frequencies (not specified) at 30 dB	4
Varying between 20 and 30 dB at 1, 2 and 4 kHz	19 (10.9)
No other frequencies	4
+ 25–35 dB at 500 Hz only	10
Other combinations	5
Total response	174 (100)
Missing or unclassifiable	21 (10.8)

TABLE 16 Staff who perform the SES (question 11)

	n (% of 178 responses ^a)
School nurse	118 (66.3)
School nurse assistant	33 (18.5)
Nursery nurses	31 (17.4)
Health care assistants/support workers/school health assistants	29 (16.3)
Audiometrician	28 (15.7)
Audiologist/assistant audiologist	18 (10.1)
Technicians	14 (7.9)
Screeners	11 (6.2)
School doctor	5 (2.8)

^a Eighty-eight services mentioned more than one grade of staff.

TABLE 17 Types of equipment and numbers used within the SES (question 12)

	n	Total responding	No. owned
a. Screening audiometer	164 (94.3)	174	1–48
b. Diagnostic audiometer	30 (29.1)	103	1–24
c. Screening tympanometer	23 (21.9)	105	1–6
d. Diagnostic tympanometer	8 (8.33)	96	
Other ^a	7		

^a Auroscope (2), otoacoustic emissions test for special schools (1), Quick tympanometer/tympanometer (1), sound level meter (2), otoscope (1).
Combinations: screening audiometer and screening tympanometer (a + c) = 12; screening and diagnostic audiometer (a + b) = 17; screening and diagnostic audiometer and screening tympanometer (a + b + c) = 8.

TABLE 18 Number of services with a recently performed audit of the SES, data management systems in place, and easily obtainable reports (questions 13, 14 and 15)

	<i>n</i>	Total responses	Missing responses
Recent audit	16 (9.5)	168 (100)	27 (13.8)
Data management system	112 (69.6)	161 (100)	34 (17.4)
Data reports easily obtainable	52 (46.4)	112 (100)	83 (42.6)

Data are shown as *n* (%).

TABLE 19 Coverage and referral rates for the SES (question 16)

	No. of service leads providing reliable data	Children				
		Mean		Median	Range	
		<i>n</i>	<i>n</i>	%	<i>n</i>	%
Children eligible for the screen	55	2704	2229	–	352–10,291	–
Children undergoing the screen	55	2512	2176	95.2	234–9483	56.3–100.0
Children referred for further assessment after failing the screen	46	246	168	7.9	23–1977	1.91–23.4

Several services commented that all equipment, is calibrated annually and that each member of staff had their own equipment, but others referred to equipment that was very old and the inability to replace equipment owing to resource limitations. The use of otoscopes only by staff qualified to use them and the need for explicit parental consent for an invasive procedure were mentioned as reasons for not using them.

Audit data

Services were asked whether an audit of the SES had taken place in the last 2 years, whether they used any sort of data management system and, if so, whether reports were easily obtainable (Table 18).

Less than 10% of services (16/168) have performed any audit of their service in the last 2 years. Approximately 70% (112/161) of services use some sort of data management system for the SES, but only half of those can easily obtain data reports from it.

Coverage, referral rates and yield

Table 18 indicated that few services were able to provide accurate data on coverage and referral rates from the SES. Table 19 indicates the data considered by respondents to be reliable. Data that were estimated or guessed by services have not been included.

Fifty-five respondents (28.2% of 195) provided data on children eligible for the screen from which coverage figures could be calculated. The percentage of eligible children who were screened (coverage) ranged from 56.3 to 100%, with a median of 95.2% (mean 91.1%). Nearly three-quarters of services (74.5%) achieved more than 90% coverage.

Forty-six respondents provided data from which referral rates overall could be calculated. The percentage of screened children who were referred for further assessment ranged from 1.91 to 23.4% with a median of 7.9% (mean 7.7%). Eleven of the 57 services (19.3%) gave values of zero for referrals.

The pattern of pass/fail criteria for these 46 services did not differ from the pattern for the services for which data are presented in Table 15. Data were unavailable on the number of children referred at different stages of the screening programme.

Comments made by respondents reflected the poor data management of the SES in general. In particular, several services commented that data were available on numbers eligible, screened and referred (Table 19), but not on outcomes, with fewer than 20 respondents able to give numbers (Table 20). Many emphasised that although (some)

TABLE 20 Yield (numbers of children) from the SES for different definitions of hearing loss (question 16)

Children identified after failing the screen with ...	No. of service leads providing reliable data	No. of children: range
... sensorineural hearing loss	18	0–56
... permanent conductive hearing loss	11	0–23
... temporary conductive hearing loss	15	0–305
... other types of hearing loss	7	0–11

TABLE 21 Percentage of children confirmed to have one of three different definitions of hearing loss after referral from the SES as a percentage of those screened and as a percentage of those referred (question 16)

Yield	No. of service leads providing reliable data	Median	Range
(a) Sensorineural hearing loss			
Percentage of those referred	18	1.44	0–12.16
<i>(excluding 0%)</i>	<i>15</i>	<i>1.71</i>	<i>0.62–12.16</i>
Percentage of those screened	17	0.11	0–0.59
<i>(excluding 0%)</i>	<i>14</i>	<i>0.12</i>	<i>0.05–0.59</i>
(b) Permanent conductive hearing loss			
Percentage of those referred	11	0	0–17.56
<i>(excluding 0%)</i>	<i>5</i>	<i>3.42</i>	<i>1.24–17.56</i>
Percentage of those screened	10	0	0–0.44
<i>(excluding 0%)</i>	<i>4</i>	<i>0.09</i>	<i>0.07–0.44</i>
(c) Temporary conductive hearing loss			
Percentage of those referred	14	35.88	0–100.0
<i>(excluding 0%)</i>	<i>13</i>	<i>35.88</i>	<i>7.74–100.0</i>
Percentage of those screened	14	2.48	0–7.56
<i>(excluding 0%)</i>	<i>13</i>	<i>2.77</i>	<i>0.72–7.56</i>

data were collected it was not easy or possible to retrieve them in any meaningful form.

From the data in *Tables 19* and *20*, figures for the yield from the SES may be calculated. *Table 21* presents the yield as the percentage of children referred and as the percentage of children screened who were subsequently identified as having sensorineural hearing loss, permanent conductive hearing loss or temporary conductive hearing loss. For example, 18 services provided data on the number of children identified with sensorineural hearing loss and the number of children referred. A median yield of 1.44% of those referred can be calculated for the data from these 18 services, with a range of 0 to 12.16%. The rows in italics refer to the data excluding services where no children were identified.

Respondents' views on the value and continued need for the SES

Services were asked whether they had any plans for development or change of the SES. For those

services who responded, 28.6% (50/175) are planning to change their current practice in some way, several in response to the implementation of the NHSP (*Table 22*).

Services were also asked to rate the overall usefulness of the SES as it was currently operated. They were asked to indicate one of ten boxes on a range from 'not useful at all' to 'very useful' (question 20) (*Table 23*).

Thus, 69.3% rated it as 8 or higher, more than one-third (33.5%) rating it as 10. Only 12 services of the 176 responding (6.8%) rated it as 4 or less.

Positive comments about the service were made by 184 services. The most common was that the screen effectively identifies children with hearing impairment; in general (38), impairment of late onset or acquired since the newborn screen (30), OME (20), unilateral (15) or unsuspected losses (seven). Other suggested benefits of the screen were that it could exclude hearing loss as a cause

TABLE 22 Proposed developments in the SES (question 19)

Development plans	n
Changes to the staff running the screen	15
Generally reviewing the screen	13
Considering changes or discontinuation in the light of the introduction of NHSP	8
Considering discontinuation	4
Considering targeting the screen	5
Further development of IT systems	3
Changing the protocol with regard to screening levels	2
Provision of modern equipment (2) or training (2)	4
Improving relationship with second tier audiology services	2
Waiting for these results	2

TABLE 23 Rates of the overall usefulness of the school entry hearing screen as it is currently operated (question 20)

	1	2	3	4	5	6	7	8	9	10	
Not useful at all	←————— ————— ————— ————— ————— ————— ————— ————— ————— ————— —————→										Very useful
No. of responses	1	5	3	3	10	9	23	35	28	59	
%	0.6	2.8	1.7	1.7	5.7	5.1	13.1	19.9	15.9	33.5	
Total n = 176.											

of a child's difficulties (24) and that it raised awareness among teachers and parents and allowed surveillance (13).

In contrast, only 54 negative comments were made about the screen service. These were concerned with poor test conditions (12), high referral rates and/or low yield (ten), the provision of a less than adequate service owing to lack of resources (seven), poor information on outcomes (six), high rates of non-attendance at follow-up (five), and uncertainties following the introduction of newborn screening (two).

Respondents were asked specifically for their suggestions for the future of the SES, either locally or nationally (question 21). Twenty respondents said that the screen must continue and only two specifically said that it should stop. The future of the screen in relation to newborn screening was again highlighted (17), with suggestions for introduction of a targeted SES. Other suggestions were for better standards/guidelines (seven), increased coverage of private schools (six), improved test conditions, and better information technology (IT) support and data collection.

Finally, respondents were invited to add any further comments. Many of these reiterated, for emphasis, comments made earlier in the questionnaire concerning inadequate conditions, IT support, training, etc.

Summary

No national protocol exists for the SES programme and there are wide variations in its implementation throughout the UK in terms of the population covered, the physical location and conditions under which the screen is implemented, the test methodology, the criteria used to determine which children to refer, the personnel and the equipment involved, and the ability to collect and then retrieve data. Much of the variation appears to be due to limitations of resources to implement the screen in the light of competing activities for the range of staff employed to run it.

Data that could be used to assess outcome/performance and thereby to determine which methods, if any, were effective in achieving the aims of the screen are not routinely available. This lack of data, combined with the wide variation, meant that the authors were unable to investigate whether significant clusters of services had similar aims, tests and operational characteristics.

Despite the difficulties and the lack of robust evidence, most service leads think the screen is useful and do not want to stop using it, even though they recognise that its worth may become even less with the advent of universal newborn hearing screening. Guidelines concerning the value of a selective screen and the population for whom it would be appropriate to target it would be welcomed by many service leads.

Chapter 3

Possible effects of newborn hearing screening on the school entry hearing screen: evidence from a series of studies in Waltham Forest

Introduction

Primary preventive measures have affected the underlying epidemiology of permanent childhood hearing impairment (PCHI). There has been a reduction in the incidence of hearing impairment attributable to rhesus incompatibility, and in that remaining as a legacy of congenital rubella, but more recent immunisations for meningococcal and *Haemophilus influenzae* type b (Hib) disease have also had a demonstrable impact which should have consequences for the occurrence of hearing impairment (see www.hpa.org.uk: Vaccine Preventable Diseases; data generated by the Centre for Infections). Meningitis, measles and mumps have all been implicated as causes of unilateral acquired hearing impairment²⁵ for which in the past the SES has been an important route to identification. Although there are current issues concerning measles, mumps and rubella (MMR) immunisation uptake, the fall in both measles and mumps notifications that followed the introduction of vaccine in 1988 was dramatic, and predictably such preventive measures have consequences for the occurrence of hearing impairment.

Improvements in secondary prevention also have implications for the SES. Since the SES became a valued (although rarely evaluated) screen in the 1960s, audiology services and programmes of early detection have developed substantially. Potentially the most significant factor has been the implementation of neonatal screening. The NHSP has been rolled out across England, but the implications for the SES will not be fully realised until the first cohorts reach school age. However, the Whipps Cross University Hospital universal newborn hearing screen (UNHS) has been implemented as an audited service since 1992 pre-dating the country-wide implementation of newborn hearing screening by more than a decade. Although the Whipps Cross UNHS and the NHSP differ in terms of protocol details [transient evoked otoacoustic emission test (TEOAE) followed if failed by a second TEOAE

test, with screen referral if either test failed for the former; TEOAE followed if failed by AABR, with screen referral if AABR failed for the latter], longitudinal data from the Whipps Cross screen provide a valuable and quasi-controlled indication of the likely implications for later screens such as the SES.

The Whipps Cross UNHS was implemented for the newborn population in the East London district of Waltham Forest. A single audiology service with long-term stability has been responsible for the screen follow-up programme and for the audiological provision for a population of around half a million in Waltham Forest and in the neighbouring district of Redbridge. Whereas the universal screen aimed at screening all newborns in Waltham Forest, a more limited targeted neonatal hearing screen (TNHS), aimed at newborns at risk of hearing impairment, was undertaken in Redbridge. Prospective follow-up of the cohorts has been undertaken by the audiology service and evaluation of process, surrogate outcomes and actual outcome reviewed.²⁶⁻³⁰ The available information has also been used to evaluate the current worth of the local SES. The necessity for a local evidence base to be available to inform local policy has been emphasised,⁵ but there are concerns about the applicability of studies with a small population base to inform wider policy. However, the Waltham Forest cohorts will be unique within the UK until the much larger long-term follow-up evaluations from the NHSP are available, and the use of the local cohorts to inform wider policy has therefore been maximised by using longitudinal population studies and by comparing the results with national studies or with data drawn from larger populations.

Changes in the programmes of early screening and identification

Approaches to screening for hearing impairment in preschool and primary age children in Waltham Forest are detailed in *Figure 1*. Changes were based

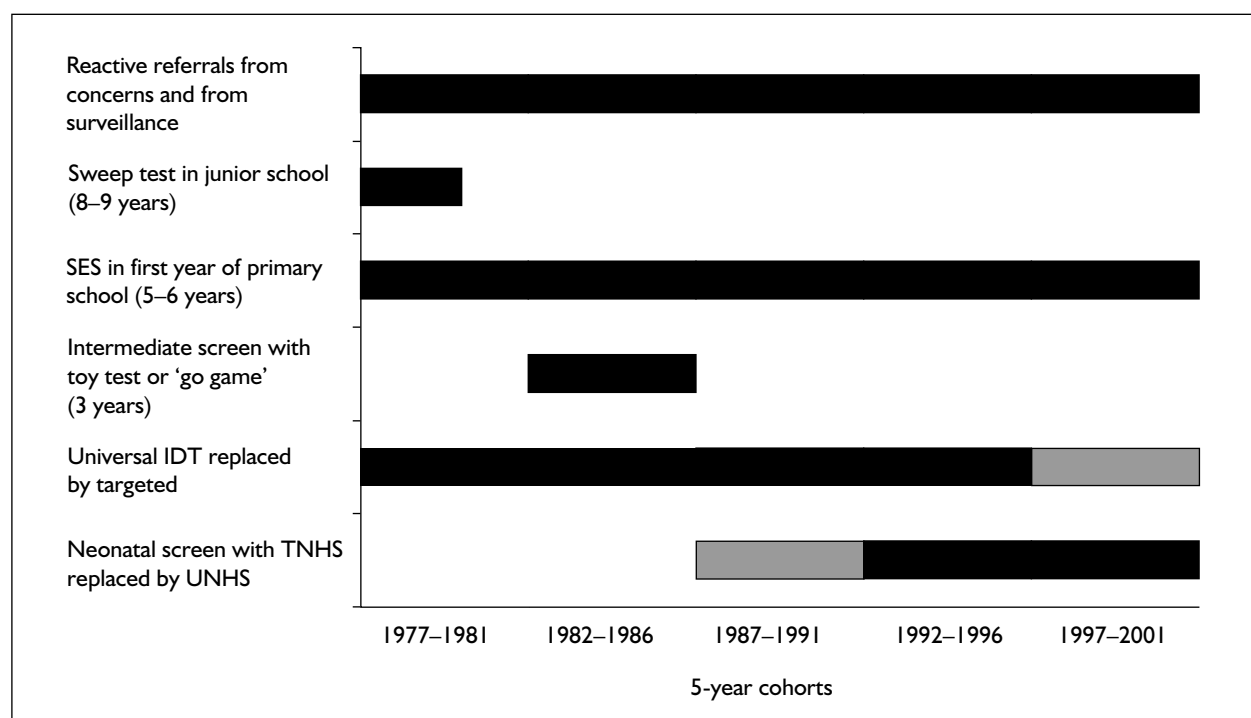


FIGURE 1 Components in the Waltham Forest programmes of secondary prevention for 5-year cohorts born over the 25 years from 1977 to 2002

on epidemiological data and screening results from the 1970s to 2002, when local implementations became centrally directed.^{26-28,31-35} Throughout the 25 years reactive referrals were received from primary health care, from paediatric and ENT services, and also directly from parents. The IDT was undertaken as a universal infant hearing screen at 8 months of age, until 1996. UNHS was introduced after a short period of targeted neonatal screening and the universal IDT was replaced by a targeted screen in 1996. Changes in Redbridge mirrored those in Waltham Forest, but the TNHS was retained from 1990 and the IDT remained a universal screen throughout the period.

The SES has always been considered an important backstop universal screen. It was typical of that used in many other districts and was a six-frequency sweep from 250 to 8000 Hz at 25 dB HL undertaken in school by school nurses. Screen positives were retested whenever possible in school, and failure at the second test prompted referral to the local clinic where threshold audiometry was undertaken. Children with OME were referred onwards to either their GP or ENT services after a period of conservative management in second tier or school nurse hearing clinics. Children with PCHI of any degree,

or any diagnostic uncertainties, were referred to the audiology services, and children with PCHI reactively referred to the ENT or paediatric service were redirected to audiology.

Screen results were initially reported to the health authority Directorate of Information, and from the 1990s were computer recorded on a Regional Interactive Child Health System (RICHES). Data retrieval was routinely undertaken through these sources and through notifications to the educational service for hearing impairment.

Evaluating the SES

The investigation consisted of two complementary evaluations.

Evaluation A

The changing worth of the SES in terms of the yield of children with a significant PCHI picked up by the screen was measured longitudinally. PCHIs are notified to educational and audiology services and their aggregated details periodically analysed. The number of PCHIs ascertained from cohorts that have received the SES, the identification methods and audiological data were available for comparison of three cohorts with different detection programmes:

- an historical 10-year Waltham Forest cohort born from January 1977 to 1987 when no neonatal hearing screen (NNHS) was in place; the size of this NNHS cohort was 31,538 (cohort 1, NNHS)
- a 10-year Redbridge cohort born from January 1990 to 2000 when a TNHS was in place; the size of this TNHS cohort was 32,890 (cohort 2, TNHS)
- an 8-year Waltham Forest cohort born from January 1992 to 2000 when a UNHS was in place; the size of this UNHS cohort was 29,132 (cohort 3, UNHS).

The three cohorts thus gave a total cohort of 93,560. Although the primary analyses are concerned with comparisons between cohorts, useful information (e.g. overall prevalence rates) can be derived from some combined analyses (combined cohort).

Evaluation B

The SES has also been argued to have an additional 'useful' yield of children with a minimal PCHI or with temporary fluctuating OME. Because these children are not routinely and invariably notified to the educational or audiology services an additional population-based cross-sectional survey was undertaken by examining the school health records of a 6-year Waltham Forest cohort (January 1993 to 1999) of 19,296 children (cohort 4) who were eligible to have received the SES in the local education authority schools, and who had previously been enrolled into the UNHS. The RICHS was interrogated for the study. It enabled local comparative evaluation of process and results with historical returns from a 3-year SES undertaken on 9301 children from 1986 to 1989 (cohort 5) and with a 5-year Waltham Forest cohort of 15,536 children who were born up to December 1982 (cohort 6) and reported after they had all received the SES;³² neither of these cohorts had had a newborn hearing screen.

Generalising the study

Generalisation requires contextualisation of the results within the local demographic. The level of deprivation³⁶ and Asian ethnic background¹³ are both factors that have been found to increase the odds ratio for PCHI within a community, and inward flow of children into the district from abroad after the newborn or infant screens, and before school entry, are characteristics that are not uniform nationwide. Nevertheless, they are pertinent to the wider interpretation of the

current data. The combined population of Redbridge and Waltham Forest in the mid-1990s was 449,500. The live birth rate in Waltham Forest was 3500 per annum, with that in Redbridge being slightly less. Although the districts are immediate neighbours, the populations have some differences. The Department of the Environment Index of Local Conditions using data from the 1991 census ranked Waltham Forest as 20th on the index of the most deprived boroughs, while Redbridge was ranked 120th out of 366 English boroughs. In both there were ethnic minority communities (31% in Waltham Forest and 28% in Redbridge). In Redbridge the largest ethnic community was Indian in origin, and Waltham Forest had the largest Pakistani community, the eighth largest black Caribbean community and the ninth largest Bangladeshi community in London. During the 1990s there was also an increasing number of refugees to both boroughs, principally Turkish, Kurdish and Somalian. The majority grouping in both was Somali. The longitudinal cohort evaluation required assessment of changes in the Waltham Forest population. Data from the 1981 census confirmed that the population in the 1980s was slightly lower than it was a decade later (214,500 in the mid-1980s), with a slightly lower birth rate of around 3000 per year. However, the overall level of deprivation measured by the 1981 census was largely unchanged when compared with later measures from the Index of Local Conditions.

Local data on the epidemiology of hearing impairment were compared with those derived from three studies with a national or wider population base:

- the prevalence of PCHI in the UK¹²
- the MRC Institute of Hearing Research epidemiology of PCHI in a cohort of 366,480 from the Trent region¹³
- evaluation of the NHSP in England.⁹

The local population-based cross-sectional survey of the SES process and result was compared with the national BACDA study of 109,505 school hearing screening tests undertaken by 43 services for the school year 2000/01 (the 1995/96 birth cohort).⁶

Changes in the SES

Although SES protocols and procedures remained unchanged throughout the longitudinal cohort comparison, the school nursing services had been

increasingly used to provide other preventive services and since 2000 this has affected the performance of the SES in Waltham Forest. During the 1980s and up until the end of the 1990s, 90% or over of schoolchildren in the first year of primary school received a SES. Enrolment to the SES for the majority of the cohorts used in the longitudinal comparisons therefore remained high. However, in the cohorts screened since 2000 the proportion receiving the SES has gradually reduced and in 2005 was below 50%. The failure rate was 7.4%, with a mean age of referral for assessment in the school clinic of 5.5 years, and this had remained stable since the 1980s. The non-attendance (DNA) rate for follow-up had also remained stable at 20%. Comparison with the BACDA survey⁶ confirmed that these results were typical of those achieved elsewhere in the UK.

Did the gradually falling SES enrolment after 2000 influence the yield? The non-screening referrals to the audiology service were examined and the age distribution and referral rate from cohort 1, NNHS, were compared with the latest year. In fact, despite all the changes in earlier screens and surveillance, the age distribution and referral rate of reactive vigilant referrals remained remarkably stable. The modal age of referral remained at between 3 and 4 years, with a mean age of referral of 4.81 years in 1986 and 4.65 years in 2005. The reduced coverage of the SES appears therefore not to have resulted in increased numbers identified through reactive referral, and there has been no influx of schoolchildren with PCHI who have been reactively referred. Longitudinal comparison of the epidemiology of hearing impairment was also made to ensure that there was no fall in the number of identified cases in recent years. Comparison was also made with the epidemiological study of PCHI in Trent.¹³ The prevalence of PCHI at school age also remained longitudinally stable.

Results

Evaluation A

Prevalence

From the combined cohort of 93,560 in their first year of primary school, 349 children with a unilateral or bilateral PCHI of mild degree or greater (>20 dB HL averaged over 0.5, 1, 2 and 4 kHz) were ascertained. Severity was classified as mild (hearing level <40 dB HL), moderate (hearing level 40–69 dB HL), severe (hearing level 70–94 dB HL) and profound (hearing level ≥95 dB HL). Bilateral PCHIs were categorised by

severity in the better hearing ear (BHE) and unilateral cases in the worse hearing ear (WHE). In those where pure tone audiometric thresholds were unavailable, sound field or electrophysiological test results were used with categorisation by degree after appropriate conversion of the decibel scale. The prevalence of moderate or worse bilateral PCHI (*Figure 2*) was 1.49 in 1000 [95% confidence interval (CI) 1.23 to 1.73], with 1.27 in 1000 (95% CI 1.03 to 1.49) appearing to be congenital. There was clear audiometric evidence of progressive hearing impairment in 18% of those with a congenital PCHI. The prevalence of hearing impairment that was acquired or late onset was 0.22 in 1000 (95% CI 0.13 to 0.32). There was no significant difference when the prevalence rates in the three cohorts in the present study were compared with each other [χ^2 0.060, degrees of freedom (df) 2, $p = 0.967$] or when they were compared with the Trent cohort (χ^2 2.532, df 1, $p = 0.112$) (see Appendix 3). Similarly, the prevalence of 1.44 in 1000 (95% CI 1.41 to 1.48) with a moderate or worse bilateral hearing impairment in the UK ascertainment study¹² was very similar to the prevalence of 1.49 (95% CI 1.24 to 1.73) measured in the current study.

The aetiologies of the hearing impairment were categorised and fell equally into one-third who had an inherited familial hearing impairment, one-third where no cause was known and one-third who had a perinatal illness, or a craniofacial abnormality or dysmorphology. Once again, there were no significant differences between the three cohorts (χ^2 0.215, df 2, $p = 0.898$; χ^2 1.469, df 2, $p = 0.480$; and χ^2 2.264, df 2, $p = 0.322$, respectively). There were also no significant differences between the Waltham Forest cohorts and the Trent cohorts in the proportions with an inherited deafness; a perinatal illness, congenital infection or craniofacial abnormality/syndrome; or an unknown or missing aetiology (χ^2 0.412, df 1, $p = 0.521$; χ^2 1.062, df 1, $p = 0.303$; and χ^2 2.224, df 1, $p = 0.136$, respectively) These epidemiological characteristics of bilateral moderate or worse PCHI were therefore longitudinally stable and they were also representative of other cohorts reported in the UK.

The prevalence of mild bilateral PCHI was 1.21 in 1000 (95% CI 0.99 to 1.43), and once again this was longitudinally stable in Waltham Forest (χ^2 0.052, df 2, $p = 0.974$), but there are no national studies for comparison.

By contrast, there were highly significant differences (χ^2 8.229, df 2, $p = 0.016$) between the

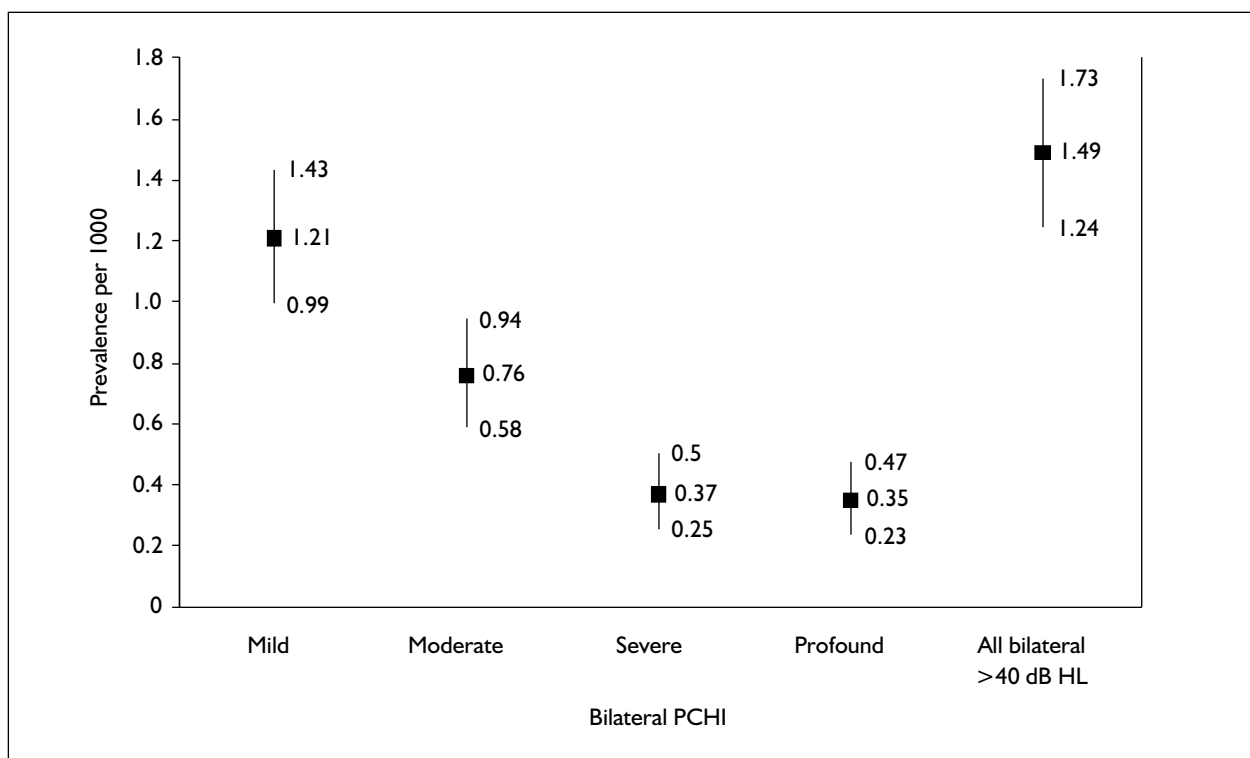


FIGURE 2 The prevalence of bilateral PCHI and 95% CIs for the combined cohorts (n = 93,560)

cohorts in the prevalence of cases (congenital and acquired) of unilateral hearing impairment (Figure 3). Note that for unilateral hearing impairment it is more reasonable, in terms of the extent of disability, to group moderate impairment with mild rather than severe. Cases of severe or profound unilateral hearing impairment (children colloquially considered to have a 'dead ear') fell from 0.95 in 1000 in cohort 1, NNHS, to 0.27 in 1000 in cohort 3, UNHS (χ^2 13.338, df 2, $p = 0.001$), with those with a milder unilateral PCHI remaining stable across the longitudinal cohorts (χ^2 0.609, df 2, $p = 0.737$). The aetiologies were investigated, but causation was usually elusive. Only 18.5% had onset definitely temporally related to illness and the remainder were considered to be congenital, although usually this was because of absence of firm evidence that the unilateral impairment was related to an illness, rather than clear evidence that it was present at birth. In the majority (65%) there was no known cause and it was in this category that there was a highly significant fall in prevalence. The prevalence decreased by a half from 0.86 in 1000 (95% CI 0.53 to 1.18) in cohort 1, NNHS, to 0.41 in 1000 (95% CI 0.18 to 0.64) in cohort 3, UNHS. Once again, comparative prevalence rates were not available from the Trent or national prevalence studies and therefore generalisation is less robust. However, the UK Child Development

Studies reported 0.8 in 1000 children aged 7 years to have such an impairment,³⁷ and other historical surveys have reported profound unilateral hearing impairment in 1 in 1000 schoolchildren (many studies cited by Bess and colleagues 1986²⁵). These were of the same order as the prevalence rate reported in the cohort 3, NNHS. The prevalence rate of all degrees of unilateral congenital hearing impairment reported from the recent NHSP evaluation was 0.64 in 1000 (95% CI 0.37 to 0.91), of whom less than half had a severe or profound unilateral PCHI.⁹ Cohort 3, UNHS, used in the current study reported a unilateral congenital prevalence of 0.65 in 1000 (95% CI 0.36 to 0.95), with 0.24 in 1000 (95% CI 0.06 to 0.42) having a severe or profound unilateral PCHI. It is therefore probable that the significant fall in prevalence of profound unilateral hearing impairment can be generalised and it is likely that this is due to measures of primary prevention.

Age of confirmation of hearing impairment

Cohort comparisons of the median and interquartile ages when the children were confirmed with a congenital PCHI are detailed in Table 24. The comparison confirmed that there had been a considerable secular reduction in the age when hearing impairment was confirmed when cohort 2, TNHS, and cohort 3, UHNS, that

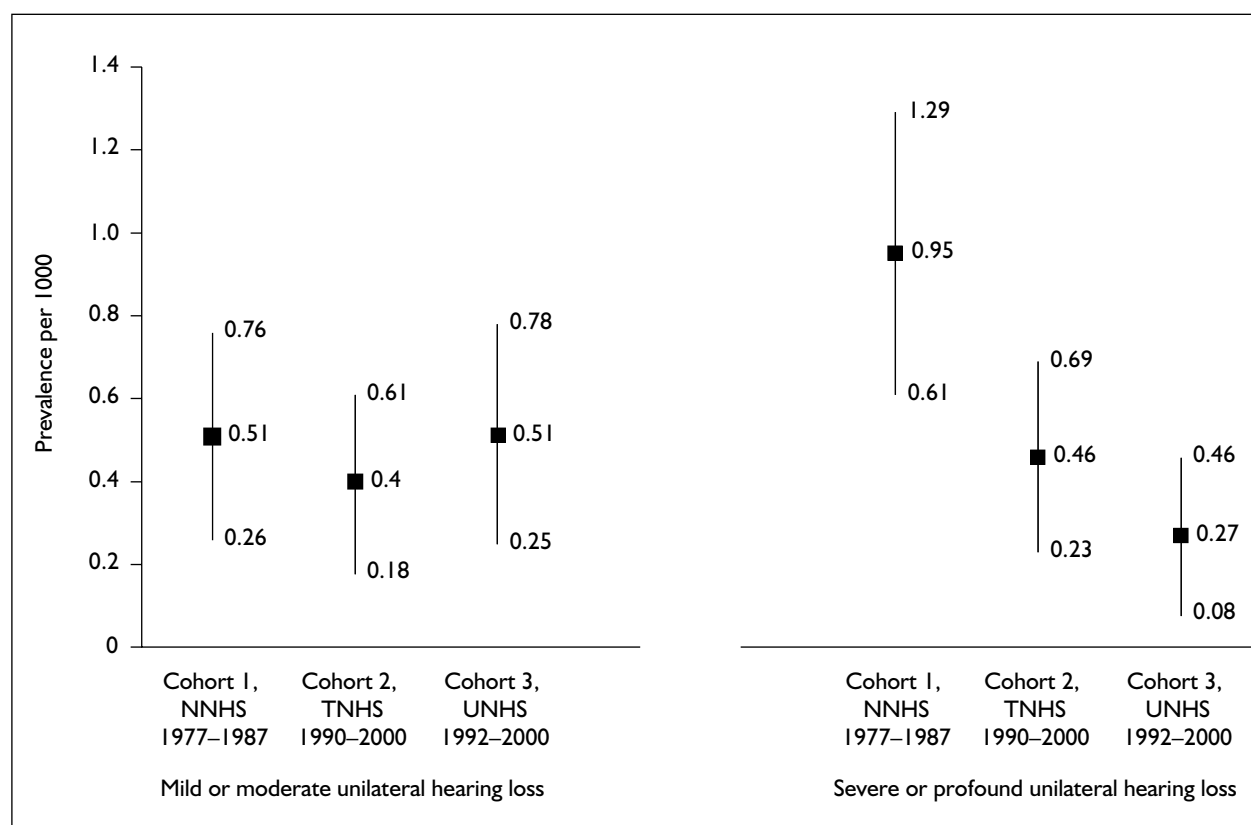


FIGURE 3 The prevalence and 95% CIs for unilateral PCHI in the three cohorts

received either a targeted or universal newborn screen were compared with cohort 1, NNHS, when newborn screening was unavailable. In cohort 1, NNHS, the median age when hearing impairment was confirmed in those with severe or profound hearing impairment was at just turned 1 year of age. However, there was a much greater delay in the confirmation of those with a mild or moderate PCHI. This occurred on average after their fourth birthday and in the months running up to primary school entry. Once universal neonatal screening had been established there was a highly significant reduction to 10 weeks of age in confirming the presence of a severe or profound hearing impairment, with those with a moderate PCHI also now being confirmed on average well within the first half of infancy. Those with a mild bilateral PCHI were now being confirmed at around 3 years of age. The same pattern of improvements was also made in the age of confirmation of those with unilateral PCHI, with a highly significant overall reduction when cohort 1, NNHS, was compared with cohort 2, UNHS. Historically, unilateral PCHI had been confirmed when the children had reached their fifth birthday, but once again this was now reduced to the first half of infancy. These were highly significant

improvements in the average age at which all degrees of hearing impairment were identified in cohort 3, UNHS.

Such improvements in the median age would be expected following the implementation of effective neonatal screens, but a statistic giving a better indication of late identifications is required to reflect the effectiveness of the system as a whole and the contribution from later screens such as the SES. The 75th centile is thus a more appropriate index for measuring improvements. The interquartile range (IQR) is included in *Table 24*. Historically, the upper quartile for the age of confirmation was within the first year of primary school for all degrees of PCHI, other than for severe and profound hearing impairment, where it was at 2.5 years of age. Had the remarkable improvements in the median age of confirmation following the implementation of the UNHS also been reflected in a lowering of the 75th centile? No child with a severe or profound bilateral PCHI remained with an unconfirmed hearing impairment after 3 years of age and the 75th centile had been reduced by over 1 year to just under 18 months. However, lesser improvements were seen in the other degrees of PCHI and the

TABLE 24 Median ages of confirmation of hearing impairment and IQR for those who had remained resident or moved in with a previously unidentified PCHI in the three cohorts (n = 266)

Degree of hearing impairment		Median age (IQR) (weeks)		
		1977–1987 Cohort 1 NNHS (n = 101)	1990–2000 Cohort 2 TNHS (n = 84)	1992–2000 Cohort 3 UNHS (n = 81)
Bilateral (PTA average BHE 500–4000 Hz)	Mild (20–39 dB HL)	218 (166–307)	187 (49–267)	150 (13–257)
	Moderate (40–69 dB HL)	218 (130–252)	119 (23–198)	18 (8–247)
	Severe/profound (≥70 dB HL)	53 (47–131)	44 (20–157)	10 (8–76)
Unilateral (PTA average WHE 500–4000 Hz)	All unilateral (>20 dB HL)	265 (194–286)	56 (15–298)	18 (11–243)

TABLE 25 Median and IQR ages of referral and delays to confirmation for those with a congenital PCHI who had remained resident or moved in with a previously unidentified PCHI in cohort 3, UNHS

Degree of hearing impairment	Median (IQR) (weeks)		
	Age of referral	Age of confirmation	Delay from referral to confirmation
Mild bilateral (n = 28)	115 (4–236)	150 (13–257)	13 (5–24)
Moderate bilateral (n = 19)	11 (5–233)	18 (8–247)	6 (2–11)
Severe/profound bilateral (n = 16)	6 (5–64)	10 (8–76)	3 (2–5)
All unilateral (n = 18)	9 (1–237)	18 (11–243)	6 (4–11)

75th centile remained within the weeks leading up to the fifth birthday. Around one-quarter of those children with any degree of PCHI other than a severe or profound hearing impairment therefore still remained to have that hearing impairment confirmed at around school entry, even though the average age of confirmation had been so drastically cut by the introduction of the neonatal screen. When the cumulative distributions of the age of confirmation were examined by severity, 22% of those with a moderate bilateral PCHI, 26% of those with a mild bilateral PCHI and 18% of those with a unilateral hearing impairment in the first year of primary school still remained to have their hearing impairment confirmed after 5 years of age. Although the introduction of neonatal screening had reduced the average age of confirmation, it appears to have done little to reduce the number of ‘stragglers’ in the overall system of detection.

Ages of referral

The cohort evaluations used the age when the congenital hearing impairments were confirmed as the most robust and stable indicator available

for the longitudinal comparison. However, confirmation delays may have reflected problems in the assessment of children who had actually received a timely referral. The age at referral was therefore examined separately for the cohort that had received the UNHS (cohort 3, UNHS, born 1992–2000) (Table 25). The delay from referral to confirmation was also measured. Median delays from referral to confirmation were less than 1 month for those with a severe or profound hearing impairment. They were slightly longer for those with a lesser degree or a unilateral hearing impairment, but even in those with a mild bilateral impairment the median delay between referral and confirmation was 3 months. The delays in confirmation therefore reflected delays in reactive referral or in referral from the screening programmes. When the cumulative distributions of the age of referral were examined by severity, 16% of those with a moderate bilateral PCHI, 18% of those with a mild bilateral PCHI and 17% of those with a unilateral hearing impairment who were in their first year of primary school still required identification and referral for hearing assessment.

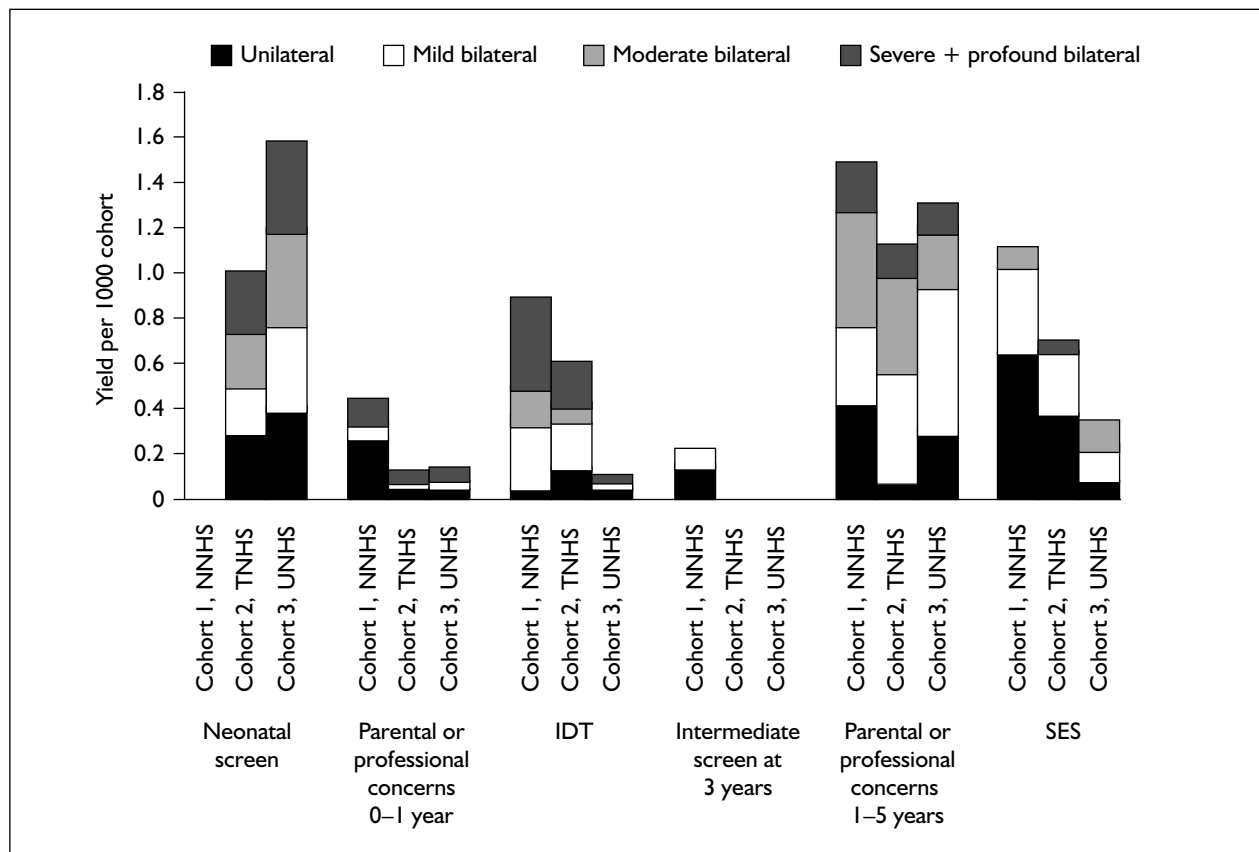


FIGURE 4 The yield per 1000 from the three cohorts by the method of identification

Routes to identification

The comparative yields per 1000 cohort for the different identification methods in the three cohorts are detailed in *Figure 4* and *Table 26*. There were significant longitudinal changes in the yields from all the identification methods, other than in the yield from parental or professional concerns from 1 to 5 years of age. (The slightly higher overall yield from cohort 1, NNHS, is accounted for by more unilateral cases, probably due to the incidence of mumps at this time.)

Electrophysiological testing was only introduced for reactive neonatal referrals towards the end of the 10-year period of cohort 1 and early identification was based around the IDT screen at 8 months. The sensitivity of the IDT screen for those with a severe or profound congenital hearing impairment who remained in the district was 77%, but the sensitivity for lesser degrees of bilateral hearing impairment was only 25%. Because this low sensitivity of the IDT screen was widely experienced, the Advisory Committee on Services to Hearing Impaired People (ACSHIP) report (1981) recommended the implementation of an intermediate universal screen around 3 years of age, and this was undertaken in the district for

a period of 5 years. However, it gave a low yield that consisted entirely of those with a mild or unilateral hearing impairment. The largest yield from cohort 1, NNHS, was from assessments undertaken because of parental or professional concern from 1 to 5 years, and this included 45% of those with a congenital bilateral moderate or worse hearing impairment and 62.5% of those with an acquired or late-onset hearing impairment of this degree. The second largest yield for these cohorts came from the SES (*Table 26*).

The implementation of a selective neonatal screen using the auditory brainstem response (ABR) testing of neonates with risk factors based on the American Joint Committee register (1982) gave an overall yield of 1.00 in 1000 neonates with PCHI, with 0.51 in 1000 having a moderate or worse bilateral PCHI. The sensitivity of the district TNHS programme for identifying moderate or worse bilateral congenital PCHI at 46.9% was typical of many other such programmes.⁵ However, the earlier identification offered by the TNHS was partly offset by a reduced yield from both parental and professional vigilance and from the IDT screen. By combining all three infant detection methods, a yield of 0.76 in 1000 with a

TABLE 26 Comparison of the yields from the identification methods in the three cohorts

Identification method	Yield of all PCHIs per 1000 in cohort (95% CI)			Pearson χ^2 (p)
	Cohort 1 No NHS	Cohort 2 Targeted NHS	Cohort 3 Universal NHS	
Neonatal screen	NA	1.003 (0.71–1.41)	1.58 (1.18–2.11)	4.03 (0.045)
Concerns 0–1 year	0.44 (0.26–0.75)	0.12 (0.05–0.31)	0.14 (0.05–0.35)	8.84 (0.01)
IDT screen	0.89 (0.61–1.28)	0.61 (0.39–0.94)	0.10 (0.04–0.30)	17.49 (<0.001)
Intermediate 3-year screen	0.22 (0.11–0.46)	NA	NA	NA
Concerns 1–5 years	1.49 (1.12–1.98)	1.12 (0.82–1.55)	1.30 (0.95–1.79)	1.65 (0.44)
SES	1.11 (0.80–1.54)	0.70 (0.41–0.99)	0.34 (0.19–0.63)	12.30 (0.002)

NA, not applicable.

moderate or worse bilateral congenital PCHI was achieved by the end of infancy. This contrasted with a yield of 0.63 in 1000 for cohort 1, NNHS, achieved by this age. The targeted neonatal programme had therefore conferred some benefit, but the largest individual yield in cohort 2, TNHS, once again was from assessments undertaken because of parental or professional concern from 1 to 5 years, with the yield from the SES being reduced by over one-third from 1.11 to 0.70 in 1000.

The implementation of universal newborn screening, as evidenced in the data from cohort 3, UNHS, resulted in a high rate of detection through this route (1.58 in 1000), with other routes predictably reducing. However, parental and professional concerns continued to deliver cases of PCHI at significant rates, especially mild and, to some extent, unilateral losses. The yield of the SES reduced to 0.34 in 1000 for all PCHI, and to less than 0.1 in 1000 for unilateral PCHI.

Effect on the SES of introducing a universal neonatal screen

It had been widely anticipated that universal neonatal hearing screening would diminish the need to retain a further universal screen at school age. It was inevitable that the yield of early identified PCHI would be increased by introducing a sensitive UNHS. Indeed, in cohort 3, UNHS, the neonatal screen contributed the largest individual yield within the overall programme, with the obvious inference being that

numbers requiring later case finding would be low. However, following up the neonatally screened cohort into primary school demonstrated that this was not the case. By primary school age the combined total prevalence of PCHI was 3.47 in 1000, with only 1.58 in 1000 (46%) being identified by the district's UNHS. This seeming contradiction was explained by the downward cascade of effectiveness of a screen undertaken at birth for identifying a condition present in the cohort several years later. *Figure 5* illustrates the incremental steps that eroded UNHS effectiveness for identifying the PCHIs present in primary school. The following factors cumulatively contributed to the need for late case finding.

- Children identified by UNHS moved away, with other preschool hearing impaired children moving in. The yield of moderate or worse PCHI confirmed by the UNHS programme before any had moved out was 1.03 in 1000. However, by school age 0.21 in 1000 of these had moved out to be replaced by 0.24 in 1000 moving in, without neonatal confirmation (note that this is not a situation that would apply once the NHSP is fully implemented nationwide). Similarly, the yield of unilateral hearing impairment confirmed by the neonatal programme was 0.55 in 1000, but of these 0.17 in 1000 moved out to be replaced by 0.10 in 1000 moving in. If those with a mild PCHI are included, then of the 3.47 in 1000 with a PCHI in primary school 0.48 in 1000 had moved in, with 86% having a previously

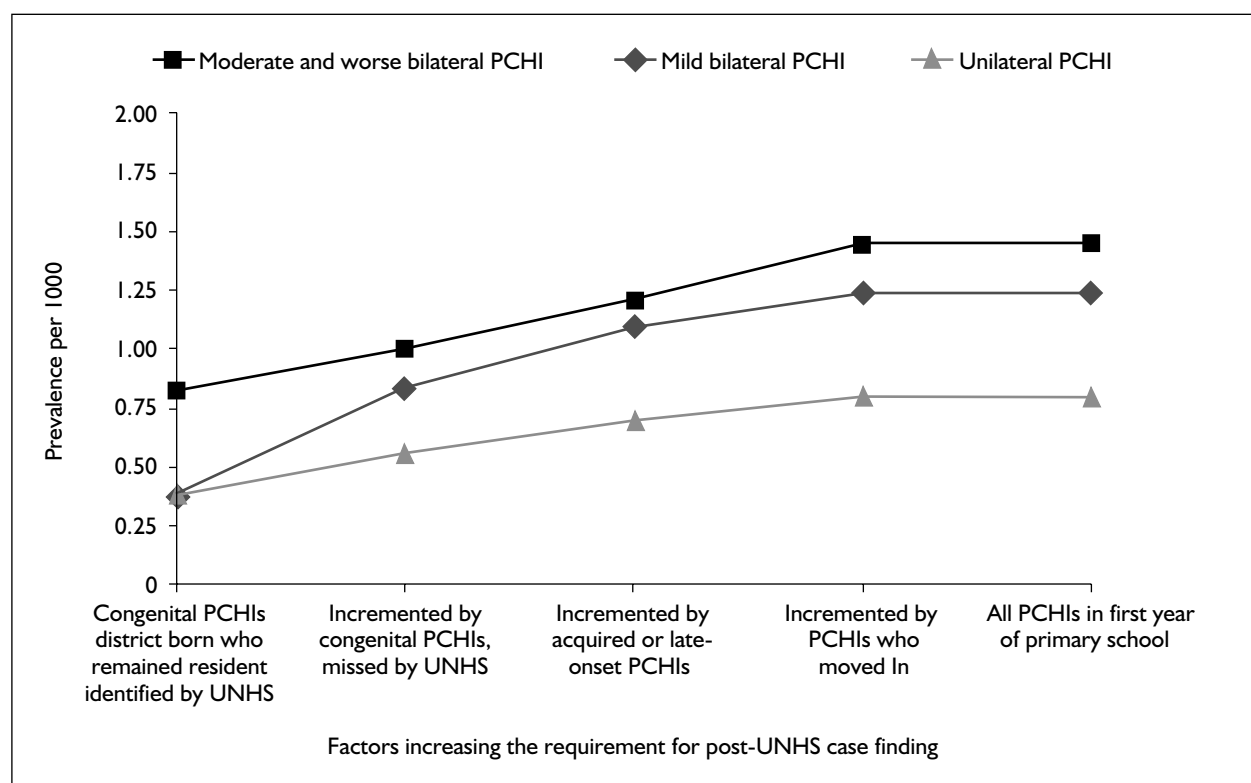


FIGURE 5 Incremental factors prompting the need for case identification after the neonatal screen and by the first year of primary school

unidentified hearing impairment. In this East London district two-thirds had moved in from abroad.

- PCHI in the school-aged cohort was acquired postnatally or of late onset in 0.20 in 1000, 0.27 in 1000 and 0.14 in 1000 for those with a moderate or mild bilateral or unilateral PCHI, respectively.
- Not all cases of congenital PCHI born and continually resident in the district had been picked up by the neonatal screen. The cases missed had implications for characterising the PCHIs that required later case finding. Of the children with a congenital PCHI, 94% had been enrolled for the neonatal screen and the test was 96% sensitive for picking up moderate or worse bilateral hearing impairment. A single child with this degree of PCHI and a reverse slope audiometric configuration was the only false negative identified over the period of the cohort. Screen sensitivity was 87% for congenital unilateral hearing impairment. Two children were false negatives – one had a similar reverse slope configuration and the other a profound unilateral impairment restricted to the high frequencies. TEOAE test sensitivity was 91% for those with a mild PCHI. Of all who had failed the cochlear emission

screen only 90% received the ABR assessment, but when undertaken, this confirmed the presence of PCHI in all apart from the mild PCHIs. In those with a mild congenital hearing impairment (they had failed the neonatal TEOAE test and/or there were no audiological and medical findings consistent with the impairment having been acquired), 42% were incorrectly assessed by the ABR to have hearing levels within the normal range. These factors combined to reduce the effectiveness of the overall programme, the sensitivity of which was 83% for moderate or worse bilateral hearing impairment, 69% for unilateral hearing impairment and 46% for mild PCHI. Therefore, despite a high screen sensitivity and a total UNHS yield of 1.58 in 1000, of the congenital PCHIs born and remaining resident, half as many again as had been picked up by the UNHS were subsequently identified after the neonatal period.

There was therefore a need for postneonatal case finding for 1.89 in 1000 of the children with PCHI in primary school. With a low yield from concerns in the first year of life and from the IDT screen, an incremental yield of 1.30 in 1000 from parental and professional concern from 1 to 5 years

remained the most productive means of case finding after the UNHS. This yield had not changed significantly compared with the earlier programmes (χ^2 1.650, df 2, $p = 0.438$) (Table 26), and consequently the place of the SES had been further eroded. There was a highly significant fall in yield to 0.34 in 1000 (χ^2 12.304, df 2, $p = 0.002$).

Identification through preschool hearing surveillance

The postneonatal yield of 1.89 in 1000 had mostly been identified because of parental or professional concerns, and the possibility of identifying a part of this yield by targeting a group for hearing assessment after the neonatal screen but before school entry was further evaluated from cohort 3, UNHS. The majority of the 0.48 in 1000 hearing-impaired children who had moved in had an unrecognised hearing impairment, and they could have been identified by referring for audiological assessment all those moving in who had not benefited from a previous hearing screen. Of the 0.62 in 1000 with an acquired or late-onset PCHI, a small number (0.1 in 1000) had no identifiable cause, although there had been strong audiometric evidence that their hearing had previously been normal. In those with an identified cause, the impairment was temporally related to a medical event (e.g. bacterial meningitis or measles) in 0.21 in 1000. A small number had a known family history of late-onset PCHI (0.06 in 1000), but the largest individual yield of 0.24 in 1000 was in those who had a late-onset hearing impairment associated with a craniofacial dysmorphism or a syndrome (e.g. Turner's, Down's, Rubinstein-Taybi). Their identification was possible through routine hearing surveillance undertaken in the child development centre. A similar targeting strategy was possible in the 0.79 in 1000 with a congenital PCHI that had not been picked up by the neonatal programme. As in the overall cohort, around two-thirds had risk factors for hearing impairment, with risk factors being absent in 0.27 in 1000. A positive family history was present in 0.21 in 1000, and 0.14 in 1000 had required admission to the special care baby unit (SCBU). However, once again the largest yield of 0.31 in 1000 consisted of children who had a dysmorphism or a neurodevelopmental condition that required the multidisciplinary care provided by the child development centre.

Of the postneonatal yield of 1.89 in 1000, identification would have been achieved in:

- 25% by referring for hearing assessment all those moving into the district

- around 10% by referral of all those with bacterial meningitis or where hearing impairment had been suspected following childhood viral illness
- around 15% by keeping a check on those with a family history of hearing impairment
- around 30% by routine hearing surveillance of all those attending the child development centre. Assessing this cohort would have been almost four times as productive as reviewing all graduates of the SCBU.

However, in 0.37 in 1000 (20%) there were no identifiable risk factors and pre-SES identification required reactive referral because of parental or professional concerns about hearing acuity or because of speech and language delay.

Evaluation B

Minimal permanent hearing impairment, OME and the SES

If the yield of children with a mild or worse PCHI is used to benchmark the worth of the SES, then this was significantly reduced (to 0.34 in 1000) in cohort 3, UNHS, that had already received a universal neonatal screen. However, irrespective of this, there is an argument that the SES might identify significant numbers of children with persistent middle ear disorders and 'minimal' PCHI that would not be identified neonatally.

There has been a recent increasing interest in the problems faced by schoolchildren with a minimal PCHI. These children have been defined in various ways, but in the severity classification used in the current study, audiometric inclusion criteria were a hearing threshold over 20 dB HL at any frequency, but with an average level of 20 dB HL or below. Such children usually present audiometrically with a hearing impairment at the extreme high frequencies or with a sensorineural dip in the middle frequencies. They have not been subjected to rigorous study in the UK, and studies reported from elsewhere have used a wide variety of inclusion criteria. Bess and colleagues¹⁴ recorded a high prevalence of 1.0% with a bilateral minimal PCHI and 1.4% with a high-frequency sensorineural hearing impairment in a sample aged 8.6–14.7 years, but in the severity classification used here many of these children would have been included in the mild category, whose results have already been discussed. However, interest in the performance and problems encountered by children with a minimal PCHI has prompted a more robust epidemiological study by the Centers for Disease Control and Prevention (Ross *et al.* Atlanta, GA,

USA: personal communication; 2005). Preliminary results have projected the prevalence in the general US population aged 6–16 years of ‘slight–mild’ bilateral hearing impairment and unilateral hearing impairment to be as high as 7.7%.

In the studies reported here, the longitudinal cohorts (cohorts 1–3) were not an appropriate tool to investigate minimal impairments because such cases would not necessarily be known to the educational or audiology services. The 1993–1999 population-based cross-sectional survey (cohort 4) described here was therefore used to investigate the role of the SES in identifying these children. Comparison was with data from cohort 1, NNHS, and with the results of the BACDA national survey.⁶ In cohort 4, of those children receiving the SES, 3.5 in 1000 had a newly identified minimal PCHI. The majority of these cases were not referred for further diagnosis or management by the secondary level services, but 70% were repeatedly followed up, 18% were referred to the third tier audiology unit and 12% were referred to their GP. The records were further scrutinised, and despite the repeated follow-up and onward referrals, habilitative or diagnostic audiological interventions were not undertaken on any of the children. The UNHS results of those who had been born in the district and remained resident were scrutinised and a small number had failed the TEOAE newborn screen. The sensitivity of the cochlear emission test for these children had been 14%, but the diagnostic ABR assessment following cochlear emission failure had been entirely unsuccessful in confirming this condition and the neonatal programme sensitivity for identifying minimal PCHI within the school cohort had been reduced to zero. It was not possible to assess retrospectively with any accuracy whether the minimal impairments had been present congenitally, but it can be assumed that some were acquired, and Bess and colleagues¹⁴ have argued that many such hearing impairments may be left as a legacy of otitis media. However, it is clear from the present study that none had emerged with a diagnosis of PCHI following the neonatal diagnostic assessment. A single child with a minimal PCHI had been referred reactively before school entry, but otherwise this condition was invariably picked up by the SES. The yield had been very much lower than that predicted from US studies, but had remained stable over time using the same screening method in Waltham Forest. The returns of minimal PCHI from 1986 to 1989 (cohort 5) had been 2.4 in 1000 screened. Although the current yield was slightly higher, this difference was not significant (χ^2 0.061, df 1,

$p = 0.805$). Clearly, the SES in its present form can give a low but relatively stable yield of minimal PCHI. Perhaps the most noteworthy outcome of this limited cross-sectional survey was that it has not been judged to be useful to provide any further intervention for such cases.

The identification of OME cases via the SES was also examined for cohort 4. The yield was 20 in 1000 of those screened. However, many were already under treatment and only 14 in 1000 were newly identified and considered true positives of the screen. Even then, when these children were conservatively managed with follow-up appointments and ‘glue ear’ reviews, only 7 in 1000 had persistent OME that required further otological management. Comparison was made with the results achieved from cohort 6 in the district and those achieved by the BACDA study. The yield of cases with OME identified historically by the SES in the district was 29 in 1000 of those screened. This is not significantly different from national data on SES performance,⁶ which suggested that 26 in 1000 receiving the SES had OME (χ^2 2.512, df 1, $p = 0.113$). It was significantly greater than the SES OME yield reported from cohort 3, UNHS, that had received UNHS (χ^2 15.342, df 1, $p < 0.001$). This suggests that, although UNHS cannot have directly affected the detection and management of middle ear problems, fewer children with previously ‘undetected glue ear’ were reaching school age. Reasons are conjectural, but increased early ENT service provision and general awareness about childhood hearing following UNHS introduction may have had some effect.

Summary

The data from the studies in Waltham Forest services demonstrate that there has been significant local change in the circumstances relating to the SES. In the 1970s and 1980s the SES provided an important backstop to a relatively insensitive IDT screen. There was a higher yield of PCHI from the SES than from the IDT. Some primary and secondary prevention measures and in particular the introduction of neonatal hearing screening have markedly changed this picture. The extent to which the findings from the Waltham Forest studies are generalisable to the UK as a whole is debatable; on the one hand, there are features of the local population (e.g. ethnicity, mobility, deprivation indices) that may be non-typical, and which may affect aspects of screening performance. On the other hand, the

Waltham Forest data are derived from relatively large cohorts, and comparisons of prevalence rates and aetiologies with well-established national data show no significant differences. Furthermore, while some aspects of screen performance (e.g. coverage) may be affected by the non-typical nature of the local population, it is not clear why the nature of the relationships between the performance of different screens (e.g. the general effect of the introduction of universal newborn screening on later screens) should be affected by these local issues. The following points are indicated by Waltham Forest data.

- The prevalence of moderate or worse permanent bilateral hearing impairment was 1.49 in 1000, which is consistent with other national reports. An additional 1.21 in 1000 children had a mild permanent bilateral impairment. These rates are apparently stable.
- The prevalence of unilateral permanent hearing impairment at school age was 0.78 in 1000, with the data indicating a significant reduction in the numbers of severe and profound cases to 0.27 in 1000, probably related to primary intervention (immunisation programmes).
- The introduction of the newborn screen was accompanied by a highly significant reduction in the median age of confirmation of permanent childhood hearing impairment; however, there was much less of a reduction in the upper quartile age of confirmation which remained in the weeks leading up to the fifth birthday. Sixteen per cent of those with a moderate bilateral PCHI, 18% of those with a mild bilateral PCHI and 17% of those with a unilateral hearing impairment who were in their first year of primary school still required identification and referral for hearing assessment at 5 years of age. Reasons for this include the relatively poor sensitivity of the preschool identification of milder hearing impairment, the occurrence of acquired and late-onset cases, and unconfirmed cases moving into the district.
- Historically, the major route to identification of all PCHI was parental or professional concern, followed by the SES (1.1 in 1000). For unilateral hearing impairment alone, it was the SES (0.63 in 1000) followed by parental or professional concern. The introduction of targeted newborn screening resulted in the newborn screen becoming the second main route to identification (second to parental and professional concern for all PCHI, and second to the SES for unilateral alone), with the SES yield reducing to 0.7 in 1000 (all PCHI) and 0.36 in 1000 (unilateral). The introduction of a universal newborn screening programme meant that newborn screening became the main route to identification for all PCHI and for unilateral PCHI, with parental and professional concern in second place (stable at 1.3 in 1000) and the SES yield reduced to 0.34 in 1000 (all PCHI) and 0.07 in 1000 (unilateral).
- The overall prevalence of all PCHI at SES age, excluding minimal but including mild and unilateral, was 3.47 in 1000, indicating a significant increase from the prevalence identified neonatally, in line with previous studies, owing to cases missed by the newborn screening programme, cases moving into district, and acquired or late-onset cases.
- Of the 3.47 in 1000 children with a PCHI at SES age, 1.89 in 1000 required identification after the neonatal screen. It would have been possible to identify 1.52 in 1000 by optimally referring from preschool surveillance a group selected as needing audiological assessment. The most effective targeting appears to be the selection for hearing assessment of the children in attendance at the Child Development Centre. However, 0.37 in 1000 of those not picked up neonatally had no discernible risk factor that would have prompted referral for a hearing assessment, and for their identification reactive referral or SES would be required.
- There has been growing interest in the prevalence and possible effects of minimal hearing impairment in childhood. In these studies, the SES showed a yield of minimal permanent hearing impairment of 3.5 in 1000. Identification did not lead to any active ongoing management in any of the children.
- Both a national study⁶ and the data from Waltham Forest have confirmed just under 3% of those primary school children screened as having OME. New cases amounted to 1.4% and of these one half (0.7%) needed further otological management.
- The DNA rate for follow-up appointments for those failing the SES in Waltham Forest was in the order of 20%; this is very similar to that reported by Fonseca and colleagues.⁶

Chapter 4

Systematic review of the effectiveness of school entry hearing screening

Background

This chapter examines the evidence base for the effectiveness of available school entry hearing screening tests or screening programmes (i.e. combination of screening tests).

As outlined by the UK National Screening Committee in their recommendations for evaluating screening programmes, Wilson and Jungner's 1968 criteria remain the benchmark³⁸ (see also Appendix 1). Those criteria that specifically relate to the screen itself include:

- There should be a reliable, valid and repeatable screening test.
- The screening test should be acceptable, safe and easy to perform.
- The screening test should be sensitive and specific.
- The cost of the screening programme should be commensurate with benefits of early detection.

With reference to these criteria, the review focuses on three broad aspects of evidence base for the effectiveness of school-based hearing screening:³⁸ screen accuracy (i.e. sensitivity: proportion of children with a hearing impairment who have a positive screen test; and specificity: proportion of children without hearing impairment who have a negative screen test), screen performance (i.e. uptake: number of children who take up screening when offered; and yield: i.e. number of cases identified by the screen) and screen effectiveness (i.e. impact of the screen on children's outcomes including language, educational ability and social interaction). The issues of cost and cost-effectiveness of school entry hearing screen are considered in Chapter 5.

The nature of the evidence necessary to address these elements of screening is potentially quite diverse. At one end of the spectrum, for child outcomes, one would want to focus on studies with a comparative design (i.e. a direct comparison of a group of children who receive the hearing screen or programme with a group of children who do not). The prospective randomised controlled trial

(RCT) provides the study design of choice.³⁹ Non-randomised (or observational or naturalistic) comparative designs, such as cohort or case-control studies, may also be useful. The assessment of sensitivity and specificity requires a study design where the hearing outcome results of a screen are compared with those of a reference test undertaken in the same group of children. The quality of this latter type of study depends on factors such as the degree of independence of the application of the screen test and reference.⁴⁰ At the other end of the spectrum, uptake and yield can often be obtained from a relatively simple non-comparative study design where a group of children is offered a screen and followed up over time.

Given that the primary focus of this chapter was to assess alternative school entry hearing screening tests or programmes, the particular approach has been to seek comparative evidence; that is, studies that directly compare the dimensions of accuracy, performance and effectiveness of two or more screening tests or programmes.

Hypotheses tested in the review (research questions)

1. How accurate are the tests used in school-based hearing screening in terms of sensitivity and specificity?
2. What is the performance of school-based hearing screening in terms of yield and uptake?
3. What is the effectiveness of school-based hearing screening in terms of language, education and social outcomes of children?
4. What are the adverse effects of school-based hearing screening?

Methods

Search strategy

Searches for systematic reviews and primary studies of school-based hearing screening were undertaken across the following bibliographic sources: Cochrane Library (Wiley) (CDSR,

CENTRAL, DARE), MEDLINE and MEDLINE In Process (Ovid), EMBASE (Ovid), CINAHL (Ovid), PsycINFO (Ovid), Science Citation Index (Web of Science), ERIC (CSA) and ongoing trial databases (National Research Register, ClinicalTrials.gov and ReFeR) from initial entries up to May 2005. The reference lists of retrieved full-text reports were also checked.

Bibliographic searches were designed and run by an experienced information specialist. Medical subject headings and text terms were chosen to maximise the comprehensiveness and sensitivity of the searches. Initial searches focused on screening related terms. These were later supplemented by additional searches designed to identify studies on test accuracy. Search strategies are listed in Appendix 4.

Inclusion/exclusion criteria

- Study design: systematic reviews and any primary study using a comparative design (i.e. randomised controlled trials, non-randomised controlled trials, cohort studies or comparisons of two or more tests or test protocols) were included.
- Population: included children aged 4–6 years. Studies of children with known hearing impairment or high-risk groups (such as Down's syndrome, cytomegalovirus infection or meningitis) were excluded.
- Screening test or programme: hearing screening comprising of any of one or more of the following tests: sweep PTA, single-frequency PTA, otoacoustic emissions, questionnaires, otoadmittance tests, tympanometry, reflectometry and speech audiometry. Tests should be undertaken in either a primary school or the community (e.g. community clinic, family home or GP surgery) setting. This could include hearing screening as a component of a multifaceted screen such as a school entry medical examination.
- Comparator: no hearing screening or hearing screening based on different tests or test protocols. Studies with no clear comparator were excluded.
- Outcomes: outcomes were sought according to the research questions: (1) test accuracy: sensitivity and specificity or equivalent; (2) screen performance: uptake (i.e. number of children who actually receive screen) and yield (i.e. number of cases identified); and (3) screen effectiveness: language skills, health-related quality of life, communications skills, social interaction and educational performance.

No language restrictions were applied to the inclusion of studies. Study selection was undertaken independently by two reviewers (JS and RT). Disagreements about selection were resolved by discussion.

Data extraction

Data were extracted from included studies on study population, sample size, study characteristics (author, year, country of publication and sample size), study design (method of sampling, details of test and comparison and listing of relevant outcomes), screening procedure (age at testing, choice and combination of screening tools, type and training of the tester, setting of the screen, failure criteria, retest frequency and interval) and study findings. For test accuracy, findings were sought as two-by-two tables so that reported sensitivities and specificities could be checked.

Data extraction was carried out using a predefined data extraction and quality assessment form by a single reviewer (JS) and checked by a second (RT). Any discrepancies were resolved by discussion and where necessary by the mediation of a third reviewer (JB or HF). Furthermore, a clinical expert (SF) in the field checked the data extracted.

Quality assessment

The methodological quality of systematic reviews was assessed using the Oxford Critical Skills Appraisal Programme (CASP) criteria (see Appendix 5).⁴¹ Depending on study design, the US Preventative Services Task Force has proposed a 'levels of evidence' rating for individual screening studies:

- level I: randomised controlled trial
- level II: non-randomised control trial
- level III: cohort or case-control study
- level IV: ecological or descriptive studies (e.g. international pattern time series)
- level V: opinions of respected authorities based on clinical experience, descriptive studies or reports of expert committees.

Given the lack of studies identified by the review that fell into the above designs it was decided to focus quality assessment on the principal issue addressed by included studies (i.e. test accuracy). The Quality Assessment of Studies of Diagnostic Accuracy (QUADAS) tool⁴² was used for this purpose and consists of 14 questions (see Appendix 6). For every study, each question was given a 'yes', 'no' or 'unsure' answer based on whether the criteria were met, not met or it was unclear, respectively. The quality of each article

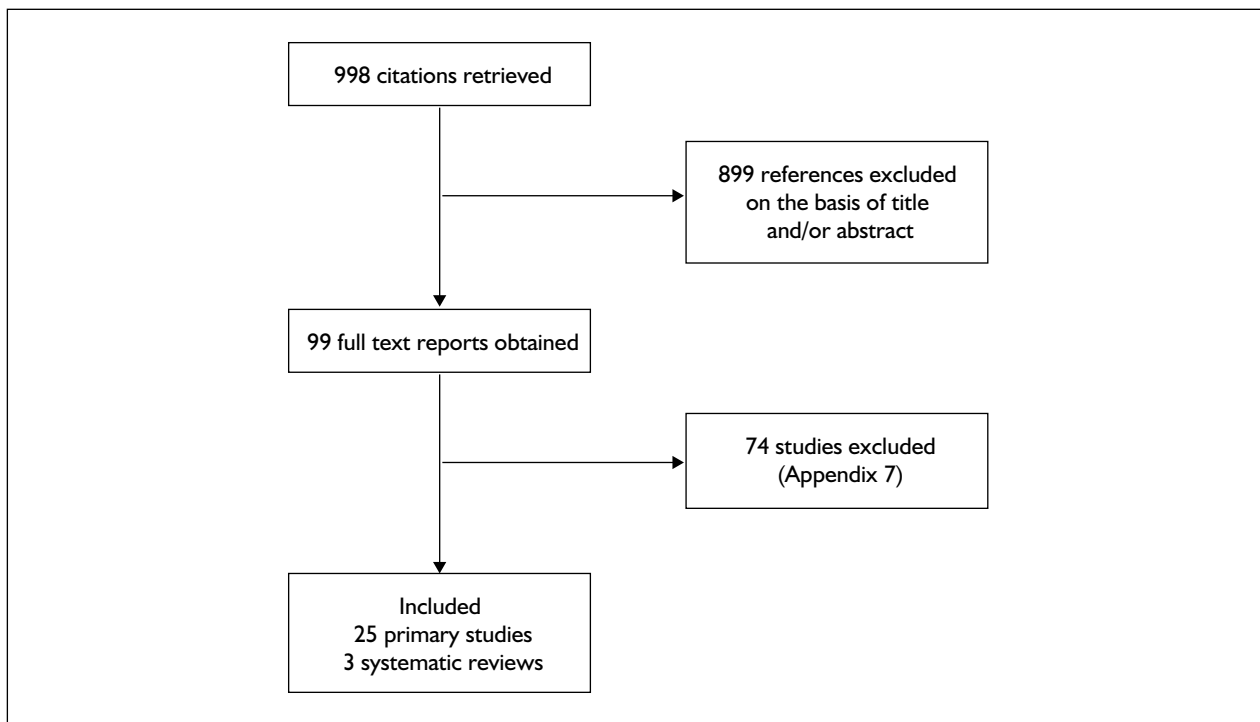


FIGURE 6 Selection process for primary studies

was scored on the basis of the total number of ‘yes’ responses and could therefore range from zero (poorest possible quality score) to 14 (highest possible quality score).

Data presentation and analysis

Results are presented separately for test accuracy, screen performance and screen effectiveness. Given the limited evidence base for screen performance and effectiveness, data pooling was not possible. To facilitate interpretation, sensitivity and specificity results are tabulated according to specific tests. In addition, sensitivity and specificity results of individual studies are graphically presented in the form of summary receiver operating characteristic (ROC) curves.

Studies included in the review

The process of selecting studies for inclusion is summarised in *Figure 6*. In total, 998 citations were identified by the bibliographic searches, the majority from three databases (MEDLINE 464, EMBASE 252 and ERIC 172). Of these citations, 899 were excluded on the basis of their title or abstract for one or more reasons, principally that the study did not address hearing impairment or

was non-comparative. Of the 99 papers retrieved in full, three systematic reviews and 25 primary studies were judged to meet the inclusion criteria of this review. Twenty-three of the included studies were identified from the initial ‘screening’-based searches and two from the follow-up ‘test accuracy’-based searches. The level of agreement in selecting studies between the two reviewers was good (weighted kappa 0.67, 95% CI 0.60 to 0.75).

Studies excluded from the review

The 74 excluded studies and reasons for exclusion are listed in Appendix 7.

Results of review

Previous systematic reviews

Three systematic reviews relating to school entry screening were identified that met the inclusion criteria (*Table 27*). A detailed summary of their quality is provided in Appendix 8. The Cochrane review of screening for OME by Butler and colleagues¹⁸ considered studies on children only up to the age of 4 years and was therefore excluded (but see discussion in Chapter 6).

Barlow and colleagues (1998)⁴³

The systematic review by Barlow and colleagues⁴³ was published in 1998 and examined the issue of the school entry medical examination (SEM). The SEM consisted of vision assessment, a hearing test and a general medical examination by a doctor. The authors also assessed the relative effectiveness of selective SEM (children assessed by a doctor only when there are concerns about their health) compared with routine SEM (all children are examined).

Overall, the methodological quality of the Barlow review was judged to be good. The review stated a clear and comprehensive search strategy that included a number of bibliographic databases including MEDLINE, EMBASE, CINAHL and DARE. Searches were initially designed to identify meta-analyses and RCTs. However, owing to a limited amount of RCT evidence the authors broadened their searches so no restrictions were placed on study design. Reference lists were examined and experts in the field were consulted. Principal investigators were contacted to obtain information about ongoing studies. There were no language restrictions placed on study inclusion. A single reviewer undertook selection of studies for inclusion and it is not stated whether another reviewer checked this or not. Two reviewers assessed and critically appraised the quality of meta-analyses and systematic reviews using published criteria. Primary studies were appraised using an adapted version of Wilson and Jungner criteria for screening programmes. No information was given as to whether studies were selected on the basis of their quality.

The authors included a total of 16 primary studies. The authors reported a 'high' uptake rate for both routine and selective SEM, but did not provide actual figures. Across the identified studies the rate of new 'problems' identified by SEM varied markedly for both routine SEM (27–45 problems per 100 children eligible) and selective SEM (two to six problems per 100 children eligible). It is virtually impossible to interpret the significance of these findings as the 'number of problems' could include any combination of vision, hearing, growth or other physical problems.

Furthermore, the review authors noted that different studies applied different threshold levels for the identification of any particular problem area, such as hearing impairment. Given that studies did not report their follow-up, it was not possible to comment on the sensitivity or specificity or yield of SEM. The one RCT

identified provided follow-up in the year succeeding the trial, of children not selected for SEM. The findings showed that from a cohort of 302 children, 12 were discovered to have serious language development problems and nine had behaviour problems. However, as this trial did not involve the re-examination of children who received SEM, it is impossible to know whether the same number of children would have been missed in the routine medical group. The percentage of children selected for SEM varied across studies from 19 to 73%, this range reflecting the widely differing selection criteria of studies.

The overall conclusion of the authors of this review was that there was insufficient evidence to assess the effectiveness and efficiency of selective or routine SEM.

New Zealand Health Technology Assessment Report (1998)⁴⁴

The New Zealand Health Technology Agency (NZHTA) undertook a review of the effectiveness of preschool and school entrant screening programmes for OME and conductive hearing impairment.⁴⁴

The review report is divided into a number of sections that examine different specific questions. The final section of the report is entitled 'Is there a suitable screening test for OME? And the associated hearing impairment?' and 'What is the evidence of effectiveness of screening programmes for OME and associated hearing impairments?' and is clearly relevant to the present report.

The methodological quality of the review was assessed to be good. Its aims were clearly stated and the authors undertook comprehensive searches of MEDLINE, HEALTHSTAR and CINAHL databases up to March 1998. In addition, reference lists were handsearched and registers of current research consulted. The report did not state whether experts in the field were contacted and the search was restricted to English-language studies and those directly applicable to the New Zealand population, thus limiting its scope. The search strategy was aimed primarily at identifying RCTs, although cohort studies and audits were also included. To be included, studies had to involve more than 30 participants and adequately report demographic details. Studies were quality assessed using a schedule developed by the Group Health Cooperative of Puget Sound in 1996 and adapted by the New Zealand Guidelines Group of the National Health Committee in 1997. The quality criteria used in this schedule were not stated in the report.

TABLE 27 Summary of previous systematic reviews

Review	Review scope			Number and type of included studies ^a	Authors' conclusions
	Population	Screening intervention	Outcomes assessed		
Barlow <i>et al.</i> , 1998 ⁴³	Children entering primary school	Routine or selective SEM with a doctor's contribution	Uptake rates, referral rates, yield, PPV, NPV, sensitivity, specificity, costs, outcome of treatment and patient satisfaction measures	One RCT, two observational comparative studies, and 13 prospective and retrospective observational studies or audits	Insufficient evidence available to assess the effectiveness or efficiency of either the routine or the selective SEM. This review demonstrates the fragility of the evidence on which the SEM is based and questions the ethical basis of this programme
NZHTA, 1998 ⁴⁴	Children aged 2–12 years	Screening techniques for the detection of OME	Sensitivity, specificity, PPV, referral rate, retest frequency	Two cohort studies, three uncontrolled studies and two audit studies	Current research cannot support or refute the effectiveness of screening programmes designed to detect OME and conductive hearing impairment in improving disability-related outcomes
Pirozzo <i>et al.</i> , 2003 ⁴⁷	Children aged 3–12 years	Whispered voice test	Sensitivity and specificity	Five cross-sectional studies	The whispered voice test is an accurate and simple test of hearing impairment that could be used by GPs but has not been adequately evaluated in primary care settings. Differences in accuracy among published studies could be explained by differences in conducting the test. The technique for conducting the test needs to be standardised to optimise the sensitivity of the test, particularly in children

^a Studies that are relevant to the children aged 4–6 years and design as stated by the authors. NPV, negative predictive value; PPV, positive predictive value.

TABLE 28 Studies using whispered voice test in children

	Groen, 1973 ⁴⁸	Dempster and Mackenzie, 1992 ⁴⁹	Prescott et al., 1999, study 1 ⁵⁰	Prescott et al., 1999, study 2 ⁵⁰
Sample size	197	141	177	201
Prevalence (%) of hearing impairment	14	13	31	9
Sensitivity (%; 95% CI)	96 (82 to 99)	90 (69 to 97)	80 (68 to 88)	83 (61 to 94)
Specificity (%; 95% CI)	92 (87 to 95)	90 (84 to 94)	96 (19 to 98)	98 (95 to 99)

Modified from Pirozzo et al. (2003), Table 2.⁴⁸

The review identified no RCTs, five cohort studies and two audits. However, three of these so-called 'cohort' studies were in fact uncontrolled descriptive studies describing the outcomes of children who had received hearing screening. Similarly, the audits describe the experiences of two New Zealand centres undertaking screening for OME. One Dutch study used tympanometry to screen for OME in 2-year-olds over a 2-year period;⁴⁵ persistent cases were randomised for surgery (grommets) versus no surgery, showing no later language outcome differences. Although the ages of the children at the close of this study were within the range of interest here, the screening was earlier. These six studies effectively provide no direct information on the comparative effectiveness of hearing screening in 4–6-year-olds.

In the remaining study, which is relevant to this review, two geographical populations of school-entry Canadian children were compared by Feldman and colleagues in 1980.⁴⁶ One group had been screened in the previous 6–12 months using two-step audiometry and the other group had not. On the basis of a non-significant difference in later audiometric outcomes between the two groups, the authors concluded that screening was ineffective.

The NZHTA authors reported the specificity and sensitivity for detecting OME with otoscopy to be highly dependent on the technique, while the specificity (53–94%) and sensitivity (78–100%) of tympanometry were generally high. Although the report mentions audiometry, TEOAE and parental questionnaire, no specificity or sensitivity values for the tests are provided. It is unclear how systematically the authors of the New Zealand report identified the evidence for OME screen test accuracy.

Pirozzo and colleagues (2003)⁴⁷

This review aimed to assess the accuracy (performance) of the whispered voice test for screening for hearing impairment in adults and children.⁴⁷

The quality of the review was judged to be moderate as no details of the selection of studies were provided. The aims of review were clearly stated. A detailed and comprehensive bibliographic search strategy including all publications until June 2002 was presented and experts were contacted about unpublished work.

To be included, studies had to be cross-sectional and include a reference test (PTA) applied to at least 80% of participants. A total of eight studies met the inclusion criteria, four of which were concerned with adults (17–89 years) and four with children (3–12 years). The authors commented that the quality of included studies was 'modest' based on the Standards for Reporting Studies of Diagnostic Accuracy (STARD) criteria. Only the findings in children will be discussed further here.

The authors stated that they did not undertake meta-analysis given the level of heterogeneity between the methods of the studies and the way in which the whispered voice test was performed. The studies in children used slightly different techniques to conduct the whispered voice test and the threshold for audiometry ranged from 20 to 35 dB HL. The studies generally showed a good level of specificity (92–98%), but a poorer sensitivity (80–96%) (Table 28). The authors of this review did not report any other long-term outcomes such as language or educational attainment.

The authors concluded that the whispered voice test is an accurate and simple test of hearing impairment. However, they note that the sensitivity is much lower for children than adults and therefore may fail to identify hearing impairment in a large proportion of children. Furthermore, differences in accuracy among published studies appeared to be explained by differences in test conduct (e.g. loudness of the whisper, and the most appropriate use of letters, numbers or words for testing and tester).

Primary studies

Scope of included studies

One of 25 included studies was a cohort study while the remainder were comparative cross-sectional studies (*Table 29*). Only the cohort study⁴⁶ attempted to address the question of the effectiveness of the hearing screen in children at school entry. The remaining cross-sectional studies were primarily concerned with the question of the test accuracy. Thirteen studies reported screen performance.^{51–62} No studies reported either screen yield or adverse effects.

Studies were included on the basis of involving children between 4 and 6 years of age. However, only four studies included exclusively children whose age fell specifically within this range.^{52,57,63,64} Most studies had varying age ranges, with some including children as young as 2.5 years and others including children as old as 14 years. Four studies failed to report a specific age, although they did describe the population as 'kindergarten' or 'preschool' and therefore were included on this basis.^{46,58,65,66} Overall, studies included similar proportions of boys and girls.

A range of different test comparisons was found. Some studies compared individual tests (e.g. tympanometry versus PTA), whereas others compared combinations of tests or different protocols for the same test. The majority of studies used PTA as the reference test because this was their current method of school entry screening testing and this is the standard test for measuring hearing threshold levels.⁶⁷

Where details were reported, screening was carried out in a variety of situations: within the school or primary care/community facility, and under tightly controlled conditions (e.g. a soundproof room) or not. In the majority of studies a qualified professional conducted the screen, such as a school (or public health) nurse or an audiologist.

The conditions being sought varied across studies; for instance, tympanometry and otoscopy are not hearing tests but predictors of conductive hearing impairment. Studies failed to describe explicitly the conditions being sought or the severity of hearing impairment identified.

Quality of included studies

Three studies were in languages other than English^{56,64,66} and were data extracted with the help of a translator. Consequently, these studies could only be partially quality assessed and so were omitted from the following quality analysis.

The median QUADAS score across the remaining 21 cross-sectional studies was 8 (out of a possible maximum score of 14), with a range of scores from 5 to 12 (*Table 29*). Based on QUADAS scores, studies were categorised as of 'poor' quality: less than 7; 'moderate' quality: 7–9; and 'good' quality: greater than 9. On this basis, one study was classed as poor quality; seven studies as moderate quality and 13 studies as good quality. The majority of low scores were the result of poor reporting where the authors had failed to describe particular methodological aspects of their study. The breakdown of quality scoring across individual studies is shown in Appendix 9.

Those criteria that were consistently met across the studies were questions 1, 4, 5, 6, and 7; that is, adequate time between index and reference test, representative sample and spectrum of children tested and tests interpreted independently of each other. Such was the level of reporting that some criteria, questions 2, 3, 10, 11, 12 and 13, were not achieved by any or very few (less than five) studies.

Question 2 considers whether or not the selection criteria have been clearly described. The fact that most studies did not describe their selection criteria, or were unclear about them, means that one has to consider that these studies may be influenced by selection bias. This brings into question the internal validity and generalisability of the studies. Question 3 considers whether the reference standard is likely to classify the target condition correctly. As there is no recognised reference test for hearing screening in children (see above, 'Scope of included studies'), all studies received an 'unclear' for this question; as none of them used a comprehensive audiological assessment as their reference standard one cannot tell whether any of the tests they used are 100% effective. This means that there may be non-differential misclassification bias within the studies. Thus, the sensitivities and specificities of these studies may be overestimates or underestimates. Questions 9 and 10 take into account whether or not the assessors were blinded to the results of the index test when carrying out the reference test and vice versa. Lack of blinding is a source of observer bias potentially leading to further misclassification and invalidity of the results. Question 12 looks at whether or not the clinical data available to the testers were the same as those that would be available in clinical practice. All studies received an 'unclear' for this question as none of them clearly states what data were available to them at the time of testing.

TABLE 29 Summary of characteristics of included studies

Source	Design	No. of children (% male)	Age, mean (SD) or range	Test(s) evaluated	Reference standard	Setting	Tester(s)	Quality score	Outcome domains
Abou Haidar et al., 2005 ⁶⁸ France	Cross-sectional	360 (58)	7.4 years	'Audio 4' picture test	PTA	NR	NR	7	Test accuracy
Feldman et al., 1980 ⁴⁶ Canada	Cohort	763 (NR)	NR	VASC	None	School; no further details	Public health nurse	NA	Screen effectiveness, screen performance
FitzZaland and Zink, 1984 ⁵⁵ USA	Cross-sectional	3510 (NR)	4 years to 7 months	Pure tone sweep test, audiometric Rhinne test, audiometric Weber test, tympanometry, acoustic reflex	Combination of history plus pure tone and air and bone conducted thresholds plus tympanometry plus acoustic reflex plus speech tests	School; previously assessed for noise level and judged to be the quietest	Public health nurses and audiometric aides	8	Test accuracy
Gomes and Lichtig, 2005 ⁶⁹ Brazil	Cross-sectional	133 (50)	3 years	Parental questionnaire	Examination plus tympanometry plus pure tone sweep test	NR	Researcher and volunteers (employees from a local nursery)	6	Test accuracy
Hamill, 1988 ⁵⁸ USA	Cross-sectional	576 (NR)	NR	VASCA and pure tone sweep test	Pure tone sweep test	NR	School vision and hearing personnel	10	Test accuracy, screen performance
Hammond et al., 1997 ⁶³ Australia	Cross-sectional	685 (NR)	4–5 years	Questionnaire	PTA, pure tone sweep test and ENT examination	NR	Nurses	8	Test accuracy
Hind et al., 1995 ⁶⁵ UK	Cross-sectional	2860 (NR)	NR	Questionnaire	Pure tone sweep test	School	Audiometricians	9	NR
Holtby et al., 1997 ⁷⁰ UK	Cross-sectional	674 (NR)	5–6 years	Tympanometry PTA	Examination	School	School nurses	11	Test accuracy, screen performance

continued

TABLE 29 Summary of characteristics of included studies (cont'd)

Source	Design	No. of children (% male)	Age, mean (SD) or range	Test(s) evaluated	Reference standard	Setting	Tester(s)	Quality score	Outcome domains
Lyons et al., 2004 ⁵³ Australia	Cross-sectional	528 (53)	6.2 years 1 month to 7 years 9 months	Tympanometry DPOAE	Pure tone sweep test	School: non-soundproofed room with ambient noise levels between 34 and 51 dBA	Audiologist	8	Test accuracy, screen performance
Maragno and Teatini, 1983 ⁶⁶ Italy	Cross-sectional	114 (NR)	NR	Speech test	Hearing assessment including PTA	Hearing centre: silent-noise controlled cabinet	NR	NA	Screen accuracy
McCurdy et al., 1976 ⁷¹ USA	Cross-sectional	93 (NR)	3 years 6 months to 4 years	Tympanometry and stapedius reflex	PTA	School: vacant classroom, not noise controlled	NR	8	Screen accuracy
Nienhuys et al., 1994 ⁶¹ Australia	Cross-sectional	180 (52)	<16 years	Otoscopy, tympanometry, pneumotoscopy, reflectometry	PTA	School: mobile soundproofed test booth	Paediatric otologist, medical officer, audiologist and a registered nurse	8	Screen accuracy, screen performance
Nozza et al., 1997 ⁷² USA	Cross-sectional	66 (NR)	5–10 years	TEOAE	Pneumatic otoscopy plus sweep test screening plus PTA	School library	Audiologist and three audiology graduate students	11	NR
Olusanya, 2001 ⁵¹ Nigeria	Cross-sectional	359 (48)	6.7 years	Questionnaire, tympanometry, otoscopic examination	Two-stage audiometry pure tone sweep test plus PTA	NR	NR	7	Screen accuracy
Orlando and Frank, 1987 ⁷³ USA	Cross-sectional	100 (NR)	2 years 6 months to 6 years	Pure tone sweep test using an audioscope, pure tone sweep test using an audiometer	PTA	Audiometric test booth	Audiologist and clinician	11	Screen accuracy

continued

TABLE 29 Summary of characteristics of included studies (cont'd)

Source	Design	No. of children (% male)	Age, mean (SD) or range	Test(s) evaluated	Reference standard	Setting	Tester(s)	Quality score	Outcome domains
Pang-Ching et al., 1995 ⁵² Hawaii	Cross-sectional	172 (50)	4 years	Pure tone sweep test, tympanometry, pneumatic otoscopy, acoustic reflectometry	Pure tone sweep test, tympanometry, pneumatic otoscopy, acoustic reflectometry	Hearing clinic: acoustically treated test room	NR	8	Screen accuracy, screen performance
Prescott et al., 1999 ⁷⁴ South Africa	Cross-sectional	205 (NR)	3–7 years	Whispered voice test	PTA	School classroom	Fourth year BSc students	8	Screen accuracy, screen performance
Ritchie and Merkein, 1972 ⁵⁷ USA	Cross-sectional	162 (64)	4–5 years	VASC, two protocols	PTA	Auditory test booth (location not stated)	NR	9	Test accuracy, screen performance
Rodriguez and Melguizo-Yepe, 1994 ⁵⁶ Columbia	Cross-sectional	80 (NR)	5–14 years	PTA, pure tone sweep test, questionnaire	Tympanometry plus pneumatic otoscopy	NR	NR	NA	NR
Roush et al., 1992 ⁵⁹ USA	Cross-sectional	204 (50)	3–4 years	Tympanometry	Otoscopy	NR	Paediatric otolaryngologist	10	Screen accuracy, screen performance
Roush and Tait, 1985 ⁶² USA	Cross-sectional	75 (NR)	3–4 years	PTA, four protocols	PTA	Day care centre with ambient noise	Graduate audiology student	7	Screen accuracy, screen performance
Sabo et al., 2000 ⁵⁴ USA	Cross-sectional	573 (55)	5–9 years	Pure tone sweep test, TEOAE	PTA	School: non-soundproofed room	School nurse, speech audiologist, volunteers	8	Screen accuracy, screen performance

continued

TABLE 29 Summary of characteristics of included studies (cont'd)

Source	Design	No. of children (% male)	Age, mean (SD) or range	Test(s) evaluated	Reference standard	Setting	Tester(s)	Quality score	Outcome domains
Schell, 1970 ⁶⁴ USA	Cross-sectional	134 (NR)	4-5 years	VASC	PTA	NR	NR	NA	Screen accuracy
Skurr and Jones, 1981 ⁷⁵ Australia	Cross-sectional	564 (50)	3-15 years	Watch tick, two tone, hand-held beeper, whispered voice test, PTA	PTA plus tympanometry	School: quietest area available	Child health nurses	5	NR
Square et al., 1985 ⁶⁰ USA	Cross-sectional	113 (NR)	2 years 6 months to 6 years	Bone conduction, PTA	Impedance audiometry	School	NR	8	Screen accuracy, screen performance
DPOAE, distortion product otoacoustic emission; NR, not reported; VASC, verbal audiometric screening for children.									

TABLE 30 Test accuracy of parental questionnaires

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Gomes and Lichtig, 2005 ⁶⁹	Parental questionnaire	Score = 0 fail Score > 0 pass	Examination, tympanometry and pure tone sweep test	Not stated	71%	64%
Olusanya, 2001 ⁵¹	Parental questionnaire	NR	PTA and pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	34%	95%
Hammond et al., 1997 ⁶³	Parental questionnaire	Positive response to ≥ 1 question	PTA, pure tone sweep test and ENT examination	>30 dB at 1 kHz >20 dB at 2 kHz >25 dB at 4 kHz	56%	52%

Finally, question 13 addresses the issue of whether any interpretable or intermediate test results were reported. The fact that most studies failed to report these could mean either that there were no uninterpretable test results or that they simply were not recorded, which in turn could be hiding any practical problems encountered in actually applying the tests.

Findings

Test accuracy

A wide variety of hearing tests (and protocols) was evaluated. Although the reference test varied, most studies used pure tone audiometric testing. Presented below are the sensitivity and specificity values, grouped and tabulated according to the screening test compared where possible to PTA with a hearing impairment cut-off ranging from 15 to 30 dB at various frequencies. Where studies reported multiple comparisons, the sensitivity and specificity values are reported for each comparison separately (Tables 30–37). The two-by-two tables for sensitivity and specificity, where available, are presented in Appendix 10.

Parental questionnaires

Three studies examined the accuracy of parental questionnaire against PTA (Table 30). Both sensitivity (34–71%) and specificity (52–95%) range widely.

Impedance audiometry/tympanometry

Nine studies reported 11 different comparisons of the accuracy of impedance audiometry/tympanometry compared with PTA, otoscopy or a combined test reference standard (Table 31). Against otoscopy the sensitivity (50–90%) and specificity (65–97%) of tympanometry was moderate to good. However, compared with

PTA, the test accuracy of tympanometry appeared to be more variable (sensitivity of 40–90% and specificity of 57–85%) and dependent on the tympanometry fail criteria used. FitzZaland and Zink⁵⁵ reported a good level of tympanometry accuracy (sensitivity 40–93% and specificity 91–100%) against a reference of multiple tests. In part, these findings reflect the differing aims of the tests: tympanometry and otoscopy assess pathology (presence of middle ear effusion) and, unlike PTA, are not tests of hearing sensitivity.

Spoken word tests

Five studies reported four comparisons of spoken word tests, VASC or SVEP (a test using speech signals), compared with PTA. Sensitivity (51–100%) and specificity (93–96.8%) were moderate to good.

Otoscopy

Two studies compared otoscopy with PTA. Both sensitivity (23–89%) and specificity values (60–93%) were highly variable.

Audiometry

Five studies reported comparisons of the pure tone sweep test with PTA. The sensitivity (86–100%) and specificity (65–100%) values were generally high. Indeed, the study by Orlando and Frank⁷³ showed that these high values were consistent across 6-month age groupings between 4 and 6 years.

TEOAE

Sabo and colleagues⁵⁴ reported the sensitivity (63%) and specificity (91%) of TEOAE compared with PTA in a smaller study with just 66 children, only 61 of whom completed. They reported

TABLE 31 Test accuracy of impedance audiometry

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Pang-Ching <i>et al.</i> , 1995 ⁵²	Tympanometry	Modified ASHA (1990) criteria	Pure tone sweep test, tympanometry, pneumatic otoscopy, acoustic reflectometry	Score ≥ 3	73%	85%
Olusanya, 2001 ⁵¹	Tympanometry	Second test non-type A tympanogram	PTA and pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	50%	83%
Lyons <i>et al.</i> , 2004 ⁵³	Tympanometry	Non-type A tympanogram	Pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	85%	91%
McCurdy <i>et al.</i> , 1976 ⁷¹	Tympanometry and stapedius reflex	Type B or C tympanogram or Type A tympanogram and no stapedius reflex	PTA	Clark (7) criteria	71%	65%
Nienhuys, <i>et al.</i> , 1994 ⁶¹	Tympanometry	Normal +100 to -99 daPa and 0.3–6 ml	PTA	>25 dB at 0.5–4 kHz	40–90% ^a	62–83% ^a
Rousch <i>et al.</i> , 1992 ⁵⁹	Tympanometry	Traditional ASHA (1990) criteria	Otoscopy	Medical attention required	27%	99%
Rousch <i>et al.</i> , 1992 ⁵⁹	Tympanometry	Modified ASHA (1990) criteria	Otoscopy	Medical attention required	64%	97%
FitzZaland and Zink, 1984 ⁵⁵	Tympanometry	1 = Type B or C with pressure ≤ 150 mmH ₂ O 2 = Type B or C with pressure ≤ 175 mmH ₂ O 3 = Type B or C with pressure ≤ 200 mmH ₂ O 4 = Type B only	Combination of history, pure tone and air and bone conducted thresholds, tympanometry, acoustic reflex and speech tests	Various according to test	1 = 93% 2 = 93% 3 = 91% 4 = 40%	1 = 91% 2 = 95% 3 = 99% 4 = 100%
Holtby <i>et al.</i> , 1997 ⁷⁶	Tympanometry and stapedius reflex	Negative pressure of ≥ -200 mm OR inability to show compliance at <0.3 ml compliance volume OR inability to show a stapedius reflex at 80 or 100 dB	Examination that included PTA, tympanometry and ear examination	Not stated	83.7%	73.6%

ASHA, American Speech and Language Hearing Association.
^a Dependent on tympanometry fail criteria used.

sensitivity in the range of 67–100% depending on the fail criterion, but these figures were based on results from just six ears.

Combined tests

The study by Lyons and colleagues⁵³ evaluated four protocols of combined DPOAE and tympanometry compared with PTA. The accuracy

of the combined test was high: sensitivity 96–98% and specificity 83–96%.

Other tests and protocols

Three studies assessed tests and protocols not considered by any other studies. FitzZaland and Zink⁵⁵ looked at the audiometric Rinne test relative to audiological assessment and found high

TABLE 32 Test accuracy of spoken word tests

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Ritchie and Merklein, 1972 ⁵⁷	VASC (protocol 1)	Two consecutive incorrect responses or two out of three incorrect responses at 15 dB	PTA	≥ 15 dB at 550 Hz in one ear ≥ 15 dB any two frequencies in one ear ≥ 20 dB for any single frequency for either ear	51%	96%
Ritchie and Merklein, 1972 ⁵⁷	VASC (protocol 2)	Two consecutive incorrect responses or two out of three incorrect responses at 15 dB or incorrect response to bird whistle	PTA	≥ 15 dB at 550 Hz in one ear ≥ 15 dB any two frequencies in one ear ≥ 20 dB for any single frequency for either ear	59%	93%
Hamill, 1988 ⁵⁸	VASC	Failure to respond at two out of three of the 19 dB presentations	Pure tone sweep test	> 20 dB at 0.5, 1, 2 and 4 kHz	87%	96%
Maragno and Teatini, 1983 ⁶⁶	SVEP test	NR	Audiological assessment including PTA	NR	100%	94%
Prescott <i>et al.</i> , 1999 ⁷⁴	Voice test	Correctly identifying less than 50% of the test words	PTA	> 35 dB HL	83.3%	96.8%

SVEP, Sweep Visual Evoked Potential.

TABLE 33 Test accuracy of otoscopy

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Olusanya, 2001 ⁵¹	Otoscopy	NR	Pure tone sweep test and PTA	> 20 dB at 0.5, 1, 2, 4 kHz	56%	62.4%
Nienhuys <i>et al.</i> , 1994 ⁶¹	Otoscopy	Wax discharge or lack of tympanic membrane activity	PTA	Clark (7) criteria 0.5–4 kHz	23–89% ^a	60–93% ^a

^a Dependent on fail criteria used.

sensitivity (91%) and specificity (99.67%). The study by Square and colleagues⁶⁰ compared bone conduction tests combined with PTA to impedance screening and found poor sensitivity (26%) and specificity (6.6%). Finally, Pang-Ching and colleagues⁵² found that reflectometry had poor sensitivity (23%) and specificity (56%) compared with otoscopy results.

Influence of testing environment

Only five of the included studies reported using a soundproofed environment in which to carry out the test.^{52,57,61,66,73} All other studies either reported a non-soundproofed environment or failed to report where the test was conducted. There appeared to be no consistent differences in the sensitivities and specificities of those studies

TABLE 34 Test accuracy of sweep audiometry

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Sabo <i>et al.</i> , 2000 ⁵⁴	Pure tone sweep test	>25 dB at 0.5 kHz and >20 dB at 1, 2 and 4 kHz	PTA	NR	87%	80%
Orlando and Frank, 1987 ⁷³	Pure tone sweep test (audioscope)	>25 dB at 0.5, 1, 2 and 4 kHz	PTA	>30 dB	4–4.5 years: 90% 4.5–5 years: 82% 5–5.5 years: 100% 5.5–6 years: 88%	4–4.5 years: 71% 4.5–5 years: 65% 5–5.5 years: 90% 5.6–6 years: 84%
Orlando and Frank, 1987 ⁷³	Pure tone sweep test (audiometer)	>25 dB at 0.5, 1, 2 and 4 kHz	PTA	>30 dB	4–4.5 years: 91% 4.5–5 years: 91% 5–5.5 years: 100% 5.5–6 years: 100%	4–4.5 years: 98% 4.5–5 years: 98% 5–5.5 years: 98% 5.5–6 years: 97%
FitzZaland and Zink, 1984 ⁵⁵	Pure tone sweep test	>25 dB at 0.5 and 4 kHz and >20 dB 1 and 2 kHz	Combination of history, pure tone and air and bone conducted thresholds, tympanometry, acoustic reflex and speech tests	Various, dependent on test	93%	99%
Holtby <i>et al.</i> , 1997 ⁷⁰	Pure tone sweep test	No response at 20 dB in either ear at any frequency	Examination including PTA, tympanometry and ear examination	NR	86%	70.2%

TABLE 35 Test accuracy of TEOAE

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Sabo <i>et al.</i> , 2000 ⁵⁴	TEAOE	Response at three frequencies of ≥ 3 above the noise floor with a minimum 70% reproducibility at each frequency and a 90% or greater stability	PTA	NR	63%	91%
Nozza <i>et al.</i> , 1997 ⁷²	TEAOE	Various	PTA	NR	67–100%	80–98%

that reported using a controlled test environment and those that did not.

Influence of study quality

There was no clear difference in sensitivity and specificity of studies judged to be of good methodological quality (i.e. QUADAS score ≥ 9) or

poor to moderate methodological quality (i.e. QUADAS score < 9).

Summary ROC curve

To provide some overall summary of screen test accuracy results, the subgroup of sensitivity and specificity values where the reference test applied

TABLE 36 Test accuracy of combined tests

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
Lyons <i>et al.</i> , 2004 ⁵³	Protocol 1 DPOAE and tympanometry	DPOAE SNR ≥ 5 dB at 1.9 kHz and tympanometry results normal	Pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	97%	86%
Lyons <i>et al.</i> , 2004 ⁵³	Protocol 2 DPOAE and tympanometry	DPOAE SNR ≥ 5 dB at 1.9 kHz and tympanometry results normal	Pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	97%	83%
Lyons <i>et al.</i> , 2004 ⁵³	Protocol 3 DPOAE and tympanometry	DPOAE SNR ≥ 11 dB at 1.9 kHz and tympanometry results normal	Pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	98%	74%
Lyons <i>et al.</i> , 2004 ⁵³	Protocol 4 DPOAE and tympanometry	DPOAE SNR ≥ 5 dB at 1.9 kHz and SNR ≥ 11 dB at 3.8 kHz and tympanometry results normal	Pure tone sweep test	>20 dB at 0.5, 1, 2 and 4 kHz	96%	95%

SNR, signal to noise ratio.

TABLE 37 Test accuracy of other tests

Source	Test evaluated	Definition of hearing impairment	Reference standard	Definition of hearing impairment	Sensitivity	Specificity
FitzZaland and Zink, 1984 ⁵⁵	Audiometric Rinne test	Reporting having heard the tone more loudly at the mastoid location	Audiological assessment	Various, dependent on test	91%	99.67%
Square <i>et al.</i> , 1985 ⁶⁰	Bone conduction and PTA	No response at +10 dB and -10 dB OR no response at = 10 dB	Impedance screening (tympanometry and reflex)	Negative peak pressure at >-150 mm/H ₂ O air pressure or having no pressure peak at all	26%	6.6%
Pang-Ching <i>et al.</i> , 1995 ⁵²	Acoustic reflectometry	Scores of 6-9	Tympanometry and pneumatic otoscopy	Score ≥ 3	23%	56%

was PTA was plotted in the ROC space (*Figure 7*). Different types of test are given different symbol shading. The diameter of the symbol reflects the sample size of the study, with larger studies having a larger symbol. 'Good' tests with both high sensitivity and specificity (e.g. tympanometry and pure tone sweep test) tend to occupy the top left of the plot. In contrast, poorer tests with lower sensitivity and specificity (i.e. parental questionnaires) tend to occupy the bottom right of the plot. It is important to recognise there are at least two caveats in interpreting this figure. First, either or both of two different conditions (middle ear pathology, hearing impairment) are being sought by different studies. Secondly, both the

referral criteria for the screening test and the criterion definition of hearing impairment for the reference test varied across studies.

Screen performance

The uptake of the screening test was reported by 13 studies (*Table 38*) and across a variety of screening tests. Regardless of the test applied the uptake rates appeared to be high, ranging from 87.5 to 100%. These high values reflect the fact that the studies are experimental test accuracy assessments rather than 'real world' and community-based screening evaluations. None of the studies looked at reported their yield of true cases.

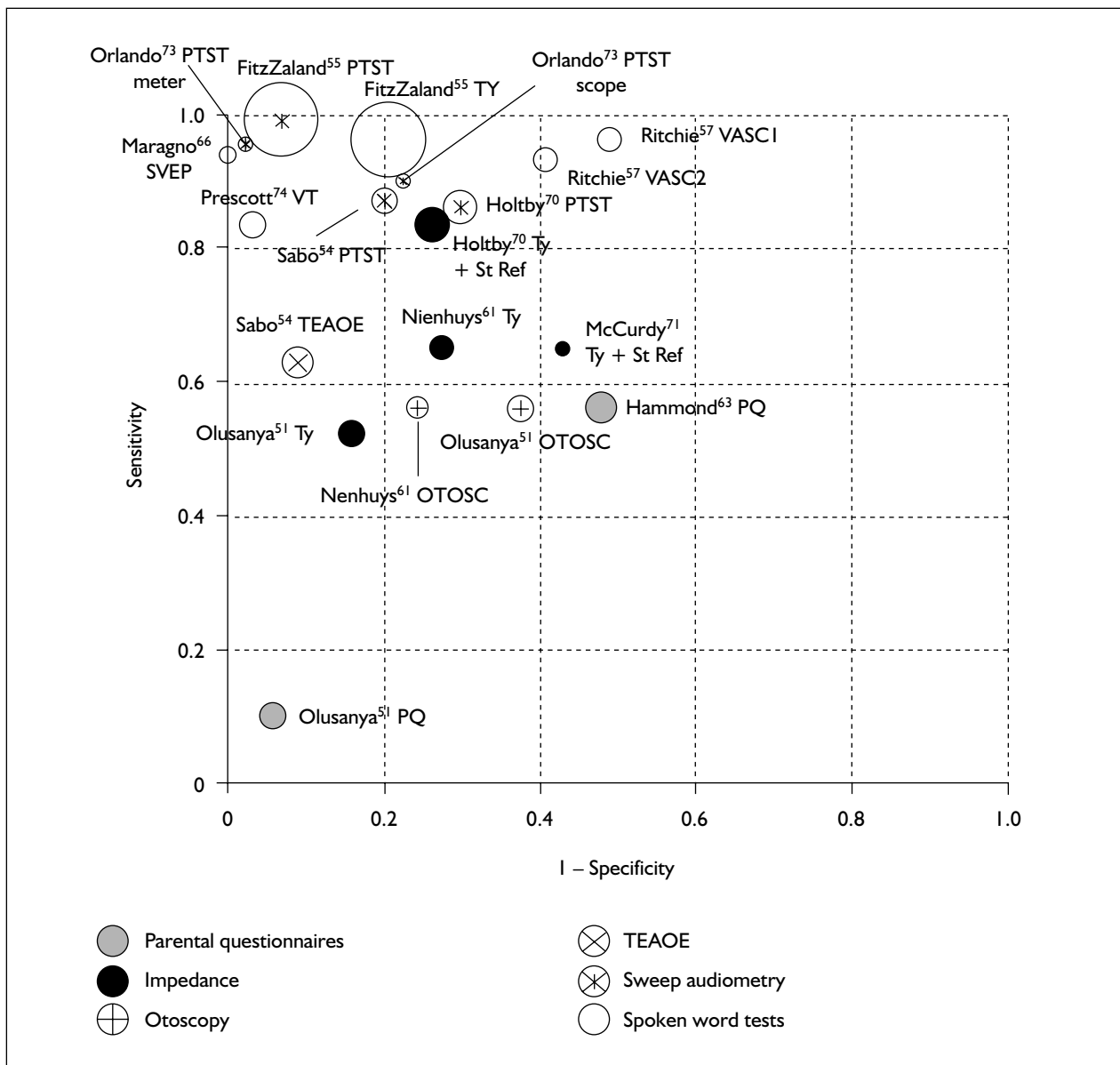


FIGURE 7 Summary ROC (should be interpreted with the results in Tables 30–37)

TABLE 38 Uptake for each study

Source	Uptake (%)
Ritchie and Merklein, 1972 ⁵⁷	100
FitzZaland and Zink, 1984 ⁵⁵	100
Holtby <i>et al.</i> , 1997 ⁷⁰	91
Lyons <i>et al.</i> , 2004 ⁵³	100
Nienhuys <i>et al.</i> , 1994 ⁶¹	71
Olusanya, 2001 ⁵¹	88
Pang-Ching <i>et al.</i> , 1995 ⁵²	100
Prescott <i>et al.</i> , 1999 ⁷⁴	94
Rodriguez and Melguizo-Yepe, 1994 ⁵⁶	87.5
Rousch <i>et al.</i> , 1992 ⁵⁹	100
Rousch and Tait, 1985 ⁶²	100
Sabo <i>et al.</i> , 2000 ⁵⁴	99
Square <i>et al.</i> , 1985 ⁶⁰	100

Screen effectiveness

It was not possible to assess the potential effectiveness of interventions for children identified by the SES since only the study by Feldman and colleagues⁴⁶ reported outcomes related to screening test effectiveness. This retrospective cohort study compared two groups of 730 children from different geographical areas in Ontario, Canada. One group received hearing screening (VASC by a public health nurse) before school entry while the other group did not. Hearing impairment was assessed (PTA at 0.5–4 kHz) in both groups at 6–12 months after the hearing screening. As the study found no statistically significant difference in the prevalence of hearing impairment in the groups after 6–12

months (unscreened group 16.8% versus screened group 14.1%) the authors concluded that preschool hearing screening was ineffective in the sense of leading to interventions which resolved the (presumably temporary) hearing impairments.

However, there are some problems with the design of this study that limit the strength of its conclusions. First, it is likely to have been underpowered to detect the small improvement in prevalence of hearing deficit seen in the screened group. Secondly, the observational nature of the study made it open to a number of potential biases. Although the authors attempted to match the children by selecting two geographical areas with similar socio-economic class, there remains considerable potential for differences in the baseline characteristics of the children, so-called selection bias. The baseline characteristics of the children in the two groups were not reported. The assessors conducting the hearing test at 6–12 months may not have been blinded to the screening status of child. In addition to a lack of effectiveness of the hearing screen, there are other potential explanations for the similar level of prevalence of hearing impairment in the two groups (e.g. non-compliance with treatment; ineffective treatment or wrong timescale).

Adverse effects

None of the included studies reported any adverse effects of screening.

Summary of findings

- There is only level III evidence for the effectiveness of preschool hearing screening, from a single, poor-quality, observational comparative study. Furthermore, this single study was inconclusive in whether preschool screening was more effective than no screening in detecting hearing impairment.
- No studies were identified that have assessed the long-term impact of preschool hearing screening on outcomes including educational, language and social outcomes, or on the

effectiveness of interventions for children identified with hearing impairment via the SES.

- Several studies with an unacceptable variability in their quality have assessed the accuracy of different hearing screening tests in preschool children. Given the unacceptable variability in methodological quality and reporting of these studies, lack of clarity in the cases of hearing impairment detected (e.g. transient versus permanent hearing impairment), variation in reference test and threshold level for hearing deficit, and range of control over the settings in which these tests were applied, it is difficult to interpret and compare their results. Accepting these caveats and selecting the subset of studies using PTA as the reference test, the findings suggest that:
 - Studies comparing various screen protocols of pure tone sweep audiometry report high sensitivity and specificity for full PTA and therefore appear to be suitable tests for screening.
 - Spoken word tests are reported to be a viable option because of their potential acceptable levels of specificity and sensitivity.
 - Depending on referral criteria, TEOAEs have potentially high specificity, but somewhat lower sensitivity.
 - Tympanometry and acoustic reflectometry have variable sensitivity and specificity.
 - Parental questionnaire and otoscopy have poor sensitivity and specificity. Therefore, these tests are likely to be less suitable for screening.
 - There is insufficient evidence to comment on the accuracy of combinations of tests.
- A small number of studies indicated a generally high uptake in this age group. However, given the experimental design of the studies, and that they were assessing test accuracy rather than programme accuracy, these findings cannot necessarily be generalised to the uptake of the screen in real-world community screening settings.
- No studies were found that assessed the potential adverse effects or yield of hearing screening for preschool children.

Chapter 5

Cost-effectiveness and cost-effectiveness acceptability of the school entry hearing screen

Methods

Aims and objectives

The overall aim of this part of the study was to estimate the cost-effectiveness and cost acceptability of alternative strategies for SES. To achieve this aim the principal objectives were:

- to conduct a systematic review of the economics literature
- to estimate the health-related quality of life and utilities associated with the SES programme
- to estimate the relative costs of the SES
- to relate the costs and health-related quality of life and utilities and compare alternative models of SES.

Overall approach

The analysis assessed the cost-effectiveness and cost acceptability of alternative models of SES. It was designed to investigate the extent to which the differences in the cost-effectiveness strategies for hearing screening result in differences in costs, resource use, health status, and hearing-related disability and quality of life. The perspectives of the NHS and education services, patients and family were used to approximate a societal perspective.

To address the research questions a decision-analytic model was developed to synthesise clinical and economic data from a number of sources. The model was used to estimate the relative cost-effectiveness of alternative SES programmes and no SES. As recommended by guidelines for economic evaluation,⁷⁷⁻⁷⁹ the alternatives for comparison were chosen to reflect the range of SES programmes reported in the survey of UK current practice (Chapter 2). A composite SES programme (SES-C) was defined that included a combination of the categories or types of SES tests reported in the survey of current practice. To define the composite, the probability of each type of test being used was estimated. Using the data from the survey of current practice, SES-C was defined as pure tone sweep audiometry (99%) and tympanometry (1%). This was used to weight the probability and cost data relating to the costs and

accuracy of individual types of test. The key alternatives compared to SES-C were:

- universal SES, using pure tone sweep audiometry only (SES-PTS)
- universal SES, using parental questionnaire only (SES-PQ)
- universal SES, using tympanometry only (SES-T)
- universal SES, using spoken word tests only (SES-SW)
- no SES
- targeted SES.

The decision model includes events relevant to the effectiveness, subsequent diagnosis and treatment/management, patient outcomes, resource use and costs of the screening packages. The outcomes assessed with the model were cost per quality-adjusted life-year (QALY) gained, and cost per year gained with no, minimal or mild hearing impairment (YNHI). The cost per QALY gained was the primary outcome measure. QALYs weight life-years by the utility or value attached to health states and improvements in health and are recommended in cases where the health outcome of interest is change in morbidity rather than simply survival.⁷⁷⁻⁷⁹ If the data on utility required to estimate QALYs are derived from inappropriate instruments or low-quality evaluations, then the estimates of QALYs may be inaccurate, which would bias the results of the analysis. An alternative measure is years with no disability due to hearing impairment. However, there were insufficient data to extrapolate from years with different levels of hearing impairment to estimate years with different levels of disability. Therefore, the cost per year with no to mild hearing impairment (YNHI) was included as an alternative measure, to assess whether the results of the economic model would differ substantially according to the method used to evaluate and value hearing levels. The outcome of cost per year with no, minimal or mild hearing impairment is a potentially less sensitive measure of health associated with hearing impairment. It assumes that minimal and mild hearing impairment have no impact on the overall utility or value of a year

with these levels of hearing impairment compared with years of life with no hearing impairment. In addition, any benefit accruing to years of life with more severe hearing impairment are not included in the estimate of outcome; in other words, the value of years of life with moderate to severe hearing impairment is set to zero. If different methods of SES affect the distribution of children between different levels of hearing impairment, and each level of hearing is associated with a different utility or value, then use of the YNHI will result in misestimation of the benefit of alternative SES programmes. The cost per true positive case of hearing impairment detected is an alternative outcome measure that is less sensitive than the cost per YNHI. It was decided to include this in the sensitivity analysis only if the conclusions of the economic model did not differ substantially between cost per QALY gained and cost per YNHI gained.

The time-horizon used for the primary analysis was from the day of screening up to 1 year. Secondary analyses explored longer time-horizons of 6 years (from the day of screening to secondary school age) and 11 years (from the day of screening to 16 years of age). As outlined in Chapter 1, children in the UK enter school at around 5 years of age and are usually screened in their first (reception) year at school. Differences between education authorities mean that they enter school any time from their fourth birthday up to their fifth birthday. So, the most appropriate population for the analysis is children 4–6 years of age; that is, 4 years and 1 day to 5 years and 364 days (i.e. the time at which school entry screening could take place in the UK). The selected age range also reflects an assumption that identification and management of previously undetected hearing impairment relatively early in a child's school life are beneficial.

Data for the model were derived from the survey of current SES practice reported in Chapter 2, the systematic review of test accuracy and effectiveness reported in Chapter 4, and two additional reviews of the economics literature and national databases and statistical sources for the UK.

Reviews of economic literature and databases

A systematic review of economic evaluations of screening for hearing impairment in children aged 4–6 years (the age range of interest for this economic evaluation) was conducted. The objectives of the review were:

- to assess the costs, effectiveness and cost-effectiveness of school entry screening
- to identify decision models reported in the literature
- to identify economic data for the model used in this study.

The review used a focused systematic search of studies and databases that report resource use, quality of life data, costs or patient outcomes associated with screening for hearing impairment.

A second review of literature that reported resource use, costs or outcomes of management interventions for hearing impairment was also conducted. The objective of this review was to identify economic data that could be used to populate the economic model.

The search strategies for both reviews were implemented in the following electronic databases:

- MEDLINE (1966 to 2005 week 3)
- EMBASE (1980 to 2005 week 31)
- Cumulative Index to Nursing & Allied Health Literature (CINAHL) (1982 to August 2005 week 5)
- Econlit (1969–2002 and 2003–2005)
- Cochrane Library (Wiley) NHS Economic Evaluation Database (NHS EED) (2005 Issue 2)
- Office of Health Economics Database (OHE HEED) (July 2005 issue).

The searches were limited to electronic databases. The detailed search strategy for the review of economic evaluations of screening is reproduced in Appendix 11. Appendix 12 details the search strategy for the management of hearing impairment review. The search strategies were developed by the project team and an information specialist with extensive experience in literature searching. The search strategy was modified and optimised for each electronic database. The economic terms used on MEDLINE, CINAHL and EMBASE were adapted from the York Centre for Reviews and Dissemination (CRD)⁷⁸ filter. The clinical terms for the screening search strategy were based on filters developed at the University of Birmingham for the review of effectiveness of SES (Chapter 4). The NHS National Electronic Library for Health (<http://libraries.nelh.nhs.uk>), the PRODIGY guideline website (www.prodigy.nhs.uk) and the Scottish Intercollegiate Guideline Network website (www.sign.ac.uk) were used to identify relevant interventions and search terms for the management of hearing impairment review. Only

interventions with a clinical benefit recommended in these guidelines were included in the search strategy for management of hearing impairment.

The syntax of the search strategies was mapped accordingly, to translate directly the thesaurus of MEDLINE, EMBASE and CINAHL. The search strategy imposed no language, date or other similar limitations.

A screening form for inclusion/exclusion was used to screen titles and abstracts and exclude any studies that did not report resource use, utility values or costs related to SES or management of hearing impairment. Articles were only rejected on initial screen if the reviewer could determine from the title and abstract that the article did not meet the prespecified inclusion/exclusion criteria. If a title/abstract could not have been rejected with certainty, the full text of the article was obtained for further evaluation to assess whether they met the inclusion/exclusion criteria. One reviewer (GV) then screened all of the retrieved papers.

The following inclusion and exclusion criteria were applied for the review of economic evaluations of screening studies (Appendix 13):

1. The studies were based on primary data or used data from systematic literature reviews, reported detailed data on costs and outcomes for extraction and use in the economic model, were conducted in a range of settings (e.g. education services, primary/secondary/tertiary healthcare, other local community services, or the family home) and were generalisable to the UK setting.
2. The paper reported data relevant to the population of interest (children between 4 and 6 years of age).
3. The evaluations compared at least two of the following interventions: PTA, tympanometry, acoustic reflex, otoadmittance tests, ABR, medical examinations (which entail a hearing screening), distraction tests, behavioural tests, speech perception tests, questionnaires, otoacoustic emissions and no screen.
4. The paper reported at least one of the following outcomes: year with no or mild/moderate disability due to hearing impairment, year with moderate or severe disability due to hearing impairment, QALYs gained, utility measure and health status measure.
5. The paper reported at least one of the following types of economic data: resource use, costs or utilities associated with hearing

screening programmes and subsequent management interventions.

6. Resource use and cost were reported separately.

The full paper was included in the review of the cost-effectiveness of screening only if it met criteria 1, 2 and 3 and at least one of 4, 5 and 6. To identify decision models and data for the economic model, studies were included if they met criterion 1 and one or more of criteria 2–6.

For the review of management strategies for hearing impairment the following inclusion and exclusion criteria were applied (Appendix 14):

1. The studies were based on primary data or used data from systematic literature reviews, reported detailed data on costs and outcomes for extraction and use in the economic model, were conducted in a range of settings (e.g. education services, primary/secondary/tertiary healthcare, other local community services, or the family home, and were generalisable to the UK setting.
2. The paper reported data relevant to children with identified hearing impairment aged from birth to 12 years, undergoing any of the following interventions: hearing aids, autoinflation, middle ear ventilation, myringotomy/grommets, adenoidectomy, speech and language therapy, hearing tactics (family, community, school), referral to specialists or cochlear implantation (only for comparative purposes).
3. Studies assessed one of the following outcomes: year with no or mild/moderate disability due to hearing impairment, year with moderate or severe disability due to hearing impairment, QALYs gained, utility or health status.
4. Studies reported resource use, costs or utilities associated with subsequent management interventions.
5. Studies reported resource use and cost separately.

To identify data on the costs or outcomes of management strategies for hearing impairment to populate the economic model, studies were included if they met criterion 1 and one or more of criteria 2–5.

A second reviewer (LD) independently screened any papers where the first reviewer (GV) was unclear of inclusion. Any uncertainties in the reviewers' assessment of the studies were resolved by discussion and, when necessary, in consultation with the rest of the project team. Reviewers were

not masked to the source and authors of the studies.

Data were extracted to populate the economic model using a data abstraction form. The form was based on the criteria to assess abstracts for the NHS EED database (Appendix 15). Data were extracted from included studies on: participants – study population; study design including type of intervention and perspective; screening procedure including comparator interventions and setting of the screen; resource use including choice and combination of screening tools; costs (direct and indirect where reported) and outcomes, including information of synthesis of costs and benefits, data on quality of life, dates to which data relate, discounting rates and side-effects; statistical/sensitivity analyses; and study findings.

All economics papers included in the analysis were quality assessed. The purpose of this assessment was to examine whether the methodology was appropriate and the results were valid and generalisable to other settings (with focus on the NHS). Quality assessment was based on the critical appraisal criteria cited in the CRD Report Number 6.⁷⁸ Quality assessment questions were included in the data extraction form (see Appendix 15) and the papers were scanned accordingly.

Economic model

The decision-analytic model was developed and validated by discussion with experts in the provision of hearing and school entry screening

services. TreeAge Pro 2005 software was used. A static decision tree model was used. The time-horizon of the model is finite and limited for both the primary and secondary analyses. A static decision-tree model works well in analysing chance events with limited recurrence or change over time (such as the probability that a person has hearing impairment or not). This is the case for most events associated with SES. The main exception is the occurrence of transient hearing impairment due to OME. OME can resolve and recur more than once in a 12-month time-frame. However, the consequences of an initial episode of OME detected by the SES (including subsequent recurrences) are not likely to last for more than 12 months. In addition, OME is not likely to result in long-term hearing impairment and only minimal or mild hearing impairment within an episode of OME. Therefore, it was decided to estimate the mean number of recurrences of transient hearing impairment and use these to model the impact on costs and outcomes of transient hearing impairment (and associated recurrences) at the end of the 1-year time-horizon (6- and 11-year time-frames for the secondary analyses).

The first split in a decision-tree model is a ‘decision node’ (sometimes called choice node) and is represented by a square box. Decision nodes reflect a choice to be made between alternatives. Later splits occur at ‘chance nodes’, which are represented by circles. Chance nodes occur when there is a number of subsequent events that could happen. Each event is assigned a

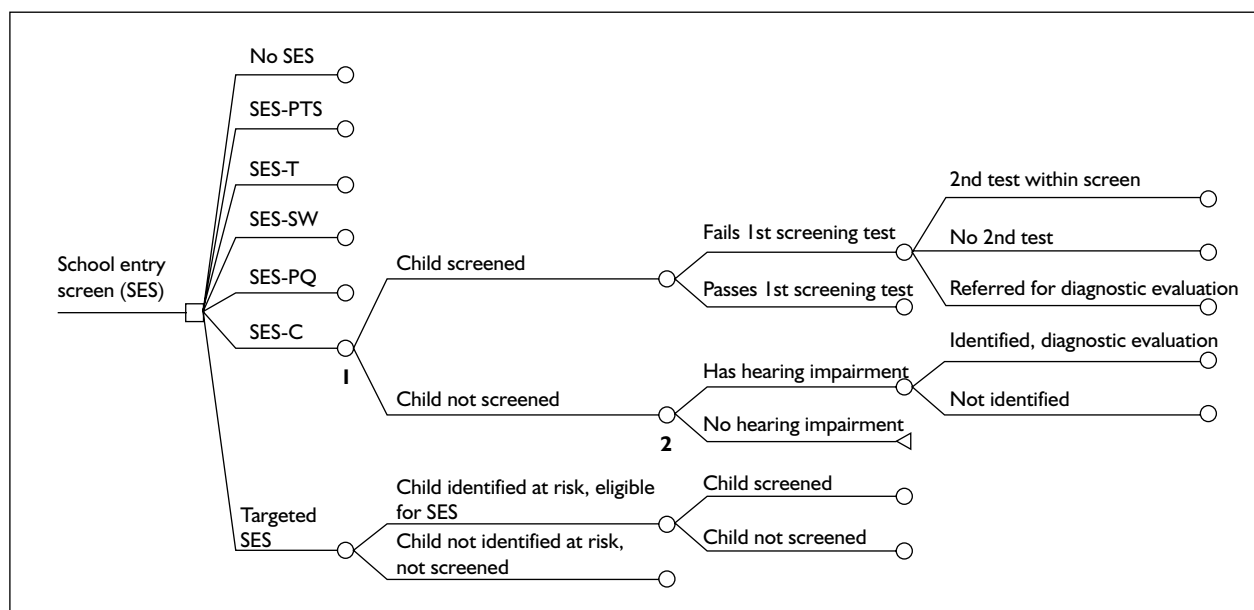


FIGURE 8 Decision tree, choice of screening method

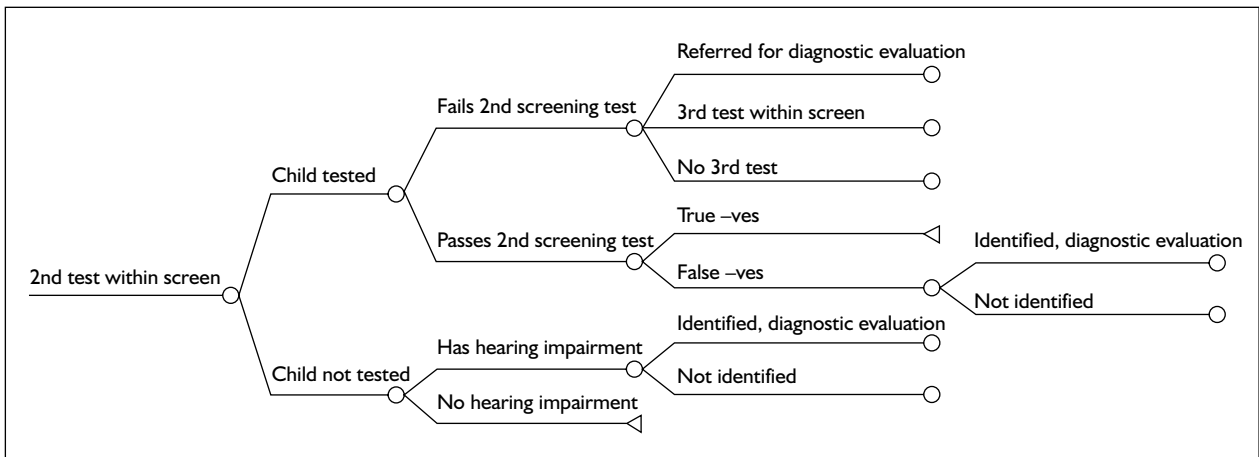


FIGURE 9 Decision subtree for third test within a screen

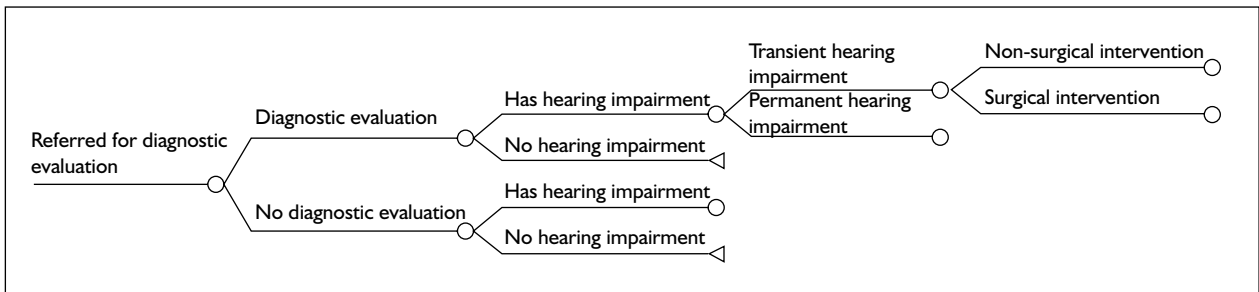


FIGURE 10 Decision subtree for children referred for diagnostic evaluation

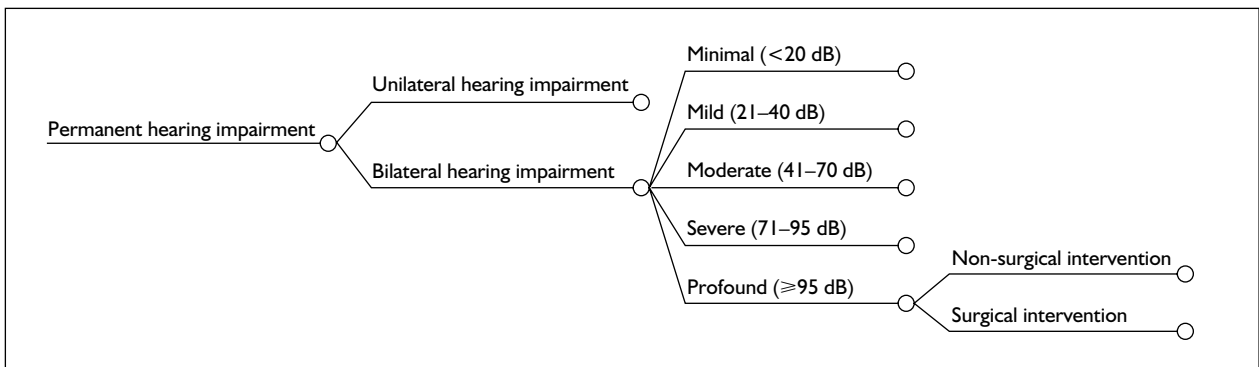


FIGURE 11 Decision subtree for permanent hearing impairment

probability that it will occur. The potential outcomes resulting from a chance node must be all-inclusive and mutually exclusive, so that the probabilities for each chance node sum to one. Triangles represent terminal nodes, to signify the last stage in the model.

A simplified structure of the decision tree is shown in Figures 8–12. These figures outline the paths followed after SES-C and targeted SES. The structure of the model is based on the UK school entry hearing screening practice, where the majority of services carry out a two-stage test

procedure for each child screened who has a positive test result with the first test, before referral for diagnostic evaluation (Figures 8 and 9). Children who have hearing impairment and who are not screened, or children who have been screened but not identified as having a hearing impairment may be identified in other ways (e.g. parental or teacher concern) and referred for a diagnostic evaluation.

The model includes up to three consecutive tests for children who fail the first and second tests (Figure 9). It is assumed that children who fail the

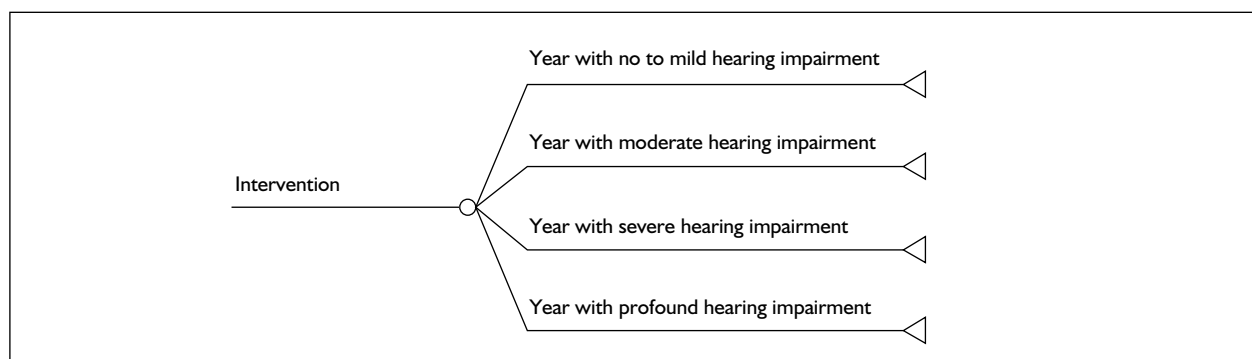


FIGURE 12 Outcomes of hearing impairment

third test within a screen will be referred for diagnostic evaluation (*Figure 10*).

Children who have hearing impairment may have permanent or transient hearing impairment (*Figure 10*). Whether permanent or transient, the hearing impairment may be unilateral or bilateral, at different levels of severity (*Figure 11*). Children with minimal to mild hearing impairment may be managed by watchful waiting or monitoring or non-surgical interventions. Children with more severe hearing impairment may be managed by surgical or non-surgical interventions (*Figure 11*).

Finally, *Figure 12* illustrates the end-points for children with hearing impairment, whether identified or not.

Variable estimation

The decision model required three categories of data: the likelihood of events occurring; the resource use and costs of those events, and the outcomes associated with those events. The overall approach and sources of data used for variable estimation for each of these categories is described below. Estimation of each variable used in the model was to some extent determined by the data available.

Likelihood of events

The probability of whether a child was screened or referred for a diagnostic evaluation was estimated from the survey of current practice reported in Chapter 2. These included the probability that: a child is screened for the first time, a child who fails the first test is scheduled for a second test within the screen or referred for diagnostic evaluation, a child who fails the second test is scheduled for a third test within the screen or referred for diagnostic evaluation, and a child scheduled for a second or third test is actually tested. Table distributions were used to derive the

mean and distribution used in the Monte Carlo simulation for the probabilistic sensitivity analysis (PSA). Each estimate from the survey was given equal weight in the table distributions.

The probability that a child failed or passed the first and subsequent screening tests and that the test results were true or false positive and true or false negative was estimated from the studies included in the systematic review of accuracy reported in Chapter 4. The probability that a child failed or passed the first test in the screen was estimated as the number of children failing divided by the number of children tested. The probability that a child passed or failed subsequent screening tests and/or diagnostic evaluation was conditional on having failed previous screening tests. If data were available from two or more studies, table distributions were used to derive the mean and distribution used for the PSA. The estimate from each study included in the distribution was weighted by the sample size of the study. This means that the estimates from larger studies were assumed to be more accurate than those from smaller studies. If data were only available from one study, theoretical minimum and maximum values were used in a triangular distribution, to reflect the high level of uncertainty associated with single estimates.

The estimates of the probability of all other events in the model were estimated from a number of sources. These included the Waltham Forest study reported in Chapter 3, published prevalence surveys, published surveys of clinical practice, published treatment guidelines, reviews and intervention studies.^{12,17,81–83} In most cases it was not possible to combine estimates from different studies into table distributions for these other events, so triangular distributions of mean or most likely estimates with minimum and maximum were used.

Resource use and costs

The costs of resources used as inputs to screening and management interventions were estimated. The costs were calculated as the product of resource use and unit costs for each screen and subsequent events. For each cost item data on resource use and unit costs were extracted from the reviewed literature and databases, nationally agreed prices, local practice, service standards and guidelines. The costs for the 6- and 11-year analyses were adjusted to net present values using the rate recommended by the UK Treasury at the time of analysis (currently 3.5% for both costs and outcomes; NICE, 2004⁷⁷). All costs were standardised to a single price year, 2004, using a health service price index.⁸³

The costs of screening and diagnosis were broken down into fixed costs, which include the capital cost of equipment, and variable costs, which include maintenance costs, the costs of supplies and consumables and the costs of staff time. The annual equivalent cost of screening and diagnostic equipment was estimated by discounting the acquisition price of the equipment over an estimated life of 8 years, at 3.5% per annum (UK Treasury recommended rate; NICE, 2004⁷⁷). The cost per case of equipment, maintenance, consumables and staff was estimated by dividing the annual equivalent cost by the throughput or number of children screened or diagnosed with the equipment. The number of children screened or diagnosed was estimated from the survey of current practice reported in Chapter 2. The costs of equipment and supplies and the throughput of children for diagnostic equipment were estimated from local purchasing data in Manchester. The salaries of staff were estimated from national unit costs of health and social service staff.⁸³

The costs of surgical interventions following a diagnosis of hearing impairment were derived from national statistics and published literature.⁸⁴ The costs of non-surgical treatment (hearing aids) included salary costs of staff to fit the hearing aids, cost of follow-up monitoring and replacement, cost of consumables, and maintenance and repair of hearing aids. The resources used to fit and monitor the use of hearing aids and follow-up maintenance were estimated from published studies, national statistics and expert opinion.^{83,84}

Where more than one estimate for each cost item was obtained, the range of values found was used to generate a distribution for the simulation analysis. The distribution for each variable included the minimum, mean or median and

maximum values found. Where possible a mean value and measure of variance (e.g. standard deviation or 95% CI) were derived and used to derive a distribution. If this information was not available, minimum and maximum estimates of cost were used to estimate a triangular distribution for the PSA.

Outcomes, utility values and QALYs

For the primary analysis, the final outcomes of years with no or mild disability due to hearing impairment, QALYs gained and true cases identified were estimated. The years with no or mild hearing impairment were estimated by giving a weight of one to final outcomes of no, minimal or mild hearing impairment and weights of zero to moderate, severe or profound hearing impairment.

The utility values to attach to no hearing impairment, minimal or mild hearing impairment were estimated from the population norms for people under the age of 25 years.⁸⁵ The utility values to attach to moderate, severe and profound hearing impairment were estimated from a published economic evaluation of the benefit of hearing aids.⁸⁶

Data analysis

Cost-effectiveness analysis compares the costs and benefits of two or more healthcare interventions with the aim of providing information that can be used to maximise the level of benefits (health effects) relative to the resources available. Incremental cost-effectiveness ratios (ICERs) are used to relate differences in consequences and costs between alternatives.⁸⁷ ICERs were calculated as: (Expected cost of A – Expected cost of B)/ (Expected outcome A – Expected outcome B). Statistical measures of variance around the ICERs were not calculated, since standard methods of analysis do not allow these to be interpreted in any meaningful way. Specifically, for positive ICERs (i.e. cost per QALY >0), a lower ICER is preferred to a higher one. For negative ICERs there is no clear decision rule. A negative ICER may occur when one intervention is both more costly and less effective than another and is not cost-effective, or when an intervention is less costly and more effective than another and is clearly cost-effective. No predefined target ceiling ratio (i.e. the maximum a decision-maker is willing to pay for a unit of effect) for cost-effectiveness was chosen. This was because there is no evidence on what a single target ceiling ratio should be. A range of ceiling ratios was used, from decision-makers being willing to pay £0 to gain 1 QALY to

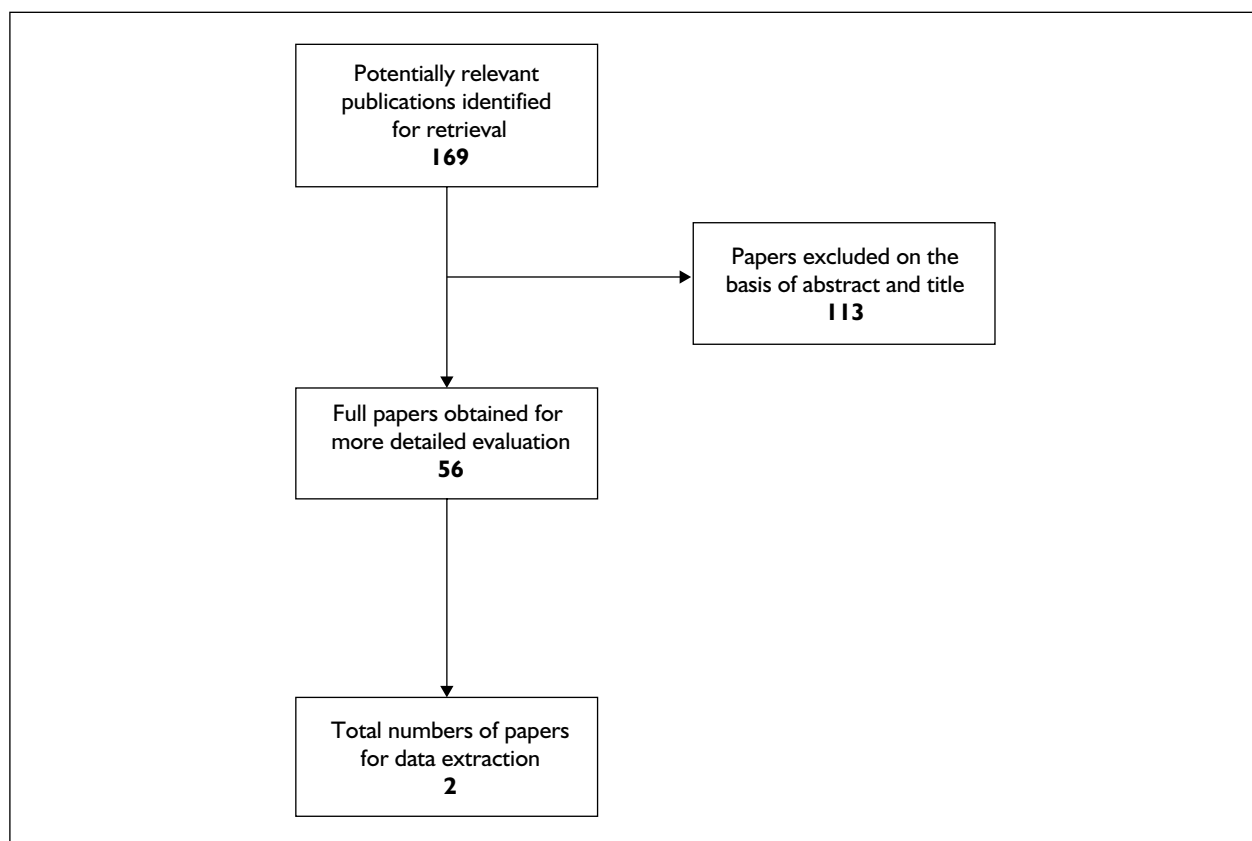


FIGURE 13 Flow diagram of included studies for the cost-effectiveness of screening

decision-makers being willing to pay £30,000 to gain 1 QALY. The ICER was the primary outcome measure used to compare each alternative with the composite universal SES programme (SES-C).

PSA was used to generate mean expected costs and outcomes and statistical measures of expected variance around the likely estimate of each variable in the model and its distribution. Each variable was assigned a base-case or average value and a distribution of possible values. The probabilistic analysis sums the results of multiple analyses (iterations). Each iteration samples values for the variables at random from the specified distributions. The sampling method used was Monte Carlo, expected value. The simulation software was TreeAge Pro 2005 plus Healthcare module.

Cost-effectiveness acceptability curves (CEACs) were plotted and used as a method of summarising the uncertainty around the generated cost-effectiveness ratios. CEACs plot the probability that an intervention is cost-effective against the value of a ceiling ratio (i.e. the maximum a decision-maker is willing to pay for a unit of effect).

The CEAC estimates the probability that SES-C is cost-effective. This is done by first bootstrapping the estimates of cost per QALY (ICER) from the PSA. The proportion of bootstrapped estimates where the cost per QALY is lower than the ceiling ratio is calculated out of the total number of bootstrapped estimates of the ICER. This is repeated for each of the ceiling ratios (in this case the ceiling ratios were £0 per QALY gained to £30,000 per QALY gained, in increments of £1000). The probability that SES-C is cost-effective is then estimated as the proportion of bootstrapped estimates of the ICER that are lower than each ceiling ratio. These estimates are plotted graphically, against each of the ceiling ratios, to derive a CEAC.

Net benefit statistics were estimated by revaluing the bootstrapped estimates of QALYs, using the ceiling ratios or willingness to pay (WTP) to gain one unit of outcome used for the CEAC analysis (i.e. £0 per QALY gained to £30,000 per QALY gained, in increments of £1000). For each WTP threshold, the net benefit (NB) is estimated as

$$NB = E * WTP - C$$

where E is the incremental QALY gained by an intervention, WTP is willingness to pay to gain 1 QALY, and C is the incremental cost of the intervention.

The CEAC summarises the information at each value of WTP to gain a QALY. The net benefit statistic gives an estimate of the monetary value of a QALY or other measure of effectiveness.

Sensitivity analysis was used to explore the impact of structural uncertainty by estimating costs, effects, ICERs and CEACs for each of the alternative outcomes estimated in the analysis, for the impact of universal newborn hearing screen on the numbers of children with unidentified hearing impairment at school entry, and for temporary versus permanent hearing impairment.

Results

Systematic review of the cost-effectiveness of screening and of management strategies

The search strategy for the cost-effectiveness of screening identified 169 articles. Of these, 113 were excluded. The full texts of 56 articles were obtained and reviewed. Data from two papers were extracted for inclusion in the economic model (*Figure 13*). Neither paper reported a full economic evaluation (*Appendix 16*).

One paper reported a cost study⁸⁸ to compare the costs associated with a TEOAE infant screening programme, a TEOAE school entry programme and a pure tone screening school-entry programme. There was no justification for the selection of the comparator programmes; however, pure tone screening is used in routine practice in the UK and so makes the comparison relevant to this review. The perspective was not clearly stated, making it difficult to judge whether all relevant costs had been assessed. The data for the screening programmes were obtained from observational studies in Brisbane. A total of 1305 infants entered the infant screening programme. The age of this population was 2 months. The school screening programme sample for TEOAE was 940 children, with a mean age of 6.2 years, recruited from 22 primary schools throughout Brisbane. No selection criteria were reported. The same schoolchildren were tested with PTA.

The results indicated that the costs for TEOAE infant screening were higher than either school screening programme. Moreover, costs for the

school TEOAE programme were marginally greater than for the school pure tone programme. The cost per hearing-impaired child with sensorineural or mixed impairment of at least a moderate degree was substantially greater than the cost per hearing-impaired child identified with any hearing-impairment, across all programmes. The ultimate yield of hearing-impaired cases was not affected by the age at screening. The difference between the costs of children with hearing impairment for all programmes is related to the low yield of cases diagnosed with sensorineural/mixed hearing impairment and the higher yield of subjects with conductive, and possibly transient pathologies. The costs for the infant programme were substantially less than most reports in the literature of TEOAE-based universal neonatal screening programmes. With regard to the first school programme, the use of TEOAE resulted in a marginally higher cost per child and cost per child with hearing impairment. Higher total programme costs were incurred in the first school programme as opposed to the second one owing, in part, to difference in the cost, maintenance and efficiency of the screening equipment. No indirect costs were included, but this omission is unlikely to affect the model's results. Costs and quantities were reported separately; however, no sensitivity analysis or any other statistical analysis was performed to evaluate the uncertainty around costs of quantities. The study did not discount costs owing to the short time-frame of the analysis. The price year was not reported.

The second paper reported an effectiveness study of hearing screening⁸⁹ that included some cost information. The aims were to evaluate impedance measurements against PTA as a screening method for the detection of middle ear changes associated with hearing impairment in infant school children. Justification of the choice of PTA as the comparator programme was that it represents usual practice. The study used an NHS perspective. Indirect costs were not included in the analysis. The cost data were collected on the sample of children observed in the study. The study did not discount costs owing to the short time-frame of the analysis. No sensitivity analysis or any other statistical analysis was used to evaluate the uncertainty around costs of quantities. The price year of the resources was 1998. The study did not formally relate costs to outcomes.

The electronic and bibliographic searches of papers reporting resource use, costs or outcomes of management interventions identified 960

TABLE 39 Probability of events related to screening and diagnosis

Item/event	Mean	Range
Child is screened	0.92	0.56–1.00
Fails first screening test	0.08 ^a	0.05–0.26 ^a
Referred for diagnostic evaluation following first screen test	0.37 ^b	0.27–0.47 ^b
Child attends diagnostic evaluation	0.80 ^c	0.60–1.00 ^c
Hearing impairment (true positives)	0.53 ^a	0.23–0.83 ^a
No hearing impairment (true negatives)	0.99 ^a	0.98–1.00 ^a
Referred for second test within screen	0.42 ^b	0.32–0.52 ^b
Misses second test within screen	0.08 ^b	0.00–0.44 ^b
Fails second screening test	0.50 ^a	0.22–0.78 ^a
Referred for diagnostic evaluation following second screen test	0.50 ^b	0.17–0.83 ^b
Third test within screen	0.10 ^b	0.00–0.20 ^b
Referred for diagnostic evaluation following third screen test	0.85 ^b	0.70–1.00 ^b

^a Systematic review of effectiveness, Chapter 4 of this report.^{54,55}
^b Survey of current practice, Chapter 2 of this report.
^c Waltham Forest study, Chapter 3 of this report.

potentially relevant titles and abstracts (Appendix 11). Of these, 824 were excluded on the basis of title and abstract and 136 full-text papers/abstracts were collected for more detailed evaluation. Of these 136 papers, no studies could be used to populate the model. Thirty-four were excluded because they did not report detailed data on costs and outcomes for extraction and use in the economic model, 33 studies were not generalisable to the UK setting, 19 papers evaluated the wrong intervention, 16 looked at a different population cohort, 12 papers assessed a different outcome measure, seven analyses did not report resource use and costs separately, five studies reported utilities not associated with management interventions, three were not based on primary data collection or systematic reviews, and seven were excluded for other reasons.

Data used as inputs to the economic model

The probability of using PTA or pure tone sweep audiometry in SES programmes in the UK was estimated as 0.99 (survey of current practice, Chapter 2) for the first and second tests within a screen. The remaining services used tympanometry. The probability of using PTA or pure tone sweep audiometry for a third test within a screen was reduced to 0.95 and the probability of tympanometry increased to 0.05 (survey of current practice, Chapter 2).

Table 39 gives the probabilities of events related to screening for and diagnosis of hearing impairment. These data were derived from the survey of current practice, the Waltham Forest

study and the systematic review of effectiveness reported in Chapters 2, 3 and 4, respectively.

Table 40 shows the probability of having hearing impairment and the distribution of hearing impairment between transient and permanent and by severity of hearing impairment. To estimate the probability of hearing impairment it was assumed that only children with bilateral hearing impairment, lasting for more than 1 month, were likely to have hearing impairment.⁹⁰ The probability for transient hearing impairment or OME lasting for more than a month was calculated from published literature.⁹¹ The prevalence of permanent hearing impairment was estimated from published studies.^{12,17,30}

Table 41 presents the probability of interventions for hearing impairment by different levels and types of hearing impairment. Children with transient hearing impairment will have either surgical or non-surgical intervention. Children with minimal, mild or moderate permanent unilateral hearing impairment will have either no intervention (or watchful waiting) or hearing aids. Children with severe or profound permanent hearing impairment will have either hearing aids or surgery (i.e. cochlear implant).

Table 42 shows the staff and equipment used for screening and the unit costs estimated for the analysis. The acquisition costs of equipment were estimated as: screening audiometer = £950 plus VAT, screening tympanometer = £2147 plus VAT (£1995–2300) and otoscope = £80. The estimate of throughput (number of children screened) for

TABLE 40 Probability and distribution of hearing impairment in the general population of children

Item/event	Mean	Range
Prevalence of hearing impairment	0.078 ^{a-c}	0–0.16 ^{a-c}
Transient hearing impairment	0.96 ^c	0.88–1.00 ^c
Unilateral transient hearing impairment, given transient hearing impairment	0.56 ^c	0.2–0.72 ^c
Permanent hearing impairment	0.04 ^{a,b}	0–0.12 ^{a,b}
Permanent unilateral hearing impairment, given permanent hearing impairment	0.6 ^a	0.3–0.9 ^a
Minimal unilateral hearing impairment (<20 dB)	0.58 ^a	0.28–0.88 ^a
Mild or moderate unilateral hearing impairment (21–70 dB)	0.20 ^a	0.15–0.25 ^a
Severe or profound unilateral hearing impairment (>71 dB)	0.22 ^a	0.18–0.27 ^a
Permanent bilateral hearing impairment, given permanent hearing impairment	0.04 ^{a,b}	0.10–0.70 ^{a,b}
Minimal permanent bilateral hearing impairment (<20 dB)	0.20 ^{a,d}	0–0.60 ^{a,d}
Mild permanent bilateral hearing impairment (21–40 dB)	0.36 ^a	0.31–0.41 ^a
Moderate permanent bilateral hearing impairment (41–70 dB)	0.23 ^{a,b}	0.18–0.28 ^{a,b}
Severe permanent bilateral hearing impairment (71–95 dB)	0.10 ^{a,b}	0.05–0.15 ^{a,b}
Profound permanent bilateral hearing impairment (≥95 dB)	0.11 ^{a,b}	0.06–0.16 ^{a,b}

^a Watkin *et al.* (2007).³⁰
^b Fortnum *et al.* (2001).¹²
^c Midgley *et al.* (2000).¹⁷
^d Niskar *et al.* (1998).⁸²

TABLE 41 Probability of interventions for hearing impairment

Item/event	Mean	Range
Transient unilateral hearing impairment		
Hearing aids	0.001 ^a	0–0.002
Surgical intervention	0.064 ^b	0–0.128
Transient bilateral hearing impairment		
Hearing aids	0.05 ^c	0–0.1
Surgical intervention	0.21 ^b	0.01–0.41
Permanent unilateral hearing impairment		
No intervention or ongoing monitoring; minimal hearing impairment	0.99 ^a	0.98–1.00
No intervention or ongoing monitoring; mild or moderate hearing impairment	0.49 ^a	0.39–0.59
Hearing aids; severe or profound hearing impairment	0.05 ^a	0.00–0.50
Permanent bilateral hearing impairment		
No intervention or ongoing monitoring; minimal hearing impairment	0.96 ^a	0.92–1.00
No intervention or ongoing monitoring; mild hearing impairment	0.70 ^a	0.40–1.00
Hearing aids; moderate hearing impairment	0.70 ^a	0.3–1.00
Hearing aids; severe hearing impairment	0.90 ^a	0.50–1.00
Hearing aids; profound hearing impairment	0.50 ^a	0.20–0.99

^a Expert opinion.
^b Mills *et al.* (2000).⁸¹
^c Ahmed *et al.* (2001).⁸⁰

the first, second and third screening tests is 628, 680 and 686, respectively (survey of current practice, Chapter 2). The total cost per screen test is also reported in *Table 42*. The estimated duration of the tests on which the salary costs were based was estimated from the survey of current practice (Chapter 2). If the reported duration of the tests includes the time for wider health checks, then the costs of the test will be overestimated.

The survey of current practice indicated that up to 60% of SES programmes are conducted as part of a wider health check. However, there was insufficient information to estimate the marginal costs of the school entry screening tests when conducted as part of a wider health check.

Table 43 shows the resource use and unit costs of staff and equipment used for the diagnostic

TABLE 42 Resource use and unit costs of staff and equipment for screening

Item/event	Resource use, mean (range)	Unit cost, mean (range) (£, 2004/05)
Duration of screening test	12 minutes (8–26) ^a	
School nurse	0.46 (probability) ^a	7 per test (4–14) ^{b,c}
School nurse assistant	0.13 (probability) ^a	3 per test (2–7) ^{b,c}
Nursery nurse	0.12 (probability) ^a	7 per test (4–14) ^{b,c}
Healthcare assistant	0.11 (probability) ^a	3 per test (2–7) ^{b,c}
Audiometrician	0.11 (probability) ^a	7 per test (4–14) ^{b,c}
Audiologist	0.07 (probability) ^a	9 per test (5–18) ^{b,c}
Screening audiometer	0.99	0.97 per test ^d
Calibration of audiometer	0.99	0.14 per test ^d
Screening tympanometer	0.01	0.8 per test ^d
Calibration of tympanometer	0.01	0.26 per test ^d
Paper roll (tympanometer)	0.01	0.04 per test ^d
Disposable ear tips (tympanometer)	0.01	0.14 per test ^d
Otoscope	1.00	0.97 per test ^d
Tips for ear examination	1.00	0.14 per test ^d
Total cost of screening	1.00	8.00 per test

^a The duration of the test and the probability that a particular staff category was used to conduct the school entry screen test were estimated from the survey of current practice, Chapter 2 of this report.

^b Curtis and Netten (2005).⁸³

^c NHS (2005).⁹²

^d Local accounts and expert opinion.

TABLE 43 Resource use and unit costs of staff and equipment used for diagnostic evaluation

Item/event	Resource use, mean (range)	Unit cost, mean (range) (£, 2004/05)
Duration of diagnostic evaluation	3.5 hours (3–4)	
Specialist practitioner (audiologist or clinical scientist) ^{a–c}	0.80 (0.60–1) (probability) ^a	144 per session (123–164) ^{b,c}
Audiology staff, AfC band 7	0.20 (0–0.40) (probability) ^a	164 per session (141–188) ^{b,c}
Diagnostic audiometer	0.80 (0.60–1) (probability)	7 per test ^{d,e}
Calibration of audiometer	0.80 (0.60–1) (probability)	0.13 per test ^{d,e}
Diagnostic tympanometer	0.20 (0–0.40) (probability)	15 per test ^{d,e}
Calibration of tympanometer	0.20 (0–0.40) (probability)	0.14 per test ^{d,e}
Paper roll (tympanometer)	0.20 (0–0.40) (probability)	0.8 per test ^{d,e}
Disposable ear tips (tympanometer)	0.20 (0–0.40) (probability)	0.26 per test ^{d,e}
Otoscope	1.00 (probability)	0.04 per test ^{d,e}
Tips for ear examination	1.00 (probability)	0.14 per test ^{d,e}
Total cost of diagnostic evaluation	1.00 (probability)	154.00 per evaluation

AfC, Agenda for Change.

^a Probability that a particular staff category was used to conduct the school entry screen test, estimated from the survey of current practice, Chapter 2 of this report.

^b Curtis and Netten (2005).⁸³

^c NHS (2005).⁹²

^d Local accounts and expert opinion.

^e Survey of current practice, Chapter 2 of this report.

evaluation for hearing impairment. The acquisition costs of equipment were estimated as: screening audiometer = £4600 plus VAT, screening tympanometer = £7750 plus VAT, and otoscope = £80.

Table 44 gives the estimated costs of surgical interventions and hearing aids. The life of hearing

aids was estimated as 3 years.⁸⁴ The long-term costs of hearing impairment were estimated as the cost of social care and the costs of education. The additional costs of special education and social care were estimated from national statistics.^{93,94} The additional costs for children with minimal or mild hearing impairment were estimated to be zero for both social care and education. The

TABLE 44 Resource use and unit costs of surgical interventions and hearing aids

Item/event	Resource use, mean (range)	Unit cost, mean (range) (£, 2004/05)
Hearing aids		
Hearing aids	1 set	70 per ear (60–80) ^a
Fitting hearing aids (audiologist)	4.5 hours ^a	185 per fitting (164–205) ^{b,c}
Follow-up	0.75 (0.5–1) hours per visit, two visits per year ^a	31 per visit (21–41) ^{b,c}
Repair of hearing aids	1–2 per year	15 per repair (10–20) ^{b,c}
Surgery		
Drainage of middle ear	–	612.71 ^d per operation
Myringotomy	–	612.709 ^d per operation
Autoinflation	–	612.709 ^d per operation
Adenoidectomy	–	691.31 ^d per operation
Cochlear implant	–	30,102.56 ^d per operation
^a Local accounts and expert opinion. ^b Curtis and Netten (2005). ⁸³ ^c NHS (2005). ⁹² ^d Department of Health and NHS IA (2003). ⁹⁵		

TABLE 45 Utilities by level of hearing impairment

Level of hearing impairment	Mean	Range
Minimal and mild hearing impairment	0.85 ^a	0.75–1.00 ^a
Moderate hearing impairment	0.677 ^b	0.652–0.702 ^b
Severe hearing impairment	0.616 ^b	0.469–0.634 ^b
Profound hearing impairment (96–105 dB)	0.497 ^b	0.469–0.525 ^b
Profound hearing impairment (> 105 dB)	0.353 ^b	0.327–0.379 ^b
^a Expert opinion and Dolan <i>et al.</i> (1995). ⁸⁵ ^b Barton <i>et al.</i> (2004). ⁸⁶		

additional costs for children with moderate hearing impairment were estimated to be £0 (range £0–7280) for social care and £6747 (range £0–8460) for education. The additional costs for children with severe hearing impairment were estimated to be £7280 (range £0–7280) for social care and £6747 (range £0–8460) for education. The additional costs for children with profound hearing impairment were estimated to be £7280 (range £7280–31,500) for social care and £18460 (range £6747–18,460) for education.

Table 45 shows the estimated utilities for different levels of hearing impairment. The utility for no hearing impairment was estimated as 0.99, which is the population norm for the general population under 25 years of age.⁸⁵ The utility values for minimal and mild hearing impairment were estimated from expert opinion. The utility values for moderate to profound hearing impairment were estimated from one study.⁸⁶ This study used the Health Utilities Index, a generic validated

measure, and associated utility weights to evaluate health status.

Results of the economic model

One-year time-horizon, alternative measures of effect

Tables 46–48 and Figure 14 show the results of the primary analysis for the 1-year time-horizon. Table 46 indicates that universal SES using current practice (SES-C) costs a total of £10 per child, which is more than no SES (less than £1 per child) and is associated with higher QALYs (0.983) than no SES (0.979). The ICER for SES-C is calculated as the net cost of SES-C (£10 minus £0.22) divided by the net QALYs of SES-C (0.983 minus 0.979), which is £2445 per QALY gained. This is within the threshold WTP to gain 1 QALY suggested by NICE guidelines (£30,000 per QALY gained).⁹⁶ The cost-effectiveness acceptability analysis suggests that SES-C was more cost-effective than no SES in more than 50% of simulations, if decision-makers are willing

TABLE 46 Expected cost and QALYs of screening, 1 year

	Expected cost (£)		Expected QALY	
	SES-C	No SES	SES-C	No SES
Mean	10	0.22	0.983	0.979
SD	6	0.19	0.005	0.007
2.5% percentile	6	0.01	0.973	0.964
97.5% percentile	27	0.74	0.992	0.991

TABLE 47 Probability that SES-C is cost-effective, 1 year, QALYs

WTP to gain 1 QALY (£)	Probability SES-C is cost-effective
1	0.00
2250	0.05
5250	0.70
30,000	0.90

to pay £2250 or more to gain 1 QALY (Table 47 and Figure 14). Given the data used, this indicates that there is a high probability that SES-C is likely to be more cost-effective than no SES.

The data in Table 48 show the results of the net benefit analysis. The net benefit statistics were estimated by revaluing the bootstrapped estimates of QALYs, using different ceiling ratios or WTP to gain one QALY. For each WTP threshold, the net benefit is estimated as the incremental QALY gained by SES-C multiplied by the WTP value, minus the incremental cost of SES-C. If decision-makers are prepared to pay less than £2250 to gain 1 QALY, then there is no net benefit associated with SES-C and no SES would be more cost-effective. If decision-makers are prepared to pay more than £2250 to gain 1 QALY, then SES-C is associated with an estimated net benefit of up to £112 per child (at a WTP threshold of

TABLE 48 Net benefit of SES-C, 1 year, QALYs

WTP (£)	Net benefit of SES-C compared with no SES (£)			
	Mean	SD	2.5% percentile	97.5% percentile
1	-10	6	-27	-5
2250	0	12	-32	19
5250	12	22	-40	53
30,000	112	110	-118	341

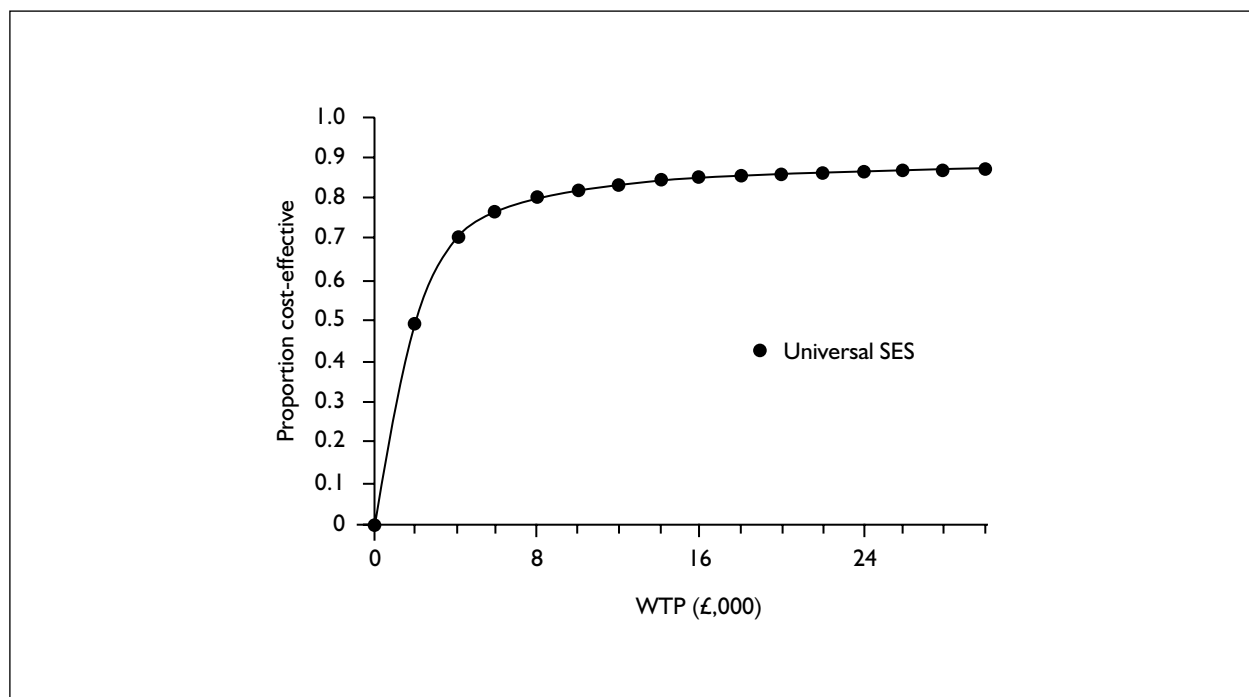


FIGURE 14 CEAC of SES-C, 1 year, QALYs

TABLE 49 Expected cost and YNHI of screening, 1 year

	Expected cost (£)		Expected YNHI	
	SES-C	No SES	SES-C	No SES
Mean	10	0.22	0.999	1.000
SD	6	0.19	0.001	0.000
2.5% percentile	6	0.01	0.997	1.000
97.5% percentile	27	0.74	1.000	1.000

£30,000 per QALY). However, the range of estimates does cross zero (i.e. the estimate for the 2.5% percentile is negative). This indicates that there may be a high level of uncertainty in the data.

Tables 49 and 50 and Figure 15 show the results of the analysis for the 1-year time-horizon, using year with no to mild hearing impairment as the measure of effect (YNHI). These data indicate that universal SES using current practice costs more, and is associated with lower YNHIs than no SES. This suggests that no SES is more cost-effective than SES-C if years gained with no to mild hearing impairment is considered the most

relevant measure of effectiveness. The cost-effectiveness acceptability analysis and net benefit analysis suggest that SES-C is not likely to be cost-effective compared with no SES. However, this analysis gives equal weight to children with no hearing impairment, minimal hearing impairment and mild hearing impairment. If minimal and mild hearing impairment adversely affect health status and health-related utility, then using YNHI as a measure of effect will underestimate the benefit of SES-C. The cost per YNHI was included as an alternative measure to assess whether the results of the economic model would differ substantially according to the method used to evaluate and value hearing levels.

The cost per true-positive case of hearing impairment detected is an alternative outcome measure that is less sensitive than the cost per YNHI. Use of this measure would give similar results to using the YNHI as an indicator of the impact of SES-C. That is, SES-C would not be cost-effective compared with no SES if the cost per true positive case were used as the outcome measure of interest.

TABLE 50 Net benefit of SES-C, 1 year, YNHI

WTP (£)	Net benefit of SES-C compared with no SES (£)			
	Mean	SD	2.5% percentile	97.5% percentile
1	-10	5	-26	-5
30,000	-28	26	-113	-10

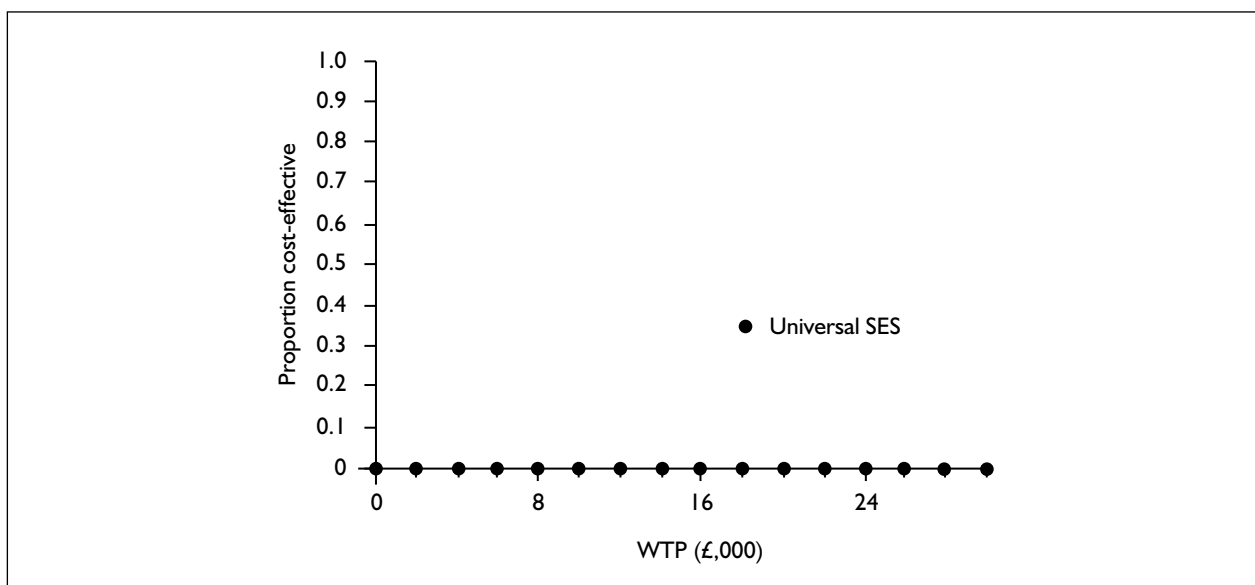


FIGURE 15 CEAC of SES-C, one year, YNHI

Six- and 11-year time-horizons, QALYs

Tables 51 and 52 and Figure 16 show the results of the primary analysis for the 6-year time-horizon. The cost-effectiveness acceptability analysis suggests that SES-C was more cost-effective than no SES in more than 99% of simulations, if decision-makers are willing to pay £2000 or more to gain 1 QALY. Given the data used, this indicates that there is a high probability that SES-C is likely to be more cost-effective than no SES.

Tables 53 and 54 and Figure 17 show the results of the primary analysis for the 11-year time-horizon. The cost-effectiveness acceptability analysis

suggests that SES-C was more cost-effective than no SES in more than 99% of simulations, if decision-makers are willing to pay £2000 or more to gain 1 QALY. Given the data used, this indicates that there is a high probability that SES-C is likely to be more cost-effective than no SES.

Comparison of SES using less accurate screen tests, 1-year time-horizon, QALYs

Tables 55 and 56 and Figure 18 show the results when less accurate screening tests (SES-T, SES-PQ and SES-SW) are compared with no SES for a 1 year time-horizon. The CEACs for SES-PQ and SES-SW suggest that they are less cost-effective than no screening. The CEAC for SES-T suggests that it is more cost-effective than no SES in 50–70% per cent of simulations, if decision-makers are willing to pay £5000 or more to gain 1 QALY.

Table 57 shows the net benefit of SES-C compared with less accurate screening tests (the cost and QALY information is given in Tables 46 and 57). Figure 19 shows the CEAC when less accurate screening tests (SES-T, SES-PQ and SES-SW) are compared with SES-C for a 1-year time-horizon.

TABLE 51 Expected cost and QALYs of screening, 6 years

	Expected cost (£)		Expected QALY	
	SES-C	No SES	SES-C	No SES
Mean	25	2	5.37	5.27
SD	10	2	0.04	0.07
2.5% percentile	11	0.36	5.28	5.13
97.5% percentile	49	6	5.44	5.39

TABLE 52 Net benefit of SES-C, 6 years, QALYs

WTP (£)	Net benefit of SES-C compared with no SES (£)			
	Mean	SD	2.5% percentile	97.5% percentile
1	-23	25	-46	-9
30,000	2853	1376	721	6034

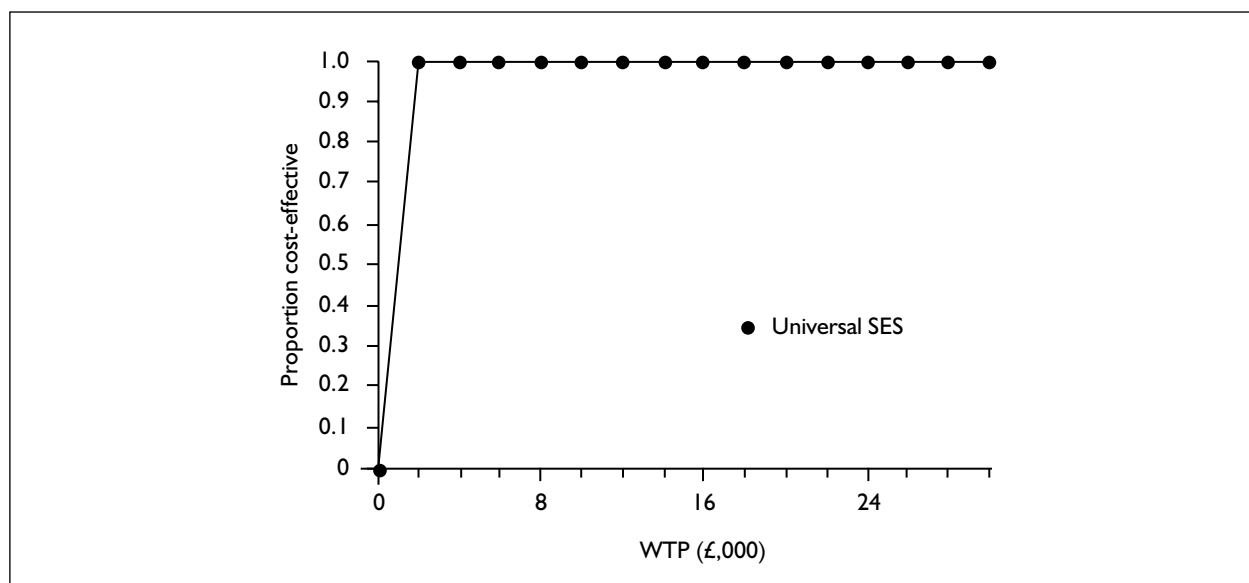


FIGURE 16 CEAC of SES-C, 6 years, QALYs

The CEACs for SES-T, SES-PQ and SES-SW suggest that they are less cost-effective than SES-C.

Comparison of SES-C with no SES using more accurate screen tests, 1-year time-horizon, QALYs
 SES using pure tone sweep only (SES-PTS) was more accurate than SES-C. The expected costs of SES-PTS were similar to those of SES-C (£10.39 SES-PTS and £9.90 SES-C) and the expected QALYs were the same at 0.983. Table 58 shows the net benefit of SES-PTS compared with SES-C. The CEAC in Figure 20 suggests that SES-C is cost-effective in 60% of simulations compared with SES-PTS.

TABLE 53 Expected cost and QALYs of screening, 11 years

	Expected cost (£)		Expected QALY	
	SES-C	No SES	SES-C	No SES
Mean	30	3	9.07	8.91
SD	13	2	0.07	0.11
2.5% percentile	12	0.45	8.93	8.68
97.5% percentile	64	9	9.19	9.10

Comparison of SES-C with low-accuracy targeted SES, 1-year time-horizon, QALYs

Tables 59 and 60 and Figure 21 show the results when targeted SES is compared with SES-C for a 1-year time-horizon. The targeted SES assumes that only children identified as being at risk of hearing impairment are screened (10%). For this analysis, the probability that children are accurately identified was set equal to the probability that parental questionnaires are an accurate screen, which was relatively low. The cost-effectiveness acceptability analysis suggests that in this case SES-C was more cost-effective than targeted SES in around 75–90% of simulations, if decision-makers are willing to pay £5000 or more to gain 1 QALY. Given the data used, this indicates that there is a high probability that SES-C is likely to be more cost-effective than targeted SES, if the accuracy of identifying children at risk is low.

Comparison of SES-C with high-accuracy targeted SES, 1-year time-horizon, QALYs

Table 61 and Figure 22 show the results when SES-C is compared with targeted SES for a 1-year time-horizon. The targeted SES assumes that only children identified as being at risk of hearing

TABLE 54 Net benefit of SES-C, 11 years, QALYs

WTP (£)	Net benefit of SES-C compared with no SES (£)			
	Mean	SD	2.5% percentile	97.5% percentile
1	-27	13	-59	-10
30,000	4867	2327	1270	10,242

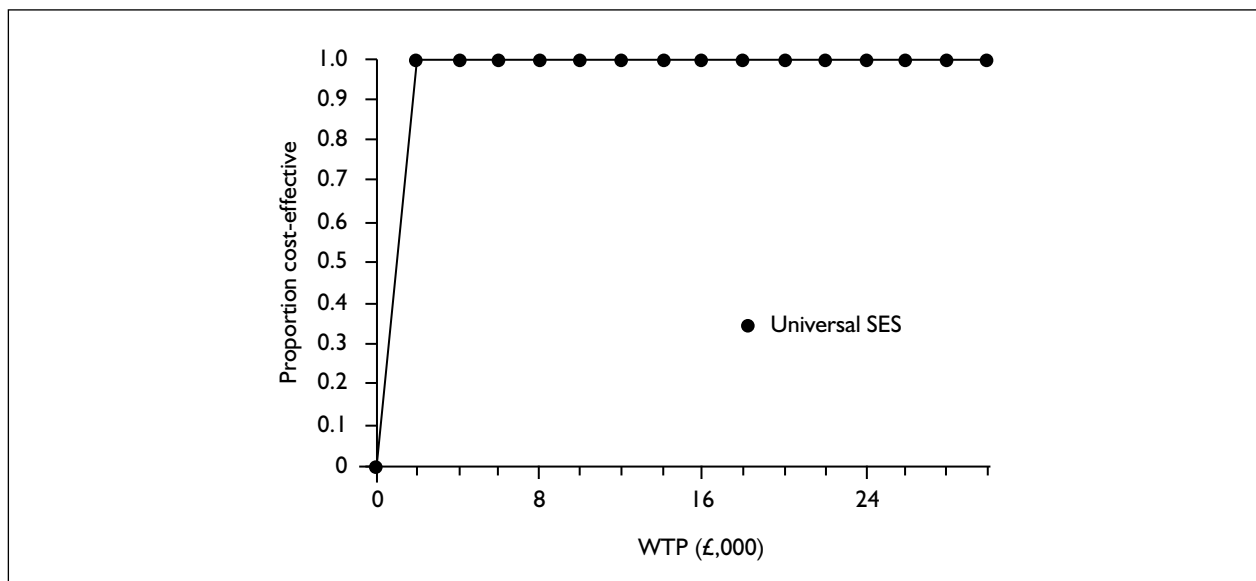


FIGURE 17 CEAC of SES-C, 11 years, QALYs

TABLE 55 Expected cost and QALYs of less effective screening, 1 year

	Expected cost (£)			Expected QALY		
	SES-T	SES-PQ	SES-SW	SES-T	SES-PQ	SES-SW
Mean	10	23	30	0.975	0.977	0.964
SD	2	12	24	0.019	0.006	0.022
2.5% percentile	6	8	9	0.912	0.964	0.907
97.5% percentile	15	44	84	0.991	0.989	0.989

TABLE 56 Net benefit of less effective screening versus no screening, 1 year, QALYs

WTP (£)	Net benefit of screening compared with no SES (£)			
	Mean	SD	2.5% percentile	97.5% percentile
SES-T				
	-10	2	-15	-6
30,000	-121	568	-1989	286
SES-PQ				
	-23	12	-44	-8
30,000	-81	145	-355	207
SES-SW				
	-30	24	-83	-9
30,000	-464	645	-2033	236

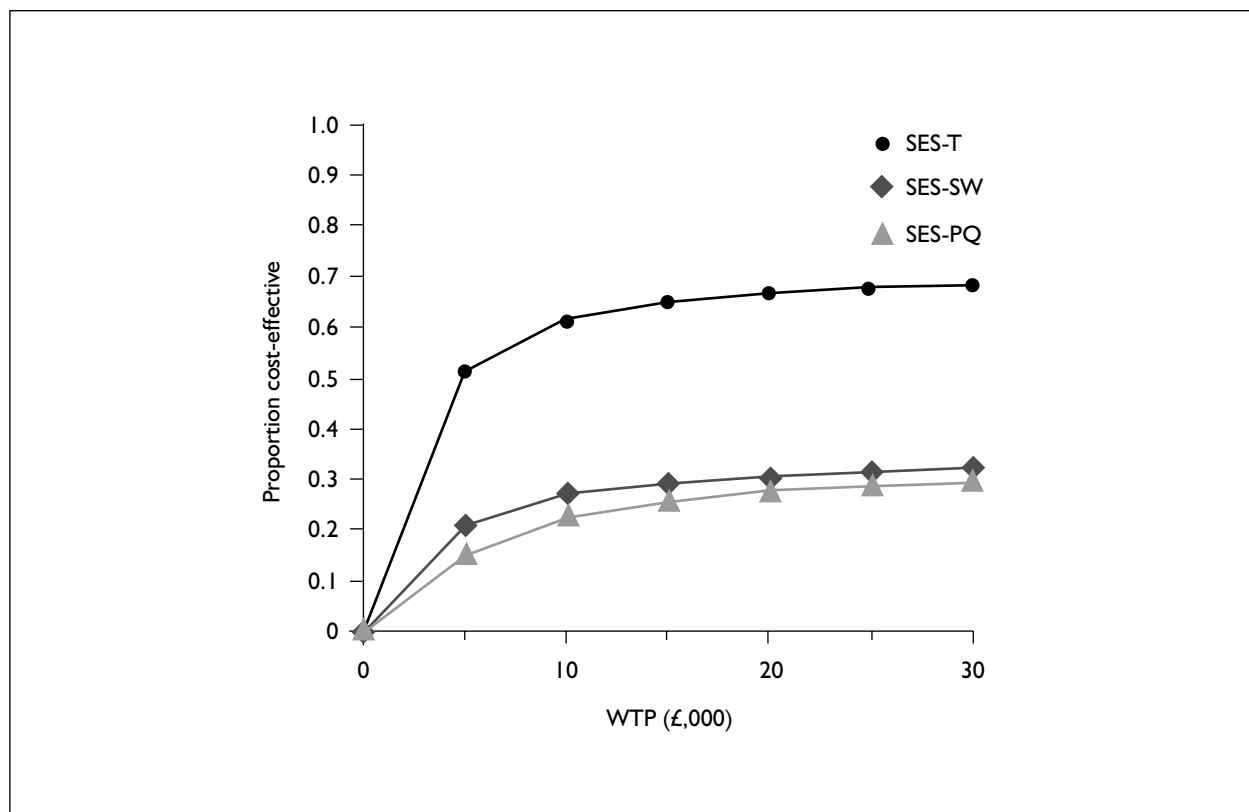


FIGURE 18 CEAC of less accurate SES programmes versus no SES, 1 year, QALYs

TABLE 57 Net benefit of less effective screening versus SES-C, 1 year, QALYs

WTP (£)	Net benefit of less accurate screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
SES-T				
	0	6	-6	16
30,000	-244	566	-2132	131
SES-PQ				
	-13	13	-35	13
30,000	-193	133	-449	64
SES-SW				
	-20	24	-74	12
30,000	-576	649	-2203	100

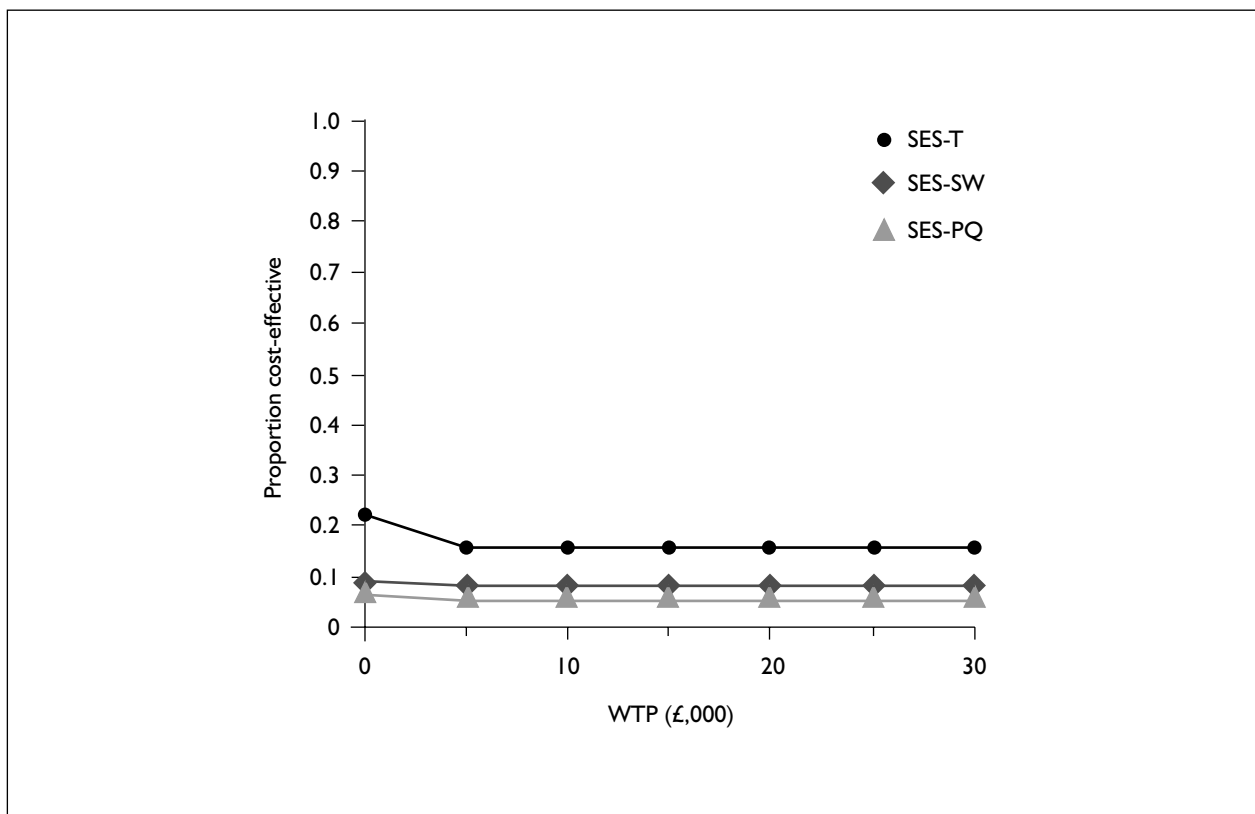


FIGURE 19 CEAC of SES-C versus less accurate tests, 1 year, QALYs

TABLE 58 Net benefit of more accurate screening versus SES-C, 1 year, QALYs

WTP (£)	Net benefit of more accurate screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
SES-PTS				
	0	8	-19	17
30,000	-4	85	-220	194

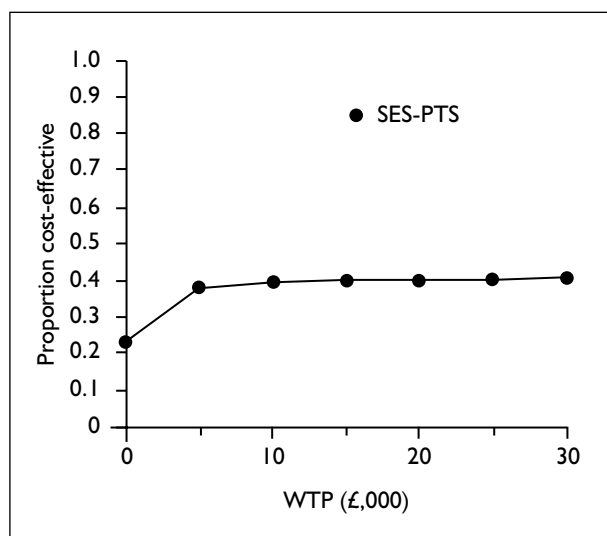


FIGURE 20 CEAC of SES-PTS versus SES-C, 1 year, QALYs

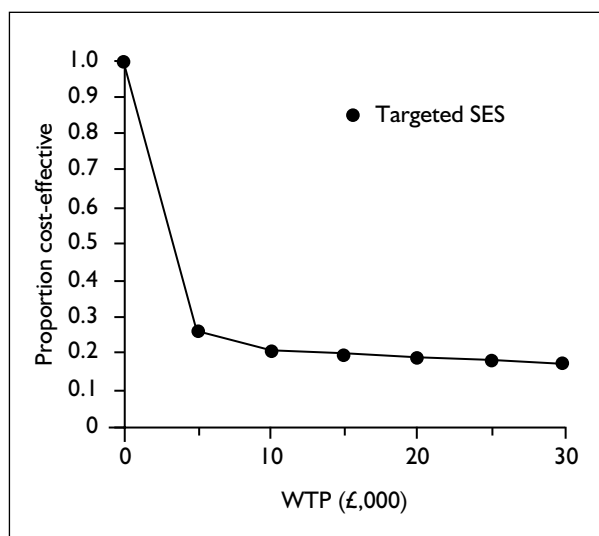


FIGURE 21 CEAC of low accuracy targeted SES versus SES-C, 1 year, QALYs

TABLE 59 Expected cost and QALYs of targeted screening versus universal SES-C, 1 year

	Expected cost (£)		Expected QALY	
	Low-accuracy targeted screening	High-accuracy targeted screening	Low-accuracy targeted screening	High-accuracy targeted screening
Mean	3	1	0.980	0.988
SD	3	1	0.006	0.004
2.5% percentile	1	0	0.967	0.981
97.5% percentile	12	2	0.992	0.996

TABLE 60 Net benefit of low accuracy targeted screening versus SES-C, 1 year, QALYs

WTP (£)	Net benefit of less accurate targeted screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
Low-accuracy targeted screening				
1	6	4	-3	20
30,000	-74	78	-251	76

TABLE 61 Net benefit of high accuracy targeted screening versus SES-C, 1 year, QALYs

WTP (£)	Net benefit of more accurate targeted screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
High-accuracy targeted screening				
1	9	5	5	25
30,000	173	77	65	364

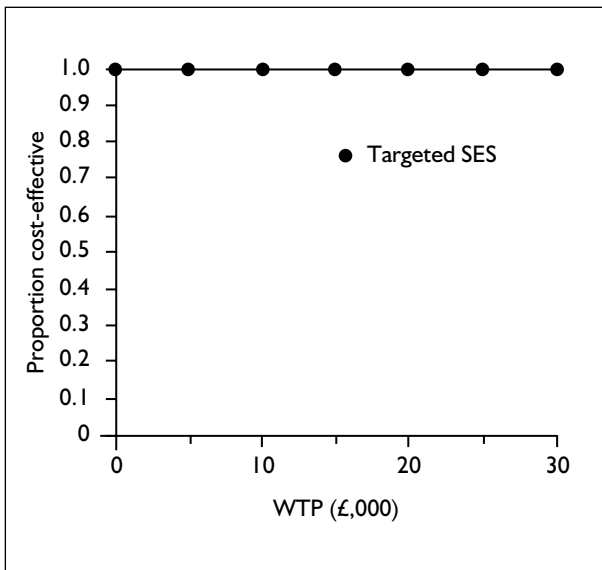


FIGURE 22 CEAC of SES-C versus high accuracy targeted SES, 1 year, QALYs

impairment are screened (10%). For this analysis, the probability that children are accurately identified was set equal to 90%. The cost-effectiveness acceptability analysis suggests that in this case targeted SES was more cost-effective than SES-C in around 80–90% of simulations, if decision-makers are willing to pay £1 or more to gain 1 QALY. Given the data used, this indicates that there is a high probability that SES-C is likely to be less cost-effective than targeted SES, if the process to identify at-risk children currently is at approximately 90%.

Comparison of SES-C with no SES, low prevalence of hearing impairment in target population, 1-year time-horizon, QALYs

Tables 62 and 63 and Figure 23 show the results when the prevalence of unidentified permanent hearing impairment is assumed to be lower, as would be the case if a proportion of cases of hearing impairment were identified via the NHSP. SES-C is compared with no SES for a 1-year time-horizon. In this analysis, the prevalence of unidentified permanent hearing impairment is reduced from 3.5 in 1000 to 0.34 in 1000. This was chosen to reflect the potential impact of the introduction of the NHSP (Waltham Forest study, Chapter 3). In addition, the probability that someone with hearing impairment has any permanent hearing impairment (including minimal and mild hearing impairment) is reduced from 0.04 to 0.01. The cost-effectiveness acceptability analysis suggests that in this case SES-C was still more cost-effective than no SES in over 50% of simulations, if decision-makers are willing to pay £5000 or more to gain 1 QALY. Given the data used, this indicates that there is a high probability that SES-C is likely to be more cost-effective than no SES, when the prevalence of unidentified permanent hearing impairment is reduced to 0.34 in 1000 and the proportion of people with hearing impairment who have any permanent hearing impairment is reduced to 1%.

Tables 64 and 65 and Figure 24 show the results if the NHSP or other previous screening programmes mean that the prevalence of any

TABLE 62 Expected cost and QALYs of SES-C versus no screening, low prevalence of hearing impairment, 1 year

	Expected cost (£)		Expected QALY	
	SES-C	No screening	SES-C	No screening
Mean	9	0.11	0.985	0.983
SD	5	0.09	0.005	0.005
2.5% percentile	5	0.007	0.976	0.973
97.5% percentile	22	0.33	0.933	0.993

TABLE 63 Net benefit of SES-C versus no screening, low prevalence of hearing impairment, 1 year, QALYs

WTP (£)	Net benefit of more accurate targeted screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
Low prevalence of hearing impairment				
1	-9	5	-22	-5
30,000	47	78	-154	176

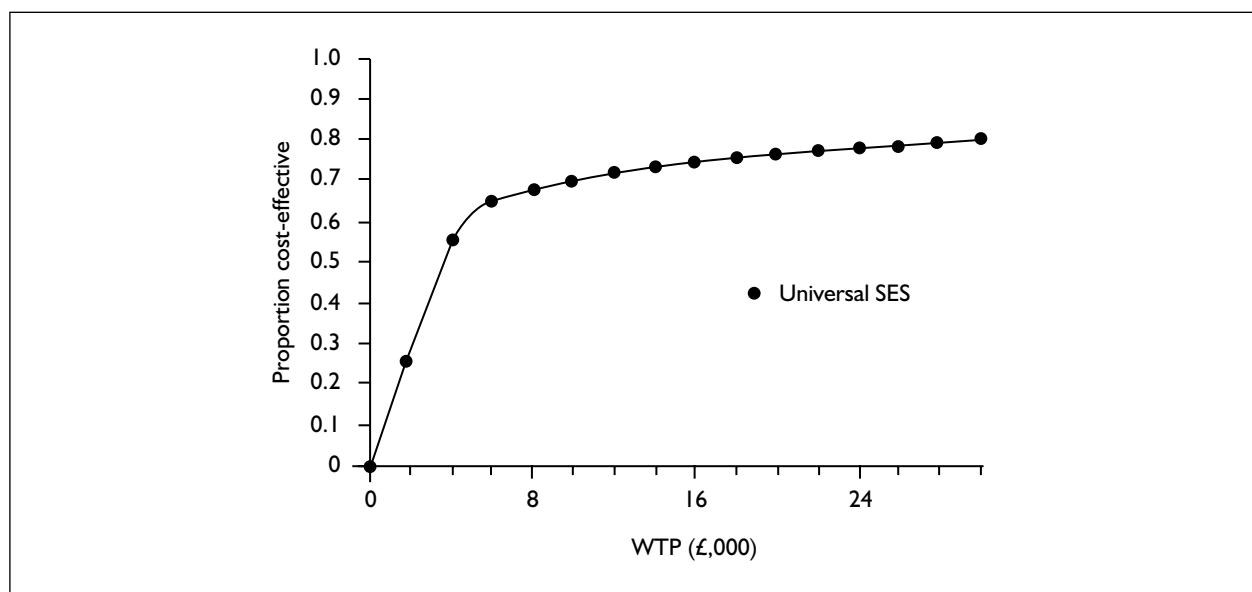


FIGURE 23 CEAC of SES-C versus no SES, lower prevalence of moderate or worse permanent hearing impairment, 1 year, QALYs

hearing impairment still to be found is halved, and the proportion of people with hearing impairment who have permanent hearing impairment is reduced to 1%. In this case, SES-C is still cost-effective compared with no SES, but the probability that it is cost-effective is reduced to 50% in 60% of simulations. The amount that decision-makers would need to be willing to pay to gain 1 QALY also increases from £2000 to over £6000. However, this is still less than the value implied by previous healthcare decisions (£30,000 per QALY gained⁹⁶).

Comparison of SES-C with no SES, 1-year time-horizon, true cases of hearing impairment detected

Tables 66 and 67 and Figures 25 and 26 present the results of comparing SES-C with no screening, using the limited outcome measure of number of true cases of hearing impairment detected by screening. When true cases of any hearing impairment are used as the effect measure, the data support the results of the primary analysis, that SES-C is likely to be cost-effective. However, if the appropriate effect

TABLE 64 Expected cost and QALYs of SES-C versus no screening, prevalence of hearing impairment halved, 1 year

	Expected cost (£)		Expected QALY	
	SES-C	No screening	SES-C	No screening
Mean	9	0.08	0.986	0.985
SD	5	0.07	0.005	0.005
2.5% percentile	5	0.05	0.977	0.975
97.5% percentile	23	0.27	0.994	0.994

TABLE 65 Net benefit of SES-C versus no screening, prevalence of hearing impairment halved, 1 year, QALYs

WTP (£)	Net benefit of more accurate targeted screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
Low prevalence of hearing impairment				
1	-8	4	-23	-5
30,000	16	73	-179	138

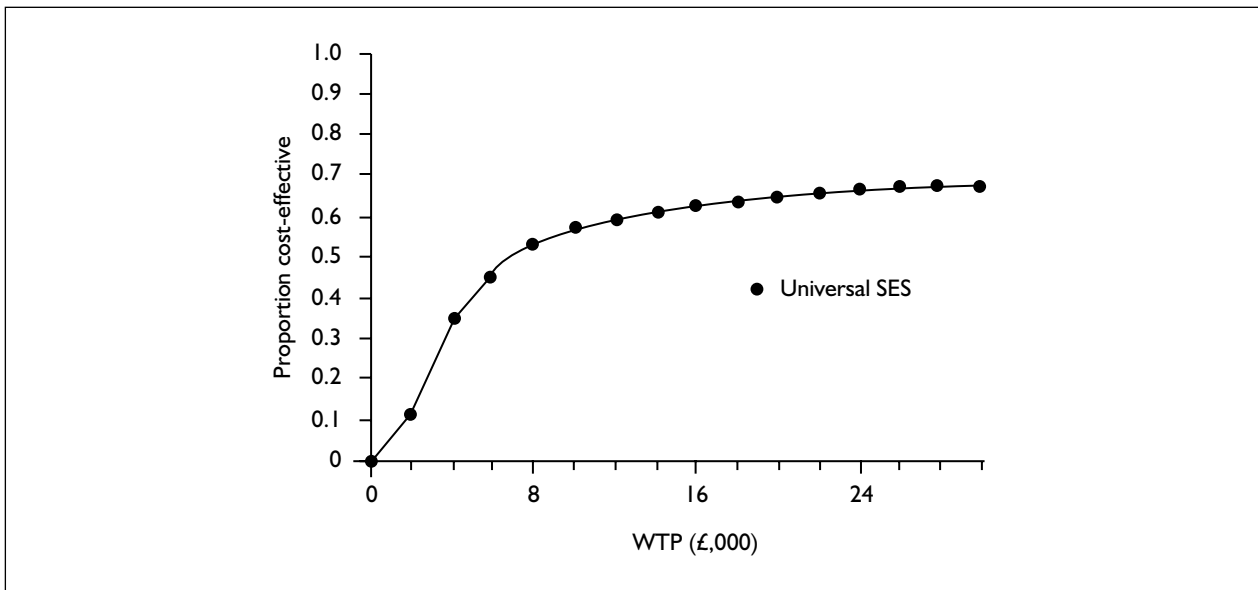


FIGURE 24 CEAC of SES-C versus no SES, prevalence of still-to-be-found hearing impairment is halved, 1 year, QALYs

TABLE 66 Expected cost and true cases of any hearing impairment detected of SES-C versus no screening, 1 year

	Expected true cases of any hearing impairment		Expected true cases of permanent hearing impairment	
	SES-C	No screening	SES-C	No screening
Mean	0.018	0.001	0.00038	0.00002
SD	0.018	0.000	0.00053	0.00002
2.5% percentile	0.007	0.000	0.00003	0.00000
97.5% percentile	0.07	0.002	0.00216	0.00009

TABLE 67 Net benefit of SES-C versus no screening, true cases of any hearing impairment detected, 1 year, QALYs

WTP (£)	Net benefit of more accurate targeted screening compared with SES-C (£)			
	Mean	SD	2.5% percentile	97.5% percentile
True cases of any hearing impairment				
1	-9	5	-24	-5
30,000	502	524	195	2058
True cases of permanent hearing impairment				
1	-9	5	24	5
30,000	2	12	-7	39

measure is thought to be true cases of permanent hearing impairment detected by screening, then SES-C is less likely to be cost-effective, with less than 50% of simulations showing SES-C as cost-effective.

Table 68 shows the total cost and number of true cases of any hearing impairment detected for a 1-year cohort of children entering school and eligible for screening in England. This is based

on the population of 1.129 million children aged between 4 and 6 years in 2005 (<http://www.statistics.gov.uk/statbase/Expodata/Spreadsheets/D9390.xls>). The data indicate that the universal screening programme represented by SES-C is likely to detect an additional 1497 true cases of any hearing impairment in 1 year compared with no screening, including an additional 32 cases of permanent hearing impairment.

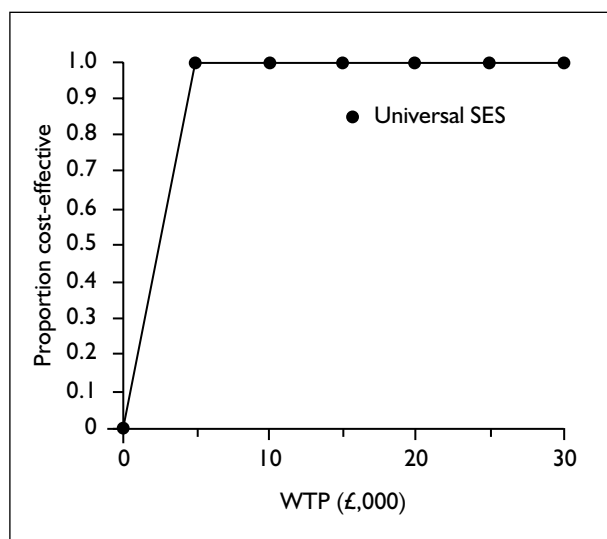


FIGURE 25 CEAC of SES-C versus no SES, 1 year, true cases of any hearing impairment detected

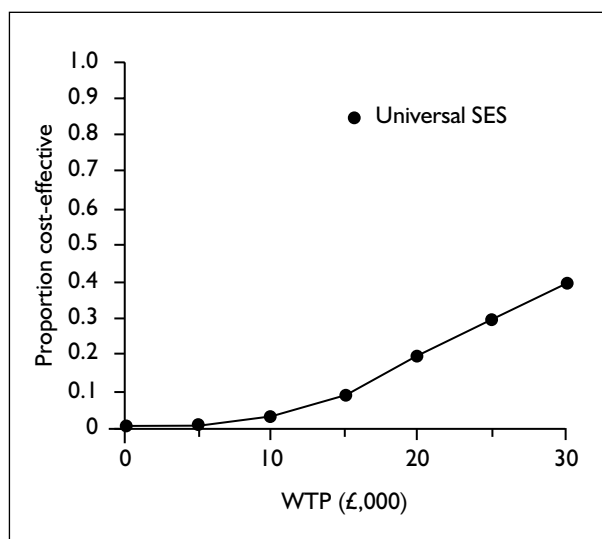


FIGURE 26 CEAC of SES-C versus no SES, 1 year, true cases of permanent hearing impairment detected

Summary

The literature was systematically searched to identify published economic evaluations that assessed the cost-effectiveness of SES. No full economic evaluations were found. Two partial economic evaluations were found. Overall, the quality of these papers was judged to be low according to the quality assessment criteria used. In particular, there were insufficient data available to judge the validity and robustness of the economic and clinical data used in the analyses, or the relevance of the data and results to the UK setting. Cost studies were also reviewed to extract any relevant resource use and unit cost data for the economic model. However, as with the economic evaluations, the quality and applicability of the data (to the UK setting) from these studies were limited.

A decision-analytic model was developed to assess the costs, effectiveness and net benefit of SES compared with no SES and SES using alternative tests within the screen. The primary source of data about the accuracy of the screening tests for the

economic model was the data included in the systematic review reported in Chapter 4. This was supplemented by data on the prevalence and distribution of hearing impairment, the probability of a child being screened, diagnosed and treated, the costs of screening, diagnosis and treatment, and the outcomes of screening from published literature, the survey of current practice, the observational study conducted in Waltham Forest, national statistics and databases, local accounts and expert opinion.

For the 1-year time-horizon used in the primary analysis, SES-C was associated with higher costs and slightly higher QALYs compared with no SES and other SES programmes. The range of expected costs, QALYs and net benefits was broad, with the 2.5th to 97.5th percentiles of differences in expected costs and outcomes crossing zero. CEACs and measures of net benefit provide a means of assessing the robustness of differences in expected costs and outcomes, when combined into ICERs. These analyses allow for the fact that there may be a relationship between resource use and costs and outcomes, so that, for example, higher

TABLE 68 Total costs and true cases detected for a 1-year cohort of children eligible for screening

	Total costs (£)	Total true cases detected	
		Any hearing impairment	Permanent hearing impairment
SES-C	880,854	1,585	33
No screening	19,379	88	2
SES-C minus no screening	861,475	1,497	32

resource use and therefore costs may be associated with improved outcomes. Overall, the primary analysis indicated that SES-C was cost-effective compared with all the other SES and no SES programmes. A number of secondary analyses was used to explore subgroups of the data and test assumptions used in the model. However, the use of subsets of the data, with relatively few studies to

combine and small sample sizes, means that these analyses can be exploratory only. The secondary analyses supported the result that SES-C is cost-effective compared with no SES and alternative SES programmes. The most cost-effective method of implementing SES is using SES-PTS. This is more cost-effective than no SES, SES-C and less accurate tests used in SES.

Chapter 6

Summary and conclusions

Introduction: strengths and weaknesses of the study

This study is the first comprehensive attempt in the UK to address issues surrounding screening for hearing impairment at school entry (around age 4–6 years). Previous studies have reported surveys of practice, audits of screen performance and test accuracy for specific conditions such as OME. This study aims to bring three strands of work together: a survey of practice across the UK (Chapter 2), a systematic review of the accuracy of alternative tests and the effectiveness of interventions (Chapter 4), and modelling costs and cost-effectiveness (Chapter 5). In addition, the authors were able to access some primary data from the team in Waltham Forest (PW) that address changes that are likely to occur owing to the introduction of newborn hearing screening (Chapter 3).

Survey of current practice

For the survey of current practice a postal questionnaire was developed for service leads for the SES across the UK. Robust results from postal survey methodology rely on both identifying the appropriate population and achieving a sufficiently high response rate to be able to generalise.

Identification of the population to which the questionnaire was to be sent raised several procedural difficulties. The aim was to survey the total population, that is, all staff responsible, as service leads, for the SES programme in the UK. The service is not provided in the same way throughout the UK, probably for historical reasons, and hence identifying the leads involved a series of different approaches (listed in Chapter 2), including advertisements in professional newsletters and cold-calling NHS trusts. This last method highlighted the difficulty, encountered often, of identifying anyone within the trust who knew about the screen and/or knew who had responsibility for it. This raises issues of managerial responsibility for the screen, and consequent local and national accountability for screen performance. After considerable effort and time a contact was secured for every trust in England, every board in Wales and Scotland and

for every LSHCG in Northern Ireland. The 244 service leads identified for the SES, many of whom covered more than one PCT, comprise a much higher number of SES leads than in previous surveys,⁶ and are likely to represent almost total coverage, although it is possible that a very few services run entirely by educational services would be missed. Research governance approval was sought from 124 R&D directorates covering 304 PCTs in England, Scotland and Wales, but in Northern Ireland it was only possible to approach two R&D offices from 15 LSHCGs. R&D procedural difficulties meant that 229 out of 244 services could be sent a questionnaire.

There was an extremely high return rate of questionnaires for this sort of study of just over 85%. Bearing in mind the data and comments from respondents, the high return rate seemed to reflect the willingness of people to take the time to tell the researchers about their service and their views, in the knowledge that the information would be used to contribute to the development of a service which they felt was important but in need of some guidance. The response rate may also be due to the fact that respondents were not asked to allocate time to report data that were not easily available. This means that some of the information provided lacks depth and that the data on yield and screen performance are based on a relatively small proportion of all services; even the data from these services may lack accuracy. Nevertheless, the survey of current practice is the most complete carried out in the UK, with wide geographical coverage and a high response rate.

Waltham Forest observational studies

The authors were fortunate to be able to access good-quality data from the Waltham Forest services. In UK terms this service is unique since it offers robust data from three sizeable cohorts, all of which had the SES, but following a universal newborn screen for one, a targeted newborn screen for the second and traditional infant screens for the third. This is a rare set of comparative cohorts, and although they do not conjointly comprise a single longitudinal study, they allow some valuable comparisons concerning the possible future impact of the now national

implementation of the NHSP on the SES, as well as some other secular data. However, there are features of the Waltham Forest populations (e.g. ethnicity, mobility) that undermine highly detailed or quantitative generalisation from this local evidence base to the national cohort covered (since March 2006) by the NHSP, once the latter has reached school entry age (in around 2010).

Nevertheless, the epidemiological characteristics of those children with a permanent hearing impairment in the Waltham Forest cohorts was not significantly different from that reported from larger and national studies undertaken in the UK, and it is probable that the population of hearing-impaired children reported is reasonably typical of that present elsewhere in the UK. The newborn screen protocol used in the Waltham Forest cohorts differed from that implemented in the NHSP; nevertheless, the yield of moderate or worse permanent bilateral hearing impairment obtained from the NHSP (1.0 in 1000, 95% CI 0.78 to 1.22)⁹ was extremely similar to the yield of 1.03 in 1000 (95% CI 0.66 to 1.40) achieved in the Waltham Forest studies. The yield of children with a permanent unilateral hearing impairment in the Waltham Forest studies was 0.55 (95% CI 0.28 to 0.82) and this was similar to the 0.64 in 1000 (95% CI 0.37 to 0.91) reported from the NHSP. These similarities suggest that generalisation is justified.

The Waltham Forest studies were ascertainment studies, that is, they depended on collecting data from all known cases of children with permanent hearing impairment in the three cohorts. Thus, cases not yet identified, for whatever reason, would not be included, and the strength of the data is crucially dependent on both the quality of the service at identifying cases and the robustness of the ascertainment procedures. Furthermore, the ability of services accurately to identify childhood hearing impairment reduces with lower levels of impairment, so in any study of this type, despite the greater number of milder cases there remains some uncertainty about their ascertainment rate.

Systematic review of accuracy and effectiveness of screening

The principal strength of the systematic review was its comprehensiveness. Compared with three previous similar reviews in this area more studies were identified, in part because these previous reviews focused more narrowly, for example on preschool screening for OME or on a particular screening test (whisper voice test).

The main limitation of the review was a lack of good-quality evidence on the effectiveness of SES on long-term outcomes, including educational, language and social outcomes. The authors acknowledge the challenge of demonstrating effectiveness at two stages, both case finding and intervention. In addition, although several studies have assessed the accuracy of screening tests, their quality and the quality of reporting were unacceptably variable. Two particular problems were the inconsistent reference standard applied and variability in the criterion definition for hearing impairment that was used. Although full diagnostic PTA was used as the reference test in many studies, this was not the case in all. A reference standard test is a key element of any study of diagnostic accuracy. A high or low level of accuracy derived from a comparison of a given test against a range of reference standards, none of which is stated to be a gold standard, is difficult to interpret. Furthermore, studies often failed to specify clearly the criterion threshold that defined hearing impairment, and different studies applied different hearing thresholds for their case definition. Without a consistent case definition it is difficult not only to interpret the accuracy of results of a given study, but also to apply these results to real-world clinical practice.

Cost-effectiveness

Overall, the evidence base to support the economic model was weak. As noted in Chapter 4, the robustness of the available evidence about the accuracy of screening tests was undermined by the variable quality of the studies investigating test accuracy. In addition, no evidence about the short- or long-term effectiveness (impact on disability and quality of life) or cost-effectiveness of SES was found. The data used to estimate other probabilities, costs and outcomes were synthesised from a variety of sources, including surveys of clinical practice and expert opinion. These may affect the robustness of the conclusions. Although the variables that used data from surveys or expert opinion were assigned wide distributions wherever possible, this also increases the uncertainty in the model parameters and reduces the likelihood or probability that an intervention is cost-effective.

The economic model was static in nature and based on a short time-frame of 1 year for the primary analysis. The time-horizon was extended to 11 years in secondary analyses. The static structure of the model was based on the assumption that the values of the variables included would not change significantly over time. There is no evidence to suggest that this is an

unreasonable assumption for the 1-year time-frame considered in the primary analysis. However, if hearing impairment identified by the school screen is progressive, or the impact on quality of life and health status of hearing impairment identified by the SES changes significantly over time, then the results of the economic model may not apply.

High and low estimates of utility and long-term costs were included in the primary and secondary analyses. The results of the cost acceptability and net benefit analyses reflected the high uncertainty about the input parameters, but broadly supported the main conclusions of the economic analyses. The structure of the model was developed from the reviews of clinical and economic evidence and discussion with experts in audiology.

The primary and secondary analyses used QALYs as the outcome measure to estimate ICERs, net benefit and CEACs. QALYs take into account differences in potential life expectancy and the impact of adverse events on overall health-related quality of life; they tend to be weighted by mortality and produce only small differences among adverse events that have a short or relatively low impact on quality of life. This may bias the analysis if one or more interventions are associated with high rates of adverse events that individually have a relatively low impact on health, but cumulatively could have a significant impact on health and health-related quality of life. Overall, the estimates of expected QALYs differentiated between different SES programmes and levels of hearing impairment. These factors would suggest that the QALY is a reasonable measure for the economic analysis (and is consistent with the approach used for reports to NICE).

Summary of findings

Survey of current practice

The SES is usually performed in the first year of primary education, in school, and usually (72%) with prior written information to parents and guardians. The survey indicated that there is wide variation in the implementation of the screen throughout the UK. This variation applies to the population covered, with 51% of respondent services not screening children entering private education and 72% not screening home-educated children; the physical location and conditions under which the screen is implemented, with little

evidence of commitment by schools to offer suitable locations, and little commitment by management to provide training and replacement equipment; test methodology, with different numbers and types of tests and retests (17% referring after the first test, 72% after a second, 10% after a third); the time of repeat tests (same day to 12 weeks); the criteria determining which children to refer, which varied from 20 dB HL across up to seven frequencies to 30 dB HL at only three frequencies (the dB scale is a logarithmic ratio scale, with 0 dB HL being the average normal hearing threshold and the difference between 20 and 30 dB HL representing a ten-fold increase in power and more than a doubling of subjective loudness); the personnel and the equipment involved; and the ability to collect and then retrieve data. There is little or non-existent robust audit at local (and therefore national) level, absent or inadequate data management systems, and a lack of explicit procedures for quality assurance. The one area of consistency concerned the pure tone sweep test, which was used by 97% of responding services as the first test in the screen.

The fact of existing protocol variation between services could provide for an evaluation of what might be the most appropriate, successful and efficient implementation of the screening programme. However, good data on yield and screen performance are necessary for such an evaluation; although nearly 70% of services claimed to have data management systems in place, only 50% of those said that they could easily obtain data reports. Coverage and referral data were available for approximately one-third of services ($n = 55$), but fewer than 20 services could provide any robust data on the numbers of children identified as hearing impaired. The uptake data from those that could provide them indicated a median uptake rate for those offered the screen of over 90%. At least half the responding services screen those children already known to some part of the service (but often not to the SES service) to have a hearing impairment, again indicative of poor information sharing.

Despite these marked difficulties and the lack of robust audit there was a very high response rate to the survey questionnaire, indicating a high level of interest; there were clear indications from comments offered that the majority of service leads regard the screen as useful and would prefer it to continue, even though it was recognised that the value of the screen may reduce with the advent of universal newborn hearing screening. A small

proportion of respondents (12.2%) have abandoned the screen, and a few noted that they were awaiting national guidance on its future. A significant number of respondents stated that they would welcome such national guidance. Other service leads would welcome guidelines on the value of a selective (targeted) screen and on the population for whom it would be appropriate. Most concerns about the continued relevance of the screen as a universal screen focused on the impact of the introduction of universal newborn hearing screening, the inadequate resources (time, personnel and facilities) available to implement the screen, and the inadequacy of systems and technological support for data management and retrieval. Support for the continuing value of the screen focused on its ability to identify children who would otherwise not be identified, either because they had been missed by previous screens or surveillance for whatever reason, or because they had entered the system having had no previous screens.

Waltham Forest observational studies

The evidence from the cohort comparisons in Waltham Forest reported here for the first time in a single source suggests strongly that there is a material effect of the introduction of newborn hearing screening on the SES, in addition to other secular changes that have occurred in recent years. Of the latter, the most important are probably the immunisation programmes that seem to have been accompanied by a significant reduction in the proportion of children with severe and profound unilateral hearing impairment. With regard to the possible changing pattern of routes to identification of permanent childhood hearing impairment, before the introduction of newborn screening the yield from the IDT, intermediate screens and parental/professional concern throughout infancy and up to school entry was around 73% of the yield of all PCHI cases resident and currently known to the service, while the final screen, the SES, accounted for the remainder at a rate of 1.11 in 1000, of which 0.63 in 1000 were unilateral impairments. The evidence suggests that since the introduction of universal newborn screening, over 90% of the cases known to services now have been identified via newborn screening and parental/professional concern in infancy and up to school entry, with only 0.34 in 1000 identified by the SES (of which only 0.07 in 1000 were unilateral).

Thus, in Waltham Forest, newborn screening has reduced the yield of the SES for permanent hearing impairment. However, postnewborn

routes to identification remain important, in large part because of late-onset and acquired cases, those who had 'moved in', and those with a congenital impairment that had not been picked up by the newborn screening programme. The prevalence of mild and greater bilateral and unilateral hearing impairment at school age was 3.47 in 1000, similar to findings from other studies. Parental and professional concern remained a steady source of identification (1.31 in 1000) postnatally, but still, at school entry 16% of moderate and greater bilateral, 18% of mild bilateral and 17% of unilateral permanent hearing impairments remained to be identified. The evidence for long-term effects of moderate or greater congenital bilateral hearing impairment is well documented,⁵ and there are known and demonstrably beneficial interventions based around the early provision of hearing aids. Long-standing beliefs in the necessity of intervention have made it impractical to conduct controlled trials on benefits of intervention for moderate hearing impairment which is first identified at school entry, and the present research has furthermore failed to identify statistically controlled studies with age of identification as a major factor that enable some conclusions to be drawn in the way that it is possible in the earlier years. Nevertheless, it is reasonable to assume that the effects, if untreated, would be marked, especially for significant but unidentified impairment at the transition to formal schooling. These arguments suggest that some sort of systematic approach to identification of moderate or greater permanent hearing impairment at, or approaching school entry age is required. The evidence on the effects of mild bilateral and unilateral hearing impairment on long-term outcomes is largely absent, and the same argument is therefore difficult to make on the basis of available evidence. However, from what is known of mild hearing impairment and the acoustics of classrooms, it would be reasonable to extend the argument to include the need to find and manage these not previously known mild and unilateral hearing impairments as well.

Systematic review of accuracy and effectiveness of screening

There was only level III evidence for the effectiveness of preschool hearing screening, from a single, poor-quality, observational comparative study. Furthermore, this single study was inconclusive in whether preschool screening was more effective than no screening in detecting hearing impairment. No studies were identified that have assessed the long-term impact of

preschool hearing screening on educational, language and social outcomes.

Several studies have assessed the accuracy of different hearing screening tests in preschool children. Given the unacceptable variability in methodological quality and reporting of these studies, the lack of clarity over the cases of hearing impairment detected (e.g. transient versus permanent hearing impairment), the variation in reference test and threshold level for hearing deficit, and the range of settings in which these tests were applied, it is difficult to interpret and compare their results.

Nevertheless, accepting these caveats and selecting the subset of studies using PTA as the reference test, the findings suggest that pure tone sweep audiometry has high sensitivity and specificity for full PTA and therefore appears to be a suitable test for screening. Other possible tests, about which more and better evidence is required, are spoken word tests and TEOAEs. For OME, tympanometry and reflectometry have variable reported sensitivity and specificity as screening tests (although note that tympanometry is a well-established and valuable diagnostic test which would be expected to be part of the follow-up diagnostic test battery), and parental report is found to have poor sensitivity and specificity. There is insufficient evidence to comment on the accuracy of combinations of tests.

A small number of studies indicated a generally high uptake in this age group. However, given the experimental design of the studies and the fact that they were assessing test accuracy rather than programme effectiveness, these findings cannot be generalised to the uptake of the screen in real-world community screening settings. The two published studies with evidence of uptake of screening at school entry in real-world settings suggest uptake in excess of 90%, reflecting the 'captive' nature of the population to be screened.^{6,97}

Cost-effectiveness

There are no good-quality published studies that assess the cost-effectiveness of SES, and no full economic evaluations. The two partial economic evaluations that were found were of poor quality and uncertain relevance.

A decision-analytic model was developed to assess the costs, effectiveness and net benefit of SES when compared with no SES and SES using alternative screening tests. The primary source of data about the accuracy of the screening tests for

the economic model was the data included in the systematic review reported in Chapter 4. This was supplemented by data on the prevalence and distribution of hearing impairment, the probability of a child being screened, diagnosed and treated, the costs of screening, diagnosis and treatment, and the outcomes of screening from published literature, the survey of current practice (Chapter 2), the Waltham Forest observational study (Chapter 3), national statistics and databases, local accounts and expert opinion.

For the 1-year time-horizon used in the primary analysis, SES-C was associated with higher costs and slightly higher QALYs compared with no SES and other SES alternatives. The ICER for SES-C is around £2500 per QALY gained. The range of expected costs, QALYs and net benefits was broad, with the 2.5th to 97.5th percentiles of differences in expected costs and outcomes crossing zero, indicating a high level of uncertainty in the conclusions. CEACs and measures of net benefit provide a means of assessing the robustness of differences in expected costs and outcomes, when combined into ICERs. These analyses allow for the fact there may be a relationship between resource use and costs and outcomes, so that, for example, higher resource use and therefore costs may be associated with improved outcomes. Overall, the primary analysis indicated that SES-C was cost-effective compared with all the other SES programmes evaluated and with no SES. The costs of individual SES tests (rather than screening programmes) were estimated to be approximately £8 per screening test (see *Table 42*, Chapter 5). It is the costs of the screening tests that dominate the total expected costs of screening. The costs of the screening tests may have been overestimated if the duration of the tests reported in the survey of current practice also included the time needed to conduct wider health checks. The survey of current practice (Chapter 2) indicated that up to 60% of programmes included the SES in wider health checks all the time; if this is the case, then the cost of each screening test may be lower and the cost-effectiveness of SES higher than estimated here.

A number of secondary analyses was used to explore subgroups of the data and test assumptions used in the model. However, the use of subsets of the data, with relatively few studies to combine and small sample sizes, means that these analyses can be exploratory only. The secondary analyses supported the result that SES-C is cost-effective compared with no SES and alternative SES models. Furthermore, the analyses using the 6- and 11-year time-horizons supported this

conclusion, with SES-C being more cost-effective in over 99% of simulations.

In the economic model, SES-C is a weighted composite reflecting the pure tone sweep test (99%) with tympanometry (1%). When SES-C was compared with the SES as it is mostly practised at present in the UK (i.e. pure tone sweep only), the latter was more cost-effective in 80% of simulations. The estimates of the costs and QALYs of SES-C were based primarily on the pure tone sweep as the screening test. The studies included in the systematic review (Chapter 4) indicate that this test has relatively high sensitivity and specificity compared with alternative tests; however, the accuracy of the pure tone sweep was assessed in trials settings, rather than in the varied and less than ideal settings encountered in routine practice. This may mean that the accuracy and therefore the cost-effectiveness of SES-C are overestimated, compared with no SES. However, the analysis comparing SES using alternative tests with lower accuracy indicated that SES might still be more cost-effective than no SES.

When SES-C was compared with targeted screening at school entry, if the targeting accurately detected 90% of children with a hearing impairment then targeted screening was more cost-effective than universal screening. However, if the identification of children at risk of hearing impairment for targeted screening was associated with low sensitivity and specificity, then universal screening was more cost-effective than targeted screening.

The economic analyses used a relatively low prevalence of moderate or worse previously unidentified permanent hearing impairment. Reducing this further to model the potential impact of newborn hearing screening reduced the proportion of simulations when SES was cost-effective to around 60%. Decision-makers also needed to be willing to pay over £6000 to gain 1 QALY for SES to be cost-effective in more than 50% of cases.

Overall, because of the lack of primary data and the necessarily wide limits for variables in the modelling, these results must be considered indicative and exploratory only.

The OME issue and some further analyses

Hearing impairment of a mild degree is also associated with transient episodes of OME, which

is much more prevalent in children than is permanent hearing impairment.¹⁵ Some people have argued the case for a screen at school entry to identify previously unknown cases of children with OME that is of a severity and/or persistence sufficient to require treatment.

There has been extensive although generally poor-quality research on the treatments for OME in children, much of which fails to address the question of which subtypes of 'OME child' benefit from treatments. A recent meta-analysis⁹⁸ accessing individual patient data from several trials confirmed the accepted conclusion that, for well-defined cases, ventilation tubes (grommets) do improve hearing for so long as they are in place. However, for the most persistent or recurrent cases (i.e. those for whom the certainty of selection for surgery is greatest) the condition tends to return, leading to the need for reinsertion(s) of grommets. Age, within the range of about 3–8 years, does not seem to be a characteristic of major importance for results, provided that children meet a criterion for persistence and severity.

The most comprehensive and sophisticated evidence on candidature for intervention in OME is emerging from the UK TARGET randomised trial,⁹⁹ of which the aspects of particular relevance here are mostly yet to be published. The trial does not contradict the above simple statements, but documents more fully the breadth and duration of benefits from adjuvant adenoidectomy and the criteria for selection of a subgroup within which the combined treatment is highly effective (Haggard M, University of Cambridge: personal communication, 2006). Professor Haggard informed the review team via several presentations given at international meetings and extensive annotated analyses which show that the largely null results on young mild cases emerging from screening did not apply to older (>3.75 years) and better selected cases, but that consistent if modest benefits are shown in the TARGET data. Thus, in relation to the Wilson–Jungner principles for screening, at one level an effective and available intervention does exist for children of school entry age. There is still no convincing and favourable evidence for types of treatment other than these surgical operations.

The implementation of an overall screening and treatment programme is less satisfactory than the above statement suggests. The evidence on this point comes from children of younger age than school entry, but there is no good reason for it not

to apply. Two trials of ventilation tubes in OME have been published in recent years, making essentially the same point.^{100,101} They are summarised here because, although restricted, they are of high internal validity and are particularly relevant to the issue of the caseloads that screening tends to find: the marginal rather than the extreme. The Rovers trial was done on children referred from the implementation of the 8-month IDT screen in The Netherlands, although by the time they had been through confirmation of fluid in the ears, they were around 2 years of age. As well as an ear status measure, there was a language test and a quality of life scale. The Paradise trial recruited slightly older children in the USA to whom it was possible to give a wider range of assessments of valued outcomes including performance tests. These children had been referred by paediatricians who exercise a highly surveillant semi-specialised form of childcare in the medically insured part of the US population.

Both of these trials found that the placement of ventilation tubes did give the known short-term benefits to ear status or hearing, but did not improve wider valued measures of outcome. These null results make useful political points for the two countries concerned, both having high intervention rates, about overtreatment in routine practice in the past, due to selection of cases that are too mild and/or insufficiently persistent to benefit. A widespread misinterpretation of these trials, imagining that they suggest that ventilation tubes 'do not work', has led to unnecessary avoidance of their real message. In many conditions it is hard to show a knock-on from either disease or treatment into valued outcomes, in relation to other powerful sources of influence on those same outcomes. The indirect knock-on from fluctuating hearing impairment and physical health problems in OME, and its treatment, into language and other developmental outcomes make that challenge particularly hard. The knock-on benefits would be expected to be rather slight. The evidence suggests that it is particularly slight in very young caseloads emerging from screening or surveillance, where the rate of spontaneous remission in untreated controls is high, and particularly so where the case entry criteria are mild.

For the preceding reasons, TARGET recruited children from the NHS who had already undergone gate-keeping. This typically includes some initial caution by the GP over the need for referral, several months (in most districts) of

waiting to be seen in secondary care, and being subject to a further 3-month formal watchful waiting period to establish persistence or recurrence. These were older children (3.5–7 years) and only randomised on meeting a severity criterion of hearing thresholds of 20 dB HL or greater in the better ear a second time after 3 months of watchful waiting. TARGET did produce some statistically significant and clinically material benefits to physical health and development over the 2 years following treatment as well as to hearing, but these benefits were rather modest taken as a whole. Given awareness of this difference in caseload, the results from TARGET and the other two trials are not inconsistent. Further analyses of the hearing level data from TARGET established that children with more severe hearing levels do indeed receive more benefit to hearing, as expected. The finding is encouraging for tests of hearing being relevant for screens for OME. The issues then for screening in OME as a fluctuating condition are: (1) how few sequential stages will suffice after some initial universal screen to define a small caseload that approximates the severity and persistence of that in TARGET, and (2) whether for the cost of such successive testing, the incremental yield over what would have been referred reactively at this age is worth that cost.

There is no published evidence that addresses the foregoing two questions. Both a national study⁶ and the historical cohort from Waltham Forest (Chapter 3) have shown that just under 3% of those screened were referred from the SES with OME. This represents a large proportion of all those referred by the SES, with data from the survey of current practice suggesting a median for positive predictive value for temporary conductive hearing impairment of 36%. In the Waltham Forest cohort, new cases (i.e. not previously known to services) amounted to 1.4% and of these half (0.7%) needed ENT referral; details thereafter were not available, and interpretation of the value of making these referrals is therefore difficult.

However, some light can be thrown on the contribution of the present SES system by a further analysis of cases seen in the TARGET recruitment stages. SES screen referrals to ENT services for possible surgery tend to go through community paediatricians who specialise in audiology (e.g. members of BACDA), rather than through GPs. This distinction is not hard and fast: not all community referrals will have originated with screens, although many will, and nearly all ex-screen referrals will arrive from this source. The

TARGET RCT had already shown on its database of over 3000 cases that referrals from community paediatricians had higher positive predictive value than those direct from GPs.⁹⁹ Because the lead-in stages to the randomised trial had recruited at a dozen ENT departments throughout the UK, the trial chief investigator (Haggard) and colleagues were asked to probe whether these analyses could be extended to say anything about the yield from SES.

A set of analyses was run on over 4000 referrals and a document annotating the results was supplied to the present team, of which the following is a summary. Although the TARGET study had not asked about screening at the level of either individual case or contributing district (clinic), it had a useful degree of indirect leverage via (1) the age distribution being centred on 5 years, the age of maximal relevance to SES and (2) a distinction between GP and community referrals. The question that this permits to be answered is one of yield: whether, taking GP-referred cases as a control set, the number or severity of community-referred children in this database increases from around 5 years of age, when contrasting the preceding 2 years of cross-sectional age with the following 2 years. This involves an interaction with age. The main-effect advantage for community mentioned above was again found to be pervasive and reflects the availability in community services of audiometry, some specialist expertise and a tendency to retest to establish persistence. Analyses of this type broken down by age and source were conducted on the severity (a marker of positive predictive value and specificity) and numbers of cases (yield). Effectiveness of screening would predict the highest values to occur in the ex-community over-5-year-olds. However, in neither numbers nor severity was there an interaction between source (GP/community) and age band (before/after modal SES age). Thus, the combination of number and severity of cases coming through the community specifically after SES age is not large enough to show up in the ENT caseload. It is therefore probably not large enough either to represent a distinct societal benefit.

Conclusions

The evidence from the national survey of current practice is that:

- the SES is in place in most areas of England, Wales and Scotland; data from Northern

Ireland were too few to draw any conclusions that might generalise there; just over 10% of respondents had abandoned the screen, while others were awaiting guidance in the light of the national implementation of newborn hearing screening

- coverage of the SES is variable, but is often over 90% for children in state schools; coverage is poor for private schools and home-educated children
- referral rates are variable, with a median of about 8%
- the test used for the screen is in all cases the pure tone sweep test; however, there is a wide variety of implementations of this, with differing frequencies, pass criteria and retest protocols; written examples of protocols were often poor and ambiguous
- there is no national approach to data collection, audit and quality assurance, and there are variable approaches at local level; a small proportion of services was able to provide audit data on coverage referral rates and yields, but these were often of doubtful quality, especially with respect to yield
- the screen is performed in less than ideal test conditions; this probably increases the referral rates and decreases accuracy
- resources for replacement equipment, calibration and screener training are said to be limited and impacting on the quality of the screen in many areas.

The evidence from the observational studies in Waltham Forest is that:

- the prevalence of permanent childhood hearing impairment continues to increase through infancy owing to acquired, late-onset and progressive hearing impairment, in line with published evidence; of the 3.47 in 1000 children with a permanent hearing impairment at school screen age, 1.89 in 1000 required identification after the newborn screen; a high proportion of these appeared to have identifiable risk factors
- the introduction of newborn hearing screening is likely to reduce significantly the yield of a universal SES for permanent bilateral and unilateral hearing impairments; the yield of the SES in Waltham Forest for such impairments has fallen from about 1.11 in 1000 before newborn screening to about 0.34 in 1000 for cohorts who have had newborn screening, of which only 0.07 in 1000 are unilateral impairments
- surveillance procedures, comprising (at least) reactive services to parental and professional

concern, are an important route to identification of cases throughout infancy and the preschool years

- in the Waltham Forest cohort studies, small but material numbers of children with permanent hearing impairment remained to be found at or before school entry; just under 20% of permanent moderate or greater bilateral, mild bilateral and unilateral impairments, known to services as 6-year-olds or older, remained to be identified around the time of school entry; some of these were late onset or acquired, some 'moved in' and some congenital cases not identified by the newborn screen.

The evidence from the systematic review of the accuracy of alternative tests for the SES and of the effectiveness of interventions is that:

- there were no good-quality published comparative trials identified of alternative screens or tests for SES
- there was one poor quality study which compares screening to no screening, but the results are inconclusive
- studies concerned with the relative accuracy (in terms of sensitivity and specificity) of alternative screening tests are difficult to compare and often flawed by differing referral criteria and differing case definitions; nevertheless, using full PTA as the reference test, the pure tone sweep test appears to have high sensitivity and high specificity for minimal, mild and greater hearing impairments, better than alternative tests for which evidence was identified (otoacoustic emissions, tympanometry, reflectometry, parental questionnaire); some evidence suggests that spoken word tests can have high sensitivity and specificity; no good-quality evidence was identified addressing the accuracy of combinations of tests
- there is insufficient evidence to draw any conclusions about possible harm of the screen as currently performed
- there were no published studies identified which examined the possible effects of SES and subsequent interventions on longer term language, educational or social outcomes.

The evidence from the cost-effectiveness and cost modelling study is that:

- no good-quality published cost-effectiveness studies or economic evaluations of SES were identified
- lack of primary data and the necessarily wide limits for variables in the modelling mean that

any conclusions must be considered indicative and exploratory only

- using decision-analytic modelling, and taking into account all types of hearing impairment, a universal SES based largely or completely on pure tone sweep tests was associated with higher costs and slightly higher QALYs compared with no screen and other screen alternatives; the ICER for such a screen is around £2500 per QALY gained; the range of expected costs, QALYs and net benefits was broad, indicating a considerable degree of uncertainty
- targeted screening can be more cost-effective than universal school entry screening; this depends on there being identifiable risk factors.

The evidence suggests that a national screening programme for permanent hearing impairment at school entry meets all but three of the criteria for a screening programme; namely, knowledge of the distribution of test values in the population with agreed cut-offs, RCTs showing that the screen reduces morbidity, and a national protocol with quality assurance and audit (Appendix 1).

With regard specifically to the issue of the value or effectiveness of the screen finding cases of transient hearing impairment associated with middle ear disorder, the evidence comes from the survey of current practice, a recently published survey,⁶ the Waltham Forest cohort studies and a recent meta-analysis, alongside data from a large and well-controlled UK study as yet largely unpublished (Haggard M, Cambridge University: personal communication, 2006). Collectively, this material suggests that:

- surgical intervention (ventilation tubes) for children with OME improves hearing levels as expected, and has modest but measurable effects on longer term outcomes (physical, developmental) for more severe and persistent cases, but not for milder and marginal cases
- about 3% of those screened are referred with OME, representing perhaps 40% of referrals; some of these cases are already known to services, and some will not require treatment; about one-quarter may require further otological management including surgery; these estimates will have wide confidence intervals
- there is no evidence that the SES is a better source of referrals of more severe and persistent cases of OME than reactive GP referrals, although data with a more rigorous sampling frame and prospective analysis are required to confirm that there is evidence for no effect

- at least six of the accepted criteria for a screening programme for OME are currently not met (Appendix 1).

Conclusions: closing comment

In 1987, Stewart-Brown and Haslum³ published the results of a survey of national practice (in England and Wales) with regard to screening for hearing impairment in childhood. In it, they commented that “the number of times that children were screened at school varied considerably ... the hearing level at which children were referred after sweep audiometry varied among districts ... only 73 [out of 165] could report the referral rate ... very few districts were collecting the sort of data that would allow them to make even the most rudimentary assessment of their screening programmes, far less any evaluation of cost consequences or benefits ... secretory otitis media fulfils few of the criteria that should be met before a screening programme can be considered likely to be either effective or ethical”. While a considerable number of lead clinicians for the SES have tried to implement an improved programme in the face of resource constraints and competing priorities, the overall similarity between the present team’s findings in 2005 and Stewart-Brown and Haslum’s in 1984³ is striking. They proceeded to comment on the need for well-controlled studies to underpin policy changes; two decades on, the lack of a good-quality evidence base to drive change in this area remains a serious problem.

Implications for practice

There is some evidence that significant numbers of children with permanent hearing impairment remain to be identified at school entry. There is evidence that the pure tone sweep test if properly implemented as a screen can have reasonable levels of sensitivity and specificity as a route to identification of these children. Although there is little evidence on the effectiveness of the SES, there is also little evidence to judge whether it is ineffective. The implications of the conclusions summarised in the section ‘Conclusions’ (p. 82) are that services already implementing the SES (the overwhelming majority in the UK) should continue to do so, pending later evidence-based policy decisions (see in the next section), but that they should make every effort to implement a clear test and screen protocol, and that they should audit the screen performance for cases of PCHI not already known to services. However, the case for using the SES as a route for finding

children with OME suitable for intervention is weak (see Appendix 1).

Recommendations for future research

It is evident that in most areas of the UK there are service leads who value the SES, and who head up services that deliver a screen in the main based on the pure tone sweep test to most children in public education. However, test and screen protocols vary between districts, there is little evidence of explicit quality assurance procedures, and data management systems are generally poor or non-existent, so that data on screen performance, let alone longer term outcomes, are largely absent.

The public health context in which the SES is delivered is changing significantly, and by about 2010 almost all babies born in the UK will have undergone newborn hearing screening. Not all PCHI will be identified by newborn screening: late-onset and acquired impairments, children moving in who have not had a newborn screen and cases not picked up by the newborn screen (largely mild and minimal hearing impairments) will remain to be identified. The justifiable means by which such cases could be identified include parental and professional concern, formal surveillance of some kind at defined ages, targeted school entry screening and universal school entry hearing screening. The evidence required to make policy decisions between such alternatives does not exist. The following recommendations for future research and audit are made with the overall aim of being able to make evidence-based policy decisions in or around 2012, when all school entry cohorts will have had newborn hearing screening.

A priority need is for the establishment of a single, agreed national protocol for those services delivering the SES to make future studies and future audit of screen performance more directly comparable. On the basis of the evidence, such a protocol should be based on detection of pure tones. It is known that the greater the level of permanent hearing impairment, the poorer the quality of life is likely to be, while the lower the pass threshold for the screening test, the poorer the test and screen specificity. Furthermore, PCHI tends to be worse in the higher frequencies, and the testing of low and mid-frequencies in the conditions under which the SES has to be performed is particularly subject to noise interference. Thus, there are persuasive arguments for a single, high-frequency (4 kHz) pure tone

detection test set at a level likely to be most effective and efficient (25 dB HL). Although the case for screening for OME at school entry is weak (see Appendix 1), there may be persistent or severe cases who remain to be identified at school entry and who would benefit from surgical intervention; a high-frequency 25 dB HL criterion for referral would, as an incidental benefit, increase the positive predictive value for such cases.

Alongside an agreed national protocol must go systems for data monitoring and quality assurance so that robust data on screen accuracy and effectiveness can be collected. Such systems are in place for newborn hearing screening, and R&D is required to develop a single data system as part of a national plan around screening for childhood hearing impairment and paediatric audiology services.

There is a need to establish with greater certainty the prevalence of permanent mild and minimal hearing impairment at school entry that could be identified by a suitable quality-assured screen protocol, and to confirm the prevalence and severity distribution of congenital unilateral hearing impairment.

Comparative trials are needed to compare the effectiveness, efficacy and efficiency of alternative approaches to the identification of permanent hearing impairment postnewborn screen.

Specifically, a comparison of reactive services, a formal surveillance procedure between fourth and fifth birthdays,⁷ targeted screening between fourth and fifth birthdays, and universal screening at school entry age would establish the necessary evidence for policy decisions. Targeted screening would be based on children who had not received or completed newborn screening, children attending child development centres, children coming from families with a history of permanent childhood deafness, and children who have suffered bacterial meningitis or a childhood viral illness leading to doubt about their hearing.

The lack of prospective controlled studies on the effectiveness of hearing screening and subsequent interventions in terms of later outcomes for children with permanent mild, minimal and unilateral hearing impairment identified at school entry represents a major gap in the evidence base. However, it is not clear that such studies would take priority over better data on alternative protocols, uptake, yield and diagnostic accuracy; furthermore, there are real problems in identifying appropriate outcome measures that have sufficient sensitivity.

The distribution of detection thresholds for pure tones in the population at school entry, and how different cut-off criteria would relate to measures of hearing disability (Wilson and Jungner criterion no. 5³⁸), are not known. Research is needed to establish these.



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Contribution of authors

John Bamford (Professor, Specialist in Paediatric Audiology and Deaf Education) led the research

team. Heather Fortnum (Associate Professor, Epidemiologist) led the survey and associated analyses of current practice and coordinated the editing of the final report. Kirsty Bristow (Research Assistant, Epidemiologist) conducted the survey and analyses of current practice, and coordinated effort across the team. Jenny Smith (Research Assistant, Epidemiologist) conducted the reviews of accuracy and effectiveness of screening tests, and coordinated the editing of the draft report. Georgios Vamvakas (Research Assistant, Health Economist) conducted the health economics reviews, modelling and analyses. Linda Davies (Reader, Health Economist) led the health economics reviews, modelling and analyses. Rod Taylor (Reader, Health Statistician) led the reviews of accuracy and effectiveness of screening tests. Pete Watkin (Consultant Community Paediatrician, Physician in Paediatric Audiology) led the data collection and analyses of the Waltham Forest studies. Sarita Fonseca (Consultant Community Paediatrician, Physician in Paediatric Audiology) advised on the survey of current practice and provided a direct link with the previous recent study of current practice. Adrian Davis (Professor, Director of NHSP, Epidemiologist) advised on all aspects of the research and provided a direct link with the newborn hearing screening programme. Sally Hind (Senior MRC Scientist, Developmental Psychologist) advised on the survey of current practice. All authors contributed to writing and editing.



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Appendix I

Screening at school entry for childhood hearing impairment: an appraisal against National Screening Committee criteria

Screening for permanent childhood hearing impairment (June 2006)

Criteria	Supporting evidence
The condition	
1. The condition should be an important public health problem	<p>Bilateral PCHI can have a devastating impact on communication skills (Conrad, 1979), educational attainment (Wood <i>et al.</i>, 1986), and quality of life (Gregory, 1995; Cheng <i>et al.</i>, 2000), with a high cost to society (Mohr <i>et al.</i>, 2000).</p> <p>Unilateral hearing impairment would be expected to affect auditory perception in various predictable ways (e.g. poor localisation of sound sources, difficulty in noisy or reverberant environments such as schools), and there is some evidence of detrimental effects on academic progress (e.g. Bess <i>et al.</i>, 1998)</p> <p>Authors' summary opinion: satisfied</p>
2. (i) The epidemiology of the condition should be known	No national register of hearing-impaired children exists for the UK, and accurate estimates of the prevalence of PCHI and of its profile across all ages and all degrees of impairment are unavailable
(ii) The natural history of the condition should be understood	PCHI of a moderate degree or greater (i.e. detection thresholds >40 dB HL averaged across 0.5, 1, 2 and 4 kHz) is present at birth at a rate of about 1.6 per 1000 live births, of which approximately 1.0 in 1000 are bilateral and 0.6 in 1000 are unilateral impairments (Davis <i>et al.</i> , 1997; Bamford <i>et al.</i> , 2006). In terms of incidence, this means that in the UK about 800 children per year will be born with permanent bilateral hearing impairment of a moderate or greater degree, and about 500 per year will be born with unilateral hearing impairment (i.e. hearing within normal limits in one ear, hearing impairment of moderate or greater degree in the other ear)
(iii) There should be a recognised latent period or early symptomatic stage	<p>There is good evidence that the prevalence of permanent bilateral moderate or greater hearing impairment increases through the first decade of childhood (Fortnum <i>et al.</i>, 2001; Fortnum, 2003). It is possible that the prevalence of bilateral moderate or greater impairment reaches 2 in 1000 by the age of about 9 years. If permanent mild hearing impairments are included, evidence from retrospective ascertainment studies in this review suggests that by school entry the prevalence is around 3.5 in 1000.</p> <p>There are four situations where children with permanent hearing impairment may not be identified by a screening test within a few days of birth:</p> <ul style="list-style-type: none"> • Some children will have no impairment at birth, but will acquire the impairment later in their life as a result of some traumatic event such as infection (usually bacterial meningitis, Fortnum, 1992), head injury (Zimmerman <i>et al.</i>, 1993), ototoxic therapy (Casano <i>et al.</i>, 1999) or chemotherapy (Littman <i>et al.</i>, 1998; Berg <i>et al.</i>, 1999). • Some children may have an impairment at birth, but of a severity insufficient to be detected by the newborn screening procedures. As the child grows this mild impairment may represent a significant disabling condition in itself, or the impairment may progress to a greater severity (Hayes and Dreith, 2000). The causes of progressive impairments include hereditary hearing loss and syndromal associations such as Alport, Waardenburg type II and Alström (Gorlin <i>et al.</i>, 1995; Zwirner and Wilichowski, 2001), infectious diseases (Williamson <i>et al.</i>, 1992), anatomical malformations (Zalzal <i>et al.</i>, 1995; Shetty <i>et al.</i>, 1997), perinatal events and treatments (Fujiwaka <i>et al.</i>, 1995; Lasky <i>et al.</i>, 1998) and ototoxic drugs (Pasic and Dobie, 1991; Borradori <i>et al.</i>, 1997).
<i>continued</i>	

Criteria	Supporting evidence
	<ul style="list-style-type: none"> • Some children develop genuine late-onset impairment that develops with no obvious causative factor and hence is not truly acquired (Parker, 1999). • A fourth group of children contributing to those who may not be identified at birth are those who should undergo the screen but who do not. Reasons for this at the time of screening include early discharge and/or parental refusal, but children who migrate into an area or country implementing a neonatal screening programme from an area/country which does not, also fall into this group. <p>Children in all four of these categories comprise those who require identification postneonatal screen and who will need some form of follow-up to be established</p> <p>The evidence on permanent unilateral hearing loss is more limited. Although it appears from NHSP data that the prevalence at birth is about 0.6 in 1000, it is not known whether there are significant numbers of later onset cases, whether some of the losses are progressive, and whether there is a tendency for congenital or postnatal unilateral hearing loss to progress to bilateral loss</p> <p>PCHI of whatever cause does not improve. It may remain stable or worsen (progressive)</p> <p>There is no latent period or early symptomatic stage in PCHI</p> <p>Authors' summary opinion: satisfied</p>
3. All cost-effective primary prevention interventions should have been implemented as far as practicable	<p>Primary prevention includes immunisation for conditions that are known to cause permanent hearing impairment, both prenatally and postnatally (e.g. rubella, mumps, meningitis); reduction in the use of, and monitoring of levels of, ototoxic antibiotics such as gentamycin in the neonatal period; and genetic counselling for people with affected children or at higher risk of having an affected child</p> <p>Authors' summary opinion: satisfied</p>
The test	
4. There should be a simple, safe, precise and validated screening test	<p>The procedures for the SES vary in their implementation, but all are relatively simple. There is no known danger to the child or to the screener in performing the test. Limited quality evidence suggests that the test has high sensitivity and specificity for full PTA (Orlando and Frank, 1987; FitzZaland and Zink, 1984; Holtby <i>et al.</i> 1997; Sabo <i>et al.</i>, 2000)</p> <p>There has until recently been a widespread if implicit consensus that "the pure tone sweep test has value educationally and as a safety net to catch any deficiencies of the earlier screening system in the overall public health provision" (Haggard, 1993), a position broadly endorsed by Hall (2003)</p> <p>The review of current practice indicated that all but 12.2% of respondents operate a universal school entry screen and use the pure tone sweep test; however, the protocols used are unacceptably variable</p> <p>Authors' summary opinion: satisfied</p>
5. The distribution of test values in the population should be known and a suitable cut-off level defined and agreed	<p>There are no published data on population values for pure tone sweep audiometry. A cut-off level has not been defined and agreed and varies across the national provision</p> <p>Population data on pure tone audiometric levels are not available for children of school entry age; adult norms are used. This may have marginal effects on case identification of mild and minimal hearing impairments, but not on moderate and greater impairments</p> <p>Authors' summary opinion: not satisfied</p>
6. The test should be acceptable to the population	<p>The pure tone sweep test and PTA are well-established tests and appear to be acceptable to the population (children) and their parents, although no data have been published that address this issue</p> <p>Authors' summary opinion: satisfied</p>

continued

Criteria	Supporting evidence
7. If the test is for mutations the criteria used to select subset of mutations to be covered by screening should be clearly set out	<p>Although hearing impairment may be caused by inherited or novel mutations, this screening test is not designed to identify them. Further diagnostic evaluations may include selective mutation screening/testing</p> <p>Authors' summary opinion: not relevant</p>
8. There should be an agreed policy on the further diagnostic investigation of individuals with a positive test and on the choices available to those individuals	<p>In 1976 the Court Report recommended that hearing screens be carried out at least twice in school (Court, 1976). However, there was no nationally agreed protocol for the screen, and implementation thus varied in small but possibly important details across services</p> <p>Guidelines on diagnostic investigation and subsequent treatment choices have been developed by the British Society of Audiology in collaboration with the National Deaf Children's Society (NDCS, 2006). For moderate and greater bilateral hearing impairments in school-age children, diagnostic procedures are well established and reliable when performed by trained paediatric audiologists. Intervention options include amplification (hearing aids), communication advice, educational support and social care support. For mild and unilateral hearing impairment, diagnostic procedures are more challenging, especially if transient middle ear conditions are also present, and require good-quality paediatric audiology services and paediatric audiologists, of which there is a national shortage. Intervention options are similar to those with more severe impairments, but evidence on the cost-effectiveness of these options is largely missing</p> <p>Authors' summary opinion: partially satisfied</p>
The treatment	
9. There should be an effective treatment or intervention for patients identified through early detection	<p>It has long been believed that earlier identification of hearing impairment must lead to better outcomes, and there is now reliable evidence that this is so in the domains of communication, educational achievement and quality of life (Davis <i>et al.</i>, 1997; Yoshinago-Itano <i>et al.</i>, 2000; Moeller, 2000). Few people now disagree with the statement that identification of congenital impairments in the first few months of life and consequent habilitation is desirable. For late-onset, progressive and otherwise not previously known impairments identified at school entry age, evidence on the effectiveness of the interventions (provision of hearing aids and regular follow-up with appropriate rehabilitative support particularly in education) is absent. It is reasonable to assume that the intervention will be effective for moderate and greater bilateral impairments; more evidence is required about intervention for mild, minimal and unilateral impairments. However, since these could be at risk for worsening impairment, identification and monitoring is arguably desirable as a minimum</p> <p>Authors' summary opinion: satisfied</p>
10. There should be agreed evidence-based policies covering which individuals should be offered treatment and the appropriate treatment to be offered	<p>There is still debate over the lower level of hearing impairment for which provision of hearing aids is beneficial. More evidence is required with respect to interventions for mild, minimal and unilateral impairments identified at school age. The evidence for the type and extent of intervention for children with moderate and greater bilateral impairments is relatively clear</p> <p>Authors' summary opinion: partially satisfied</p>
11. Clinical management of the condition and patient outcomes should be optimised by all healthcare providers prior to participation in a screening programme	<p>There is evidence from the NHSP (www.nhsp.info) that the quality of paediatric audiology services in the UK is unacceptably variable. This is likely to be a resource and training issue and is receiving attention</p> <p>Authors' summary opinion: partially satisfied</p>

continued

Criteria	Supporting evidence
12. There should be evidence from high-quality RCTs that the screening programme is effective in reducing mortality and morbidity	<p>There is no evidence from high-quality RCTs that the screening programme is effective in reducing morbidity</p> <p>Authors' summary opinion: not satisfied</p>
13. There should be evidence that the complete screening programme (test, diagnostic procedures, treatment/intervention) is clinically, socially and ethically acceptable to health professionals and the public	<p>Screening and intervention for childhood hearing impairment are clinically, socially and ethically acceptable to all health professionals concerned with the issue and to most of the public. The majority of health professionals who took part in the survey of current practice were strongly in favour of the SES. There is an important minority of the Deaf community who subscribe to a social/cultural model of deafness and who do not support some of the 'corrective' interventions for severe/profound hearing impairment; however, this does not apply to the mild/moderate levels detected by the SES</p> <p>Authors' summary opinion: satisfied</p>
14. The benefit from the screening programme should outweigh the physical and psychological harm (caused by the test, diagnostic procedure and treatment)	<p>The extent of the beneficial effects of early identification for children with permanent hearing impairment on developmental outcomes in general and communication in particular has been demonstrated for young preschool infants (e.g. Yoshinaga-Itano <i>et al.</i>, 1998); there is no evidence of either benefit or harm associated with the SES</p> <p>Authors' summary opinion: partially satisfied</p>
15. The opportunity cost of the screening programme (including testing, diagnosis and treatment) should be economically balanced in relation to expenditure on medical care as a whole	<p>There is no good-quality published evidence of the costs and effectiveness of the screen. The cost-effectiveness modelling carried out suggested that each screen costs £8, and that a universal school entry screen based largely or completely on pure tone sweep tests was associated with higher costs and slightly higher QALYs compared with no screen and other screen alternatives; the ICER for such a screen is around £2500 per QALY gained; the range of expected costs, QALYs and net benefits was broad, indicating a considerable degree of uncertainty</p> <p>Authors' summary opinion: partially satisfied</p>
16. There should be a plan for monitoring and managing the screening programme and an agreed set of quality assurance standards	<p>Several reviews (Stewart-Brown and Haslum, 1987; Haggard and Hughes, 1991; Davis <i>et al.</i>, 1997; Fonseca <i>et al.</i>, 2005) have recommended monitoring and management strategies for the SES; the evidence from the survey of current practice suggests that none has been widely implemented, and that little has changed since the review of Stewart-Brown and Haslum. There is no national protocol or quality assurance plan</p> <p>Authors' summary opinion: not satisfied</p>
17. Adequate staffing facilities for testing, diagnosis, treatment and programme management should be available prior to the commencement of the screening programme	<p>Resources to perform the screen vary across different implementations of the programme nationally. Many responses to the survey in the current report highlight deficiencies in staff numbers and experience, facilities and equipment</p> <p>Authors' summary opinion: partially satisfied</p>
18. All other options for managing the condition should have been considered (e.g. improving treatment, providing other services)	<p>Suggestions for universal screens for hearing impairment at different ages have been made and implemented. The most important is the universal newborn hearing screen, fully implemented in England as the NHSP with effect from March 2006</p> <p>Interventions have remained stable in recent years</p> <p>Authors' summary opinion: satisfied</p>

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Zwirner P, Wilichowski E. Progressive sensorineural hearing loss in children with mitochondrial encephalomyopathies. *Laryngoscope* 2001;**111**:515–21.

Screening for temporary childhood hearing impairment (June 2006)

Criteria	Supporting evidence
The condition	
1. The condition should be an important public health problem	Temporary hearing impairment associated with middle ear fluid (OME, sometimes known as glue ear) is a common condition in childhood, particularly up to about 8 years of age (Casselbrant and Mandel, 2003). The hearing loss may be unilateral or more commonly bilateral, minimal, mild or occasionally moderate in degree; evidence for long-term effects is sparse Authors' summary opinion: partially satisfied
2. (i) The epidemiology of the condition should be known	The period prevalence (0–8 years) for OME is around 80% (Casselbrant and Mandel, 2003), while the point prevalence may be as high as 20% at 2 and 4 years of age (Zielhuis <i>et al.</i> , 1990). Risk factors include socio-economic group, passive smoking, bottle feeding, upper respiratory tract infections, craniofacial anomalies and time on the neonatal intensive care unit at birth (Casselbrant and Mandel, 2003)
(ii) The natural history of the condition should be understood	The natural history of the condition is only partly understood. Spontaneous remission is common, with no long-term effects. About 5% of cases exhibit severity of the hearing impairment and persistence/recurrence of the condition sufficient to cause concern
(iii) There should be a recognised latent period or early symptomatic stage	Hearing impairment is a major symptom, caused by middle ear fluid impeding the passage of acoustic energy from outer to inner ear. The degree of impairment varies with the presence and viscosity of the fluid Authors' summary opinion: satisfied
3. All cost-effective primary prevention interventions should have been implemented as far as practicable	Public health initiatives for children and families with young infants are likely to have an important effect on the condition Authors' summary opinion: satisfied
The test	
4. There should be a simple, safe, precise and validated screening test	The procedures for the SES vary in their implementation, but all are relatively simple. There is no known danger to the child or to the screener in performing the test. Limited quality evidence suggests that the test has high sensitivity and specificity for full PTA (Orlando and Frank, 1987; FitzZaland and Zink; 1984, Holtby <i>et al.</i> , 1997; Sabo <i>et al.</i> , 2000) There has until recently been a widespread if implicit consensus that "the pure tone sweep test has value educationally and as a safety net to catch any deficiencies of the earlier screening system in the overall public health provision" (Haggard, 1993), a position broadly endorsed by Hall (2003)

continued

Criteria	Supporting evidence
	<p>The review of current practice indicated that all but 12.2% of respondents operate a universal school entry screen and use the pure tone sweep test; however, the protocols used are unacceptably variable</p> <p>Authors' summary opinion: satisfied</p>
<p>5. The distribution of test values in the population should be known and a suitable cut-off level defined and agreed</p>	<p>There are no published data on population values for pure tone sweep audiometry. A cut-off level has not been defined and agreed and varies across the national provision. Evidence on a suitable cut-off level for severity of hearing impairment associated with OME is emerging from the TARGET trial data (Haggard M, University of Cambridge: personal communication, 2006), but the requirement of persistence as a marker of potential to benefit requires repeat tests (screen or follow-up)</p> <p>Population data on pure tone audiometric levels are not available for children of school entry age; adult norms are used. This may affect case identification of mild and minimal hearing impairments</p> <p>Authors' summary opinion: not satisfied</p>
<p>6. The test should be acceptable to the population</p>	<p>The pure tone sweep test and PTA are well-established tests and appear to be acceptable to the population (children) and their parents, although no data have been published that address this issue</p> <p>Authors' summary opinion: satisfied</p>
<p>7. If the test is for mutations the criteria used to select subset of mutations to be covered by screening should be clearly set out</p>	<p>Authors' summary opinion: not relevant</p>
<p>8. There should be an agreed policy on the further diagnostic investigation of individuals with a positive test and on the choices available to those individuals</p>	<p>Further diagnostic investigation to confirm hearing impairment and middle ear fluid involves air and bone conduction PTA and acoustic impedance measures, with otoscopy and ENT examination; these are standard procedures.</p> <p>Authors' summary opinion: satisfied</p>
<p>The treatment</p>	
<p>9. There should be an effective treatment or intervention for patients identified through early detection</p>	<p>The main treatment option is surgery to drain the fluid and insert ventilation tubes (grommets), possibly with adjuvant adenoidectomy. This restores hearing to normal, but there is no evidence for longer term benefits in marginal cases (Rovers <i>et al.</i>, 2005). In severe and persistent cases there is emerging evidence for modest benefit from treatment on physical and developmental measures (Haggard M: personal communication). It is reasonable to argue that reactive services will know of these cases before school entry, but evidence is lacking. Education-based interventions may also be effective in reducing temporary disability</p> <p>Authors' summary opinion: partially satisfied</p>
<p>10. There should be agreed evidence-based policies covering which individuals should be offered treatment and the appropriate treatment to be offered</p>	<p>In severe and persistent cases there is emerging but as yet unpublished evidence for modest longer term benefit from surgical intervention, as well as the immediate expected benefit of hearing restored to near normal (Haggard M: personal communication). More work is required on case definition and markers of likely benefit from surgery</p> <p>Authors' summary opinion: partially satisfied</p>

continued

Criteria	Supporting evidence
11. Clinical management of the condition and patient outcomes should be optimised by all healthcare providers prior to participation in a screening programme	<p>There is considerable practice variability between services in the UK; management and outcomes therefore vary in ways not related to evidence</p> <p>Authors' summary opinion: not satisfied</p>
12. There should be evidence from high-quality RCTs that the screening programme is effective in reducing mortality and morbidity	<p>There is no evidence from high-quality RCTs that the screening programme is effective in reducing morbidity (Simpson <i>et al.</i>, 2003)</p> <p>Authors' summary opinion: not satisfied</p>
13. There should be evidence that the complete screening programme (test, diagnostic procedures, treatment/intervention) is clinically, socially and ethically acceptable to health professionals and the public	<p>There is no evidence that screening and intervention for childhood hearing impairment associated with OME are clinically, socially and ethically unacceptable to health professionals concerned with the issue and to the public. Universal SES is undertaken by all but 12.2% of service-lead respondents in the current nationwide survey of current practice; surgical intervention for OME is relatively straightforward and risk free, and is very common; parental refusal is thought to be rare. Robust evidence for these statements is sparse, however</p> <p>Authors' summary opinion: partially satisfied</p>
14. The benefit from the screening programme should outweigh the physical and psychological harm (caused by the test, diagnostic procedure and treatment)	<p>The extent of the beneficial effects of early identification for children with transient hearing impairment on developmental outcomes in general and communication in particular has not been demonstrated; there is no evidence of either benefit or harm associated with the SES and treatment for OME, other than evidence of some postsurgical changes noted to the tympanic membrane in some cases who have repeat ventilation tubes (Rosenfeld, 2003)</p> <p>Authors' summary opinion: not satisfied</p>
15. The opportunity cost of the screening programme (including testing, diagnosis and treatment) should be economically balanced in relation to expenditure on medical care as a whole	<p>There is no good-quality published evidence of the costs and effectiveness of the screen. The cost-effectiveness modelling carried out suggested that each screen costs £8, and that a universal school entry screen based largely or completely on pure tone sweep tests was associated with higher costs and slightly higher QALYs compared with no screen and other screen alternatives; the range of expected costs, QALYs and net benefits was broad, indicating a considerable degree of uncertainty</p> <p>Authors' summary opinion: satisfied</p>
16. There should be a plan for monitoring and managing the screening programme and an agreed set of quality assurance standards	<p>Several reviews (Stewart-Brown and Haslum, 1987; Haggard and Hughes, 1991; Davis <i>et al.</i>, 1997; Fonseca <i>et al.</i>, 2005) have recommended monitoring and management strategies for the SES; the evidence from the survey of current practice suggests that none has been widely implemented, and that little has changed since the review of Stewart-Brown and Haslum. There is no national protocol or quality assurance plan</p> <p>Authors' summary opinion: not satisfied.</p>
17. Adequate staffing facilities for testing, diagnosis, treatment and programme management should be available prior to the commencement of the screening programme	<p>Resources to perform the screen vary across different implementations of the programme nationally. Many responses to the survey in the current report highlight deficiencies in staff numbers and experience, facilities and equipment. Resources in paediatric otology departments are variable, with variable linkage with good-quality paediatric audiology; waiting times for surgery for OME in children are variable</p> <p>Authors' summary opinion: partially satisfied</p>

continued

Criteria	Supporting evidence
18. All other options for managing the condition should have been considered (e.g. improving treatment, providing other services)	<p>The most common route for referral of children with this condition is via GPs. It is not clear from the published evidence that improvement of reactive services and surveillance programmes in the preschool period (Hall and Elliman, 2003) would not result in the identification of the children who would benefit from surgery (i.e. those with severe and persistent symptoms)</p> <p>Authors' summary opinion: not satisfied</p>

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Sabo MP, Winston R, Macias JD. Comparison of pure tone and transient otoacoustic emissions screening in a grade school population. *Am J Otol* 2000;**21**: 88–91.

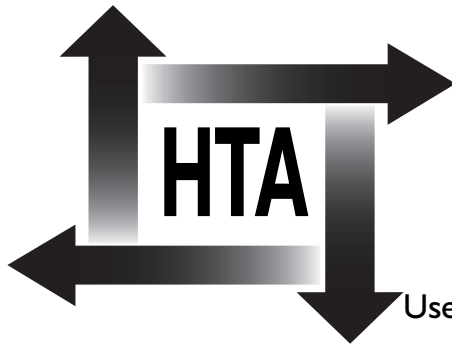
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Appendix 2

Questionnaire used in the survey of national practice



**Health Technology
Assessment Programme
Sponsored Project**

Use of this logo does not constitute endorsement

**Current practice, accuracy, effectiveness and
cost-effectiveness of the
School Entry hearing Screen (SES)**

*A research project commissioned by the
NHS R&D Health Technology Assessment Programme*

District / Area / PCT

Service Lead Name

**Name of person completing the
questionnaire (if different)**

Who should fill in this questionnaire?

This questionnaire should be filled in by the person who is considered the clinical service lead for the school entry hearing screen in your area.

If this is not you then please pass the questionnaire on to the most appropriate person for completion.

Confidentiality

Your answers will be stored on a computer at the University of Manchester and will meet the conditions of the Data Protection Act.

Your answers will be anonymised before they are inputted into the computer. All responses will be kept confidential and they will be seen only by members of the research team.

Questions

If you have any questions or would like to receive a summary report of our findings then please do not hesitate to contact:

Kirsty Bristow

Human Communication and Deafness
School of Psychological Sciences
Humanities Building (Devas)
University of Manchester
Oxford Road
Manchester
M13 9PL

Telephone: 0161 275 8575

e-mail: kirsty.bristow@manchester.ac.uk

BEFORE YOU START

Does your area have a written protocol for the School Entry Hearing Screen
AND
can a copy of this protocol be sent to us?

NO

If you have answered **NO** please go to page 3 and answer as many questions as possible. If you are not sure how to answer a question then please give the best answer you can and write additional comments if you want to.

- YES, a hard copy of the written protocol has been included with the completed questionnaire
- YES, a hard copy of the written protocol has been sent separately
- YES, an electronic version of the written protocol has been emailed to Kirsty Bristow (kirsty.bristow@manchester.ac.uk)

If you have answered **YES** to the above question please read the following statement –

Some of the questions within this questionnaire may already be answered within your written School Entry Hearing Screen protocol.

So....

If the answer is adequately covered by information already given in the protocol just write 'protocol'. However, this will not always be the case and some questions may need more detail or ask for your opinion. Therefore we ask that you read all sections of the questionnaire as carefully as possible.

Please feel free to add comments on any question in the spaces provided at the end of each section or on additional pages.

All comments will be read, so please write as many as you wish

WHO DO YOU TEST?

These questions are designed to find out the kinds of children that are routinely tested by the School Entry Hearing Screen in your area.

1 Please indicate which children are routinely entered into the school entry hearing screen in your area. Please tick one box for each category.

	All	Some	None
Children in state schools			
Children in private schools			
Children who are home educated			
Children in special schools with known physical or sensory disability (excluding hearing loss)			
Children in special schools with known mental disability (excluding children who also have hearing loss)			
Children known to have hearing loss			
Other (please specify in the space below)			

*If you have answered **some** or **none** to any of the above categories it would be very helpful if you could give further details in the space below*

.....

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.....

2 What arrangements (if any) are in place within your area to screen children, for whom you have consent, who did not attend the screen for any reason (e.g. through school absence, had a cold)? Please tick one box for each category

	All of the time	Most of the time	Some of the time	Rarely	Never
Revisit to the school					
Appointment arranged at school health clinic					
Appointment arranged at Audiology clinic					
No arrangement made					
Other (please specify in the space below)					

3 In which school year is the school entry hearing screen routinely performed in your area? Please tick as many answers as apply.

- Preschool
- Reception/Primary 1
- Year 1/Primary 2
- Year 2/Primary 3
- Other (*please specify in the space below*)

Any Comments?

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.....

.....

.....

HOW DO YOU PERFORM THE SCHOOL ENTRY HEARING SCREEN?

For the purposes of this study we need to know what audiological tests are used in the School Entry Hearing Screen, under what conditions these tests are performed and in what locations. By answering all the questions in this section you will enable us to understand these factors for your area

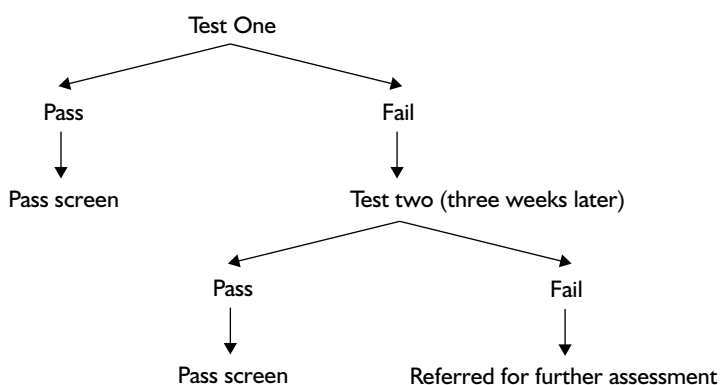
Before completing this section please be aware that for the purposes of this questionnaire we are applying the following definitions –

Screen – the entire remit of tests that a child undergoes before either passing or being referred for further hearing assessment

Test – the individual assessments, which when taken together, form a screen

For example, the diagram below details **one screen** that consists of **either one or two tests** –

Screen Example –



**4 Does your area have any written documentation for the following?
(please tick one box for each category)**

	Documentation?	
	Yes	No
Parent/guardian agreement for the screen		
Information provided to the parent/guardian prior to screening		
Information provided to the parent/guardian prior to referral		
Test protocol		
Re-test protocol		
Referral protocol		

If you answered yes to any of the above categories, could a copy of the document(s) be sent to us?

- Yes, they have been included with this completed questionnaire
- Yes, they have been sent separately
- Copy unavailable

5 Where is the first test within the school entry hearing screen typically carried out in your area?
(please tick one box for each location)

	All of the time	Most of the time	Some of the time	Rarely	Never
School					
Community clinic					
Home					
GP clinic					
Other (<i>please specify in the space below</i>)					

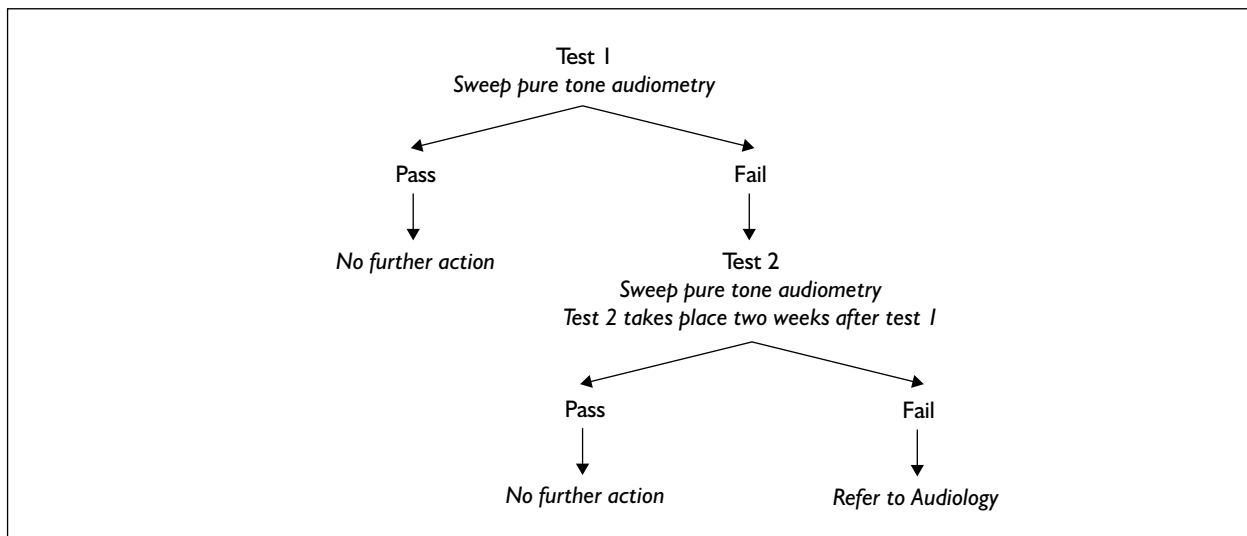
6 Under what conditions is the school entry hearing screen in your area performed? (please tick one box for each category)

	All of the time	Most of the time	Some of the time	Rarely	Never
Soundproof booth					
Sound treated room					
Quiet office					
Noisy office					
Quiet classroom/area					
Noisy classroom/area					
Other (<i>please specify in the space below</i>)					

7 Which tests are used as part of the school entry hearing screen in your area? How are these tests combined into a whole screen protocol? Please describe via words or flow diagram.

To indicate the required level of detail an example is shown in the box below. The information we require is shown in *italics* in the diagram and detailed in the following bullet pointed list –

- the tests used at each stage of the screen
- how much time passes between each test
- how many times the child is tested before referral takes place
- which service the child is referred on to



Your text or diagram –

8 Please indicate at what level a child will be said to have not passed each test you use in the screen.

	Frequency screened and level at which a child will not have passed
Test 1	
Test 2	
Test 3	

9 When screening children at school entry do you
(please tick one box for each category)

	All of the time	Most of the time	Some of the time	Rarely	Never
... screen for hearing loss only					
... screen for hearing loss as part of a wider health check					

10 Please estimate the minimum, average and maximum numbers of children that could be screened under normal circumstances during the course of a one day visit to a school in your area.

	When screening for hearing loss only	When screening for hearing loss as part of a wider health check
Minimum number		
Average number		
Maximum number		

Any Comments?

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.....

WHO PERFORMS THE TESTING?

These questions are designed to find out which staff are involved in the implementation of the School Entry Hearing Screen in your area.

11 Please indicate which staff perform the school entry hearing screen tests in your area. Please tick all that apply to your area.

- School nurse
- Audiometrician
- Audiologist
- Health Visitor
- School Doctor
- Other *(please specify in the space below)*

.....

Any Comments?

.....
.....
.....
.....

WHAT EQUIPMENT DO YOU USE?

12 Please indicate the types of equipment used within the school entry hearing screen as run in your area. Please then say, as accurately as possible, the number of each you have.

	Use?		If yes, how many are in use?
	No	Yes	
Screening audiometer			
Diagnostic audiometer			
Screening tympanometer			
Diagnostic tympanometer			
Other (please specify)			

Any Comments?

.....

.....

.....

.....

AUDIT DATA

In order for us to make evidence-based recommendations for the future of the School Entry Hearing Screen to the NHS we need to know the referral rates and yield of the screen. Your answers to these questions are therefore extremely important.

However, we do NOT expect you to undertake an exhaustive and time consuming note review process. Please answer the questions below with details of audit data ONLY if it can be easily obtained.

13 Has an audit of the School Entry Hearing Screen in your area been performed in the last two years AND can a copy of this audit be made available to us?

- No *please go to question 14*
- Yes, a hard copy of the audit has been included with this questionnaire *please go to question 18*
- Yes, a hard copy of the audit has been sent separately *please go to question 18*
- Yes, an electronic version of the audit has been emailed to Kirsty Bristow (kirsty.bristow@manchester.ac.uk) *please go to question 18*

14 Please indicate if the school entry hearing screen in your area employs a data management system.

- No *please go to question 17*
 - Yes, an IT system
 - Yes, paper system
 - Yes, other
If other, please specify below
-

15 Can you easily get data reports from this data management system?

- No *please go to question 17*
- Yes

16 Please indicate the following for the most recent academic year possible – (e.g. 2003–2004, 2002–2003 etc. Please indicate below which year)

For the academic year

	Number
How many children were eligible for the school entry hearing screen in your area?	
How many children were screened with the school entry hearing screen in your area?	
How many children were referred for further audiological assessment following failure of the school entry hearing screen in your area?	
How many children with sensorineural hearing loss were identified by the school entry hearing screen in your area?	
How many children with permanent conductive hearing loss were identified by the school entry hearing screen in your area?	
How many children with temporary conductive hearing loss were identified by the school entry hearing screen in your area?	
How many children with other types of hearing loss were identified by the school entry hearing screen in your area? <i>(please specify types of hearing loss)</i>	

17 Do you have any other documentation that you feel may be of use to us?

No

Yes

If yes, could a copy this documentation be made available to us?

Yes, a hard copy has been included with this completed questionnaire

Yes, a hard copy has been sent separately

Yes, an electronic version has been emailed to Kirsty Bristow (kirsty.bristow@manchester.ac.uk)

No

18 Is information regarding the costing of the School Entry Hearing Screen within the last two years routinely available in your district?

- No
- Yes

If yes, please provide the contact details of a member of staff that could provide us with this costing data in the space below

Name

Position

Address

.....

E-mail

Telephone

Any Comments?

.....
.....
.....

YOUR VIEWS

The questions so far may not have addressed all the issues you wish to raise. This section allows you to say what you think about the usefulness and future of the School Entry Hearing Screen.

19 Are there any plans for development or change of the school entry hearing screen in your area?

- No
- Yes

If yes, please give details below

.....

.....

.....

.....

20 Please cross one of the boxes below to indicate how useful overall you think the School Entry Hearing Screen is in your area as it is currently operated.

Not useful
at all

--	--	--	--	--	--	--	--	--	--

Very
useful

Please add further comments to explain your answer

.....

.....

.....

.....

21 Please add below any suggestions for the future of the School Entry Hearing Screen either in your area or nationally

.....

.....

.....

Any further comments?

.....
.....
.....
.....
.....

Thank you for taking the time to complete this questionnaire

We may need to contact you again to discuss some matters raised in your questionnaire. We will endeavour to do so only when strictly necessary and will keep any correspondence with you to an absolute minimum. Do we have your permission to contact you if required?

- No
- Yes

If yes could you please give your preferred form of contact below–

Telephone Number

OR

E-mail Address

.....
.....

OR

Post Address

.....
.....

Please return this completed questionnaire, and any other supporting documents you wish to include, in the pre-paid envelope supplied to–

Kirsty Bristow
Human Communication and Deafness
School of Psychological Sciences
Humanities Building (Devas)
University of Manchester
Oxford Road
Manchester
M13 9PL

Appendix 3

Prevalence rates for permanent childhood hearing impairment for three cohorts in Waltham Forest and Redbridge Districts and for one cohort in Trent Region of the UK

TABLE 69 Prevalence of PCHI by the end of the first year in primary school in cohort 1, NNHS: 31,538 children born from January 1977 to 1987 (n = 131)

Degree of hearing impairment		All PCHI n Prevalence/1000 (95% CI)	Congenital PCHI n Prevalence/1000 (95% CI)	Acquired PCHI n Prevalence/1000 (95% CI)
Bilateral (PTA average 500–4000 Hz) in BHE	Mild (20–39 dB HL)	37 1.17 (0.8 to 1.55)	32 1.01 (0.66 to 1.37)	5 0.16 (0.02 to 0.3)
	Moderate (40–69 dB HL)	24 0.76 (0.46 to 1.07)	18 0.57 (0.31 to 0.83)	6 0.19 (0.04 to 0.34)
	Severe (70–94 dB HL)	12 0.38 (0.17 to 0.6)	10 0.32 (0.12 to 0.51)	2 0.06 (0 to 0.15)
	Profound (≥95 dB HL)	12 0.38 (0.16 to 0.6)	12 0.38 (0.17 to 0.6)	0 0.00 (0.00 to 0.00)
	All (≥40 dB HL)	48 1.52 (1.09 to 1.95)	40 1.27 (0.88 to 1.66)	8 0.25 (0.08 to 0.43)
	All bilateral	85 2.69 (2.12 to 3.27)	72 2.28 (1.76 to 2.81)	13 0.41 (0.19 to 0.64)
Unilateral (PTA average 500–4000 Hz) in WHE	Mild/moderate (20–69 dB HL)	16 0.51 (0.26 to 0.76)	15 0.48 (0.23 to 0.72)	1 0.03 (0 to 0.09)
	Severe/profound (≥70 dB HL)	30 0.95 (0.61 to 1.29)	23 0.73 (0.43 to 1.03)	7 0.22 (0.06 to 0.39)
	All unilateral	46 1.46 (1.04 to 1.88)	38 1.21 (0.82 to 1.59)	8 0.25 (0.08 to 0.43)
Total all degrees		131 4.15 (3.44 to 4.86)	110 3.49 (2.84 to 4.14)	21 0.67 (0.38 to 0.95)

TABLE 70 Prevalence of PCHI by the end of the first year in primary school in cohort 2, TNHS: 32,980 children born from January 1990 to 2000 (n = 117)

Degree of hearing impairment		All PCHI n Prevalence/1000 (95% CI)	Congenital PCHI n Prevalence/1000 (95% CI)	Acquired PCHI n Prevalence/1000 (95% CI)
Bilateral (PTA average 500–4000 Hz) in BHE	Mild (20–39 dB HL)	40 1.22 (0.84 to 1.59)	29 0.88 (0.56 to 1.2)	11 0.33 (0.14 to 0.53)
	Moderate (40–69 dB HL)	24 0.73 (0.44 to 1.02)	19 0.58 (0.32 to 0.84)	5 0.15 (0.02 to 0.29)
	Severe (70–94 dB HL)	14 0.43 (0.2 to 0.65)	12 0.36 (0.16 to 0.57)	2 0.06 (0 to 0.15)
	Profound (≥95 dB HL)	11 0.33 (0.14 to 0.53)	11 0.33 (0.14 to 0.53)	0 0.0 (0.00 to 0.00)
	All (≥40 dB HL)	49 1.49 (1.07 to 1.91)	42 1.28 (0.89 to 1.66)	7 0.21 (0.06 to 0.37)
	All bilateral	89 2.71 (2.14 to 3.27)	71 2.16 (1.66 to 2.66)	18 0.55 (0.29 to 0.8)
	Unilateral (PTA average 500–4000 Hz) in WHE	Mild/moderate (20–69 dB HL)	13 0.39 (0.18 to 0.61)	10 0.30 (0.12 to 0.49)
Severe/profound (≥70 dB HL)		15 0.46 (0.23 to 0.69)	12 0.36 (0.16 to 0.57)	3 0.09 (0 to 0.19)
All unilateral		28 0.85 (0.54 to 1.17)	22 0.67 (0.39 to 0.95)	6 0.18 (0.04 to 0.33)
Total all degrees		117 3.56 (2.91 to 4.2)	93 2.83 (2.25 to 3.4)	24 0.73 (0.44 to 1.02)

TABLE 71 Prevalence of PCHI by the end of the first year in primary school in cohort 3, UNHS: 29,132 children born from January 1992 to 2000 (n = 101)

Degree of hearing impairment		All PCHI n Prevalence/1000 (95% CI)	Congenital PCHI n Prevalence/1000 (95% CI)	Acquired PCHI n Prevalence/1000 (95% CI)
Bilateral (PTA average 500–4000 Hz) in BHE	Mild (20–39 dB HL)	36 1.24 (0.83 to 1.64)	28 0.96 (0.61 to 1.32)	8 0.27 (0.08 to 0.46)
	Moderate (40–69 dB HL)	23 0.79 (0.47 to 1.11)	19 0.65 (0.36 to 0.95)	4 0.14 (0 to 0.27)
	Severe (70–94 dB HL)	9 0.31 (0.11 to 0.51)	9 0.31 (0.11 to 0.51)	0 0.00 (0.00 to 0.00)
	Profound (≥95 dB HL)	10 0.34 (0.13 to 0.56)	8 0.27 (0.08 to 0.46)	2 0.07 (0 to 0.16)
	All (≥40 dBHL)	42 1.44 (1.01 to 1.88)	36 1.24 (0.83 to 1.64)	6 0.21 (0.04 to 0.37)
	All bilateral	78 2.68 (2.08 to 3.27)	64 2.20 (1.63 to 2.7)	14 0.48 (0.25 to 0.78)
	Unilateral (PTA average 500–4000 Hz) in WHE	Mild/moderate (20–69 dB HL)	15 0.51 (0.25 to 0.78)	12 0.41 (0.18 to 0.64)
Severe/profound (≥70 dB HL)		8 0.27 (0.08 to 0.46)	7 0.24 (0.06 to 0.42)	1 0.03 (0 to 0.1)
All unilateral		23 0.79 (0.47 to 1.11)	19 0.65 (0.36 to 0.95)	4 0.14 (0 to 0.27)
Total all degrees		101 3.47 (2.79 to 4.14)	83 2.85 (2.21 to 3.42)	18 0.62 (0.36 to 0.95)

TABLE 72 Prevalence of PCHI for the three cohorts combined and for the Trent cohort, and an analysis of the difference in the prevalence rates between the three cohorts and between the combined cohort and the Trent cohort

Degree of hearing impairment		Combined cohort Prevalence/1000 (95% CI)	Comparison of prevalence in three cohorts Pearson χ^2 (p-value)	Trent cohort Prevalence/1000 (95% CI)	Comparison of prevalence in Trent and combined cohorts Pearson χ^2 (p-value)
Bilateral (PTA average 500–4000 Hz) in BHE	Mild (20–39 dB HL)	1.21 (0.99 to 1.43)	0.052 (0.974)	Not available	
	Moderate (40–69 dB HL)	0.76 (0.58 to 0.94)	0.073 (0.964)	0.68 (0.61 to 0.78)	0.583 (0.445)
	Severe (70–94 dB HL)	0.37 (0.25 to 0.5)	0.568 (0.753)	0.28 (0.23 to 0.34)	2.151 (0.142)
	Profound (\geq 95 dB HL)	0.35 (0.23 to 0.47)	0.108 (0.948)	0.31 (0.26 to 0.37)	0.463 (0.496)
	All (\geq 40 dB HL)	1.49 (1.24 to 1.73)	0.060 (0.967)	1.27 (1.16 to 1.39)	2.532 (0.112)
	All bilateral	2.69 (2.36 to 3.03)	0.005 (0.998)	Not available	
Unilateral (PTA average 500–4000 Hz) in WHE	Mild/moderate (20–69 dB HL)	0.47 (0.33 to 0.61)	0.609 (0.737)	Not available	
	Severe/profound (\geq 70 dB HL)	0.57 (0.41 to 0.72)	13.338 (0.001)***	Not available	
	All unilateral	1.04 (0.83 to 1.24)	8.229 (0.016)*	Not available	

*** $p < 0.001$, * $p < 0.05$.

Appendix 4

Search strategies used in the systematic review

Ovid MEDLINE(R) 1966 to April week 4 2005

- 1 hearing loss\$.mp. or exp Hearing Loss/
- 2 (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp.
- 3 exp Hearing Disorders/
- 4 or/1-3
- 5 exp Child, Preschool/ or school entry.mp.
- 6 exp Child Development/
- 7 early detect\$.mp.
- 8 infant school\$.mp.
- 9 exp Schools, Nursery/ or exp Nurseries/ or exp Child Day Care Centers/ or kindergarten\$.mp.
- 10 nursery school\$.mp.
- 11 or/5-10
- 12 screen\$.mp. or exp Mass Screening/
- 13 4 and 12
- 14 13 and 11
- 15 (school entry adj3 (screen\$ or exam\$)).mp.
- 16 (medical exam\$ adj2 school\$).mp.
- 17 or/15-16
- 18 14 or 17
- 19 randomized controlled trial.pt.
- 20 controlled clinical trial.pt.
- 21 randomized controlled trials.sh.
- 22 random allocation.sh.
- 23 double blind method.sh.
- 24 single-blind method.sh.
- 25 or/19-24
- 26 (animals not human).sh.
- 27 25 not 26
- 28 clinical trial.pt.
- 29 exp clinical trials/
- 30 (clin\$ adj25 trial\$).ti,ab.
- 31 ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj25 (blind\$ or mask\$)).ti,ab.
- 32 placebos.sh.
- 33 placebo\$.ti,ab.
- 34 random\$.ti,ab.
- 35 research design.sh.
- 36 or/28-35
- 37 36 not 26
- 38 37 not 27
- 39 comparative study.sh.
- 40 exp evaluation studies/
- 41 follow up studies.sh.
- 42 prospective studies.sh.
- 43 (control\$ or prospectiv\$ or volunteer\$).ti,ab.
- 44 or/39-43
- 45 44 not 26

- 46 45 not (27 or 38)
- 47 27 or 38 or 46
- 48 18 and 47

Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations 6 June 2005

- 1 hearing loss\$.mp. or exp Hearing Loss/
- 2 (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp.
- 3 exp Child, Preschool/ or school entry.mp.
- 4 early detect\$.mp.
- 5 infant school\$.mp.
- 6 exp Schools, Nursery/ or exp Nurseries/ or exp Child Day Care Centers/ or kindergarten\$.mp.
- 7 nursery school\$.mp.
- 8 screen\$.mp. or exp Mass Screening/
- 9 (school entry adj3 (screen\$ or exam\$)).mp.
- 10 (medical exam\$ adj2 school\$).mp.
- 11 randomized controlled trial.pt.
- 12 controlled clinical trial.pt.
- 13 clinical trial.pt.
- 14 (clin\$ adj25 trial\$).ti,ab.
- 15 ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj25 (blind\$ or mask\$)).ti,ab.
- 16 placebo\$.ti,ab.
- 17 random\$.ti,ab.
- 18 (control\$ or prospectiv\$ or volunteer\$).ti,ab.
- 19 or/1-2
- 20 or/3-7
- 21 or/8-10
- 22 or/11-18
- 23 19 and 20
- 24 21 and 23
- 25 22 and 24

EMBASE 1980 to 2005 week 19

- 1 hearing loss\$.mp. or exp Hearing Loss/
- 2 (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp.
- 3 exp Hearing Disorder/
- 4 or/1-3
- 5 school entry.mp.
- 6 (pre adj school).mp.
- 7 (nursery adj school\$).mp.
- 8 exp nursery school/
- 9 kindergarten\$.mp. or exp kindergarten/
- 10 exp Day Care/
- 11 infant school\$.mp.

- 12 early detect\$.mp.
- 13 exp Child Development/
- 14 or/5-13
- 15 screen\$.mp.
- 16 exp mass screening/ or exp screening/ or exp auditory screening/ or exp screening test/
- 17 or/15-16
- 18 (school entry adj3 (screen\$ or exam\$)).mp.
- 19 (medical exam\$ adj2 school\$).mp.
- 20 or/18-19
- 21 4 and 17
- 22 21 and 14
- 23 22 or 20
- 24 randomized controlled trial/
- 25 exp clinical trial/
- 26 exp controlled study/
- 27 double blind procedure/
- 28 randomization/
- 29 placebo/
- 30 single blind procedure/
- 31 (control\$ adj (trial\$ or stud\$ or evaluation\$ or experiment\$)).mp.
- 32 ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj5 (blind\$ or mask\$)).mp.
- 33 (placebo\$ or matched communities or matched schools or matched populations).mp.
- 34 (comparison group\$ or control group\$).mp.
- 35 (clinical trial\$ or random\$).mp.
- 36 (quasiexperimental or quasi experimental or pseudo experimental).mp.
- 37 matched pairs.mp.
- 38 or/24-37
- 39 23 and 38

CINAHL – Cumulative Index to Nursing & Allied Health Literature 1982 to May week 1 2005

- 1 hearing loss\$.mp. or exp Hearing Loss/
- 2 (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp.
- 3 exp Hearing Disorder/
- 4 or/1-3
- 5 school entry.mp.
- 6 (pre adj school).mp.
- 7 (nursery adj school\$).mp.
- 8 exp nursery school/
- 9 kindergarten\$.mp. or exp kindergarten/
- 10 exp day care/
- 11 infant school\$.mp.
- 12 early detect\$.mp.
- 13 exp child development/
- 14 or/5-13
- 15 (school entry adj3 (screen\$ or exam\$)).mp.
- 16 (medical exam\$ adj2 school\$).mp.
- 17 or/15-16
- 18 exp health screening/ or screen\$.mp.
- 19 exp hearing screening/

- 20 or/18-19
- 21 4 and 18
- 22 21 or 19
- 23 22 and 14
- 24 23 or 17

Cochrane Library (Wiley) 2005 Issue 2

- #1 exp Hearing Loss/
- #2 Hearing next loss
- #3 hearing next disorder*
- #4 hearing next difficult*
- #5 hearing next problem*
- #6 hearing next impair*
- #7 exp Hearing disorders/
- #8 #1 or #2 or #3 or #4 or #5 or #6 or #7
- #9 screening
- #10 exp Mass screening/
- #11 #9 or #10
- #12 school next entr*
- #13 early detect*
- #14 infant next school*
- #15 nursery next school*
- #16 kindergarten*
- #17 exp Schools, Nursery/
- #18 exp Child day care centers/
- #19 exp Child, Pre School
- #20 exp Child Development
- #21 #12 or #13 or #14 or #15 or #16 or #17 or #18 or #19 or #20
- #22 #8 and #11
- #23 #21 and #22
- #24 school next entr*
- #25 medical near/3 exam* near/3 school*
- #26 #24 or #25
- #27 #23 or #26

ERIC (Cambridge Scientific Abstracts) Searched 7 June 2005

- #1 school entry or preschool
- #2 screen* or test*
- #3 hearing loss or hearing impair*
- #4 #1 and #2 and #3

Science Citation Index (Web of Knowledge) Searched 7 June 2005

- #1 school entry or preschool
- #2 screen* or test*
- #3 hearing loss or hearing impair*
- #4 #1 and #2 and #3

PsycINFO 1985 to May week 1 2005

- 1 (hearing adj (disorder\$ or impair\$ or problem\$ or difficul\$)).mp.
- 2 exp Hearing Disorders/ or hearing loss.mp.

- 3 or/1-2
- 4 exp Screening Tests/ or screen\$.mp.
- 5 1 and 4
- 6 Exp CHILD DAY CARE/
(School Adj Entry).Mp
- 8 (Pre Adj School).Mp.
- 9 Nurser\$.Mp.
- 10 Exp Kindergarten Students/ Or Exp
KINDERGARTENS/ Or Kindergarten\$.Mp.
- 11 Exp Preschool Students/ Or Exp Preschool
Education/ Or Exp Nursery School Students/
Or Infant School\$.Mp.
- 12 Early Detect\$.Mp.
- 13 (School Entry Adj3 (Screen\$ Or Exam\$)).Mp.
- 14 Child\$.Mp.
- 15 Or/6-14
- 16 5 And 15
- 17 (School Entry Adj3 (Screen\$ Or Exam\$)).Mp.
- 18 (Medical Exam\$ Adj2 School\$).Mp.
- 19 Or/17-18
- 20 16 Or 19

Search strategies: accuracy of diagnostic tests

Ovid MEDLINE(R)

1966 to January week 4 2006

- 1 audiometry.mp. or audiometry/ or exp
audiometry, pure-tone/
- 2 exp otoacoustic emissions, spontaneous/ or
otoacoustic emission\$.mp.
- 3 exp acoustic impedance tests/ or acoustic
impedance.mp.
- 4 exp hearing tests/is, mt [instrumentation,
methods]
- 5 hearing test\$.mp.
- 6 sweep audio.mp.
- 7 sweep test\$.mp.
- 8 (hearing adj2 questionnaire\$).mp.
- 9 cmedhq.mp.
- 10 conventional audiometry.mp.
- 11 conditioned play audiometry.mp.
- 12 cpa.mp.
- 13 exp audiometry, evoked response/
- 14 audiologic\$ assessment\$.mp.
- 15 acoustic intermittance.tw.
- 16 tympanometry.mp.
- 17 otoscopy.mp. or exp otoscopy/ or exp
diagnostic techniques, otological/
- 18 otological exam\$.mp.
- 19 acoustic reflex test\$.mp.
- 20 teoae.mp.
- 21 dpoeae.mp.
- 22 (impedance adj screening).mp.
- 23 (impedance adj method\$).mp.
- 24 fixed frequency audio.mp.

- 25 (speech adj2 noise).mp.
- 26 reflectometry.mp.
- 27 acoustic impedance.mp.
- 28 or/1-27
- 29 exp hearing loss, sensorineural/pc, di
- 30 exp hearing disorders/di, pc
- 31 exp otitis media/pc, di
- 32 exp hearing loss, high-frequency/pc, di
- 33 exp hearing loss/di, pc
- 34 hearing impairment\$.mp.
- 35 exp hearing loss, conductive/di, pc
- 36 (hearing adj3 screen\$).mp.
- 37 exp "sensitivity and specificity"/
- 38 exp predictive value of tests/
- 39 (diagnos\$ adj2 accura\$).mp.
- 40 or/37-39
- 41 exp child, preschool/ or school entry.mp.
- 42 exp child development/
- 43 early detect\$.mp.
- 44 infant school\$.mp.
- 45 exp schools, nursery/ or exp nurseries/ or exp
child day care centers/ or kindergarten\$.mp.
- 46 nursery school\$.mp.
- 47 or/41-46
- 48 screen\$.mp. or exp mass screening/
- 49 (school entry adj3 (screen\$ or exam\$)).mp.
- 50 (medical exam\$ adj2 school\$).mp.
- 51 or/49-50
- 52 28 and 40
- 53 47 or 51
- 54 or/29-36
- 55 52 and 53
- 56 40 and 54
- 57 53 and 56
- 58 55 or 57

EMBASE

1980 to 2006 week 11

- 1 exp pure tone audiometry/ or exp audiometry/
or audiometry.mp.
- 2 otoacoustic emission\$.mp. or exp spontaneous
otoacoustic emission/ or exp otoacoustic
emission/
- 3 acoustic impedance.mp. or exp acoustic
impedance/
- 4 hearing test\$.mp.
- 5 exp hearing test/
- 6 sweep audio.mp.
- 7 sweep test\$.mp.
- 8 (hearing adj2 questionnaire\$).mp.
- 9 cmedhq.mp.
- 10 ((conventional or conditioned play) adj
audiometry).mp.
- 11 cpa.mp.
- 12 exp evoked response audiometry/
- 13 (audiologic\$ adj assessment\$).mp.
- 14 (acoustic adj intermittance).mp.

- 15 tympanometry.mp. or exp tympanometry/
- 16 otoscopy.mp. or exp otoscopy/
- 17 (otological adj2 technique\$.mp.
- 18 (otological adj2 (exam\$ or technique\$)).mp.
- 19 teoae.mp.
- 20 dpoae.mp. or exp distortion product
otoacoustic emission/
- 21 (impedance adj (screen\$ or method\$)).mp.
- 22 fixed frequency audio.mp.
- 23 (speech adj2 noise).mp.
- 24 reflectometry.mp. or exp reflectometry/
- 25 or/1-24
- 26 hearing loss/pc, di [prevention, diagnosis]
- 27 hearing disorder/pc, di [prevention, diagnosis]
- 28 otitis media/pc, di [prevention, diagnosis]
- 29 hearing impair\$.mp.
- 30 hearing impairment/pc, di [prevention,
diagnosis]
- 31 (hearing adj3 screen\$.mp.
- 32 exp "sensitivity and specificity"/
- 33 (predictive adj2 test\$.mp.
- 34 (diagnos\$ adj2 accura\$.mp.
- 35 or/32-34
- 36 or/26-31
- 37 25 and 35
- 38 school entry.mp.
- 39 pre-school.mp.
- 40 child/
- 41 exp child development/
- 42 early detect\$.mp.
- 43 infant school\$.mp.
- 44 nursery school\$.mp. or exp nursery school/
- 45 exp child care/ or child day care.mp.
- 46 kindergarten\$.mp. or exp kindergarten/
- 47 or/38-46
- 48 screen\$.mp.
- 49 exp mass screening/
- 50 (school entry adj3 (screen\$ or exam\$)).mp.
- 51 (medical exam\$ adj2 school\$.mp.
- 52 or/48-49
- 53 or/50-51
- 54 25 and 35 and 47
- 55 35 and 36 and 47
- 56 54 or 55
- 57 or/52-53
- 58 47 or 57
- 59 35 and 36 and 58
- 60 56 or 59

**CINAHL
1982 to March week 3 2006**

- 1 audiometry.mp. or exp audiometry, evoked
response/ or exp audiometry/ or exp
audiometry, pure-tone/
- 2 otoacoustic emission\$.mp. or exp otoacoustic
emissions, spontaneous/

- 3 exp acoustic impedance tests/ or acoustic
impedance.mp.
- 4 hearing test\$.mp. or exp hearing tests/
- 5 sweep test\$.mp.
- 6 sweep audio.mp.
- 7 (hearing adj2 questionnaire\$.mp.
- 8 cmedhq.mp.
- 9 (conventional adj2 audiometry).mp.
- 10 (conditioned adj2 audiometry).mp.
- 11 cpa.mp.
- 12 evoked response.mp. or exp evoked
potentials/
- 13 (audiologic\$ adj assessment\$.mp.
- 14 (acoustic adj intermittance).mp.
- 15 tympanometry.mp.
- 16 otoscopy.mp.
- 17 (otological adj2 technique\$.mp.
- 18 (otological adj2 exam\$.mp.
- 19 teoae.mp.
- 20 dpoae.mp. or exp otoacoustic emissions,
evoked/
- 21 (impedance adj (screen\$ or method\$)).mp.
- 22 fixed frequency audio.mp.
- 23 (speech adj2 noise).mp.
- 24 reflectometry.mp.
- 25 or/1-24
- 26 hearing disorders/di, pc
- 27 hearing impair\$.mp.
- 28 exp hearing screening/
- 29 (hear\$ adj2 screen\$.mp.
- 31 exp "sensitivity and specificity"/
- 32 exp "predictive value of tests"/
- 33 (predictive adj2 test\$.mp.
- 34 (diagnos\$ adj2 accura\$.mp.
- 35 or/31-34
- 36 school entry.mp.
- 37 exp child, preschool/ or pre-school.mp.
- 38 exp child development/
- 39 early detect\$.mp.
- 40 infant school\$.mp. or exp infant development/
- 41 nursery school\$.mp. or exp schools, nursery/
- 42 child day care.mp. or exp child day care/
- 43 kindergarten\$.mp.
- 44 or/36-43
- 45 screen\$.mp.
- 46 exp hearing screening/
- 47 exp school admissions/
- 48 (school entry adj2 (screen\$ or exam\$)).mp.
- 49 (medical exam\$ adj2 school\$.mp.
- 51 or/47-49
- 52 25 and 35 and 44
- 53 30 and 35 and 44
- 54 52 or 53
- 55 50 or 51 or 44
- 56 30 and 35 and 55
- 57 54 or 56

PSYCINFO**1967 to March week 4 2006**

- 1 exp bone conduction audiometry/ or exp audiometry/ or audiometry.mp.
- 2 otoacoustic emission\$.mp.
- 3 acoustic impedance.mp.
- 4 hearing test\$.mp.
- 5 sweep audio.mp.
- 6 sweep test\$.mp.
- 7 (hearing adj2 questionnaire\$.mp.
- 8 cmedhq.tw.
- 9 cpa.mp.
- 10 evoked response audiometry.mp.
- 11 audiologic\$ assessment\$.mp.
- 12 acoustic intermittance.tw.
- 13 tympanometry.mp.
- 14 otoscopy.mp.
- 15 (otological adj2 diagnos\$.mp.
- 16 otological exam\$.mp.
- 17 acoustic reflex test\$.mp.
- 18 teoae.mp.
- 19 dpoae.mp.
- 20 (impedance adj screening).mp.
- 21 (impedance adj method\$.mp.
- 22 fixed frequency audio.mp.
- 23 (speech adj2 noise).mp.
- 24 reflectometry.mp.
- 25 acoustic impedance.mp.
- 26 or/1-25
- 27 (sensitivity adj2 specificity).mp.
- 28 (predictive value adj2 test\$.mp.
- 29 (diagnos\$ adj2 accurac\$.mp.
- 30 or/27-29
- 31 ((hearing loss\$ or hearing disorder\$ or hearing impair\$ or otitis media) adj3 (diagnos\$ or screen\$)).mp.

- 32 30 or 31
- 33 26 and 32
- 34 child\$.mp. or exp child day care/
- 35 exp early childhood development/ or exp preschool education/ or exp preschool students/ or pre-school.mp. or exp nursery schools/
- 36 kindergarten\$.mp. or exp kindergartens/
- 37 nursery school\$.mp.
- 38 exp elementary school students/ or infant school\$.mp.
- 39 exp early intervention/ or early detect\$.mp.
- 40 or/34-39
- 41 33 and 40

**ERIC (CSA)
1966 to present**

Search date: 28 March 2006

(hearing or otitis) and (diagnos* or screen* or test*) and (school*or nurser* or infant*) and (accur* or predictive or sensitiv*)

**Science Citation Index (Web Of Knowledge)
1970 to present**

Search date: 28 March 2006

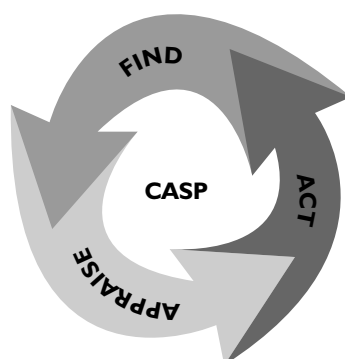
(Hearing or otitis or deaf*) and (screen* or test* or diagnos*) and (accura* or predictive or sensitive) and (pre-school or infant* or nurser* or kindergarten*)

Appendix 5

Quality criteria for systematic reviews

CRITICAL APPRAISAL SKILLS PROGRAMME

Making sense of evidence about clinical effectiveness



10 questions to help you make sense of a review

These questions consider the following:

Are the results of the review valid? (SECTION A)

What are the results? (SECTION B)

Will the results help locally? (SECTION C)

A number of italicised prompts are given after each question. These are designed to remind you why the question is important. There will not be time in the small groups to answer them all in detail!

These materials were developed by the Critical Appraisal Skills Programme (CASP) and we thank them for permission to use the materials.

A/ Are the results of the review valid?

Screening Questions

<p>1. Did the review address a clearly focused question?</p> <p><i>HINT: An issue can be 'focused' in terms of</i></p> <ul style="list-style-type: none"> • the population studied • the intervention given • the outcome considered 	<p>Yes</p> <p><input type="checkbox"/></p>	<p>Can't tell</p> <p><input type="checkbox"/></p>	<p>No</p> <p><input type="checkbox"/></p>
<p>2. Did the authors look for the appropriate sort of papers?</p> <p><i>HINT: The 'best sort of studies' would</i></p> <ul style="list-style-type: none"> – address the review's question – have an appropriate study design (usually RCTs for papers evaluating interventions) 	<p>Yes</p> <p><input type="checkbox"/></p>	<p>Can't tell</p> <p><input type="checkbox"/></p>	<p>No</p> <p><input type="checkbox"/></p>

Is it worth continuing?

Detailed Questions

<p>3. Do you think the important, relevant studies were included?</p> <p><i>HINT: Look for</i></p> <ul style="list-style-type: none"> – which bibliographic databases were used – follow-up from reference lists – personal contact with experts – search for unpublished as well as published studies – search for non-English language studies 	<p>Yes</p> <p><input type="checkbox"/></p>	<p>Can't tell</p> <p><input type="checkbox"/></p>	<p>No</p> <p><input type="checkbox"/></p>
<p>4. Did the review's authors do enough to assess the quality of the included studies?</p> <p><i>HINT: The authors need to consider the rigour of the studies they have identified. Lack of rigour may affect the studies' results ('All that glisters is not gold' Shakespeare, the Merchant of Venice, Act II)</i></p>	<p>Yes</p> <p><input type="checkbox"/></p>	<p>Can't tell</p> <p><input type="checkbox"/></p>	<p>No</p> <p><input type="checkbox"/></p>
<p>5. If the results of the review have been combined, was it reasonable to do so?</p> <p><i>HINT: Consider whether</i></p> <ul style="list-style-type: none"> – the results were similar from study to study – the results of all the included studies are clearly displayed – the results of the different studies are similar – the reasons for any variations in results are discussed 	<p>Yes</p> <p><input type="checkbox"/></p>	<p>Can't tell</p> <p><input type="checkbox"/></p>	<p>No</p> <p><input type="checkbox"/></p>

B/ What are the results?

<p>6. What are the overall results of the reviews?</p> <p><i>HINT: Consider</i></p> <ul style="list-style-type: none"> – if you are clear about the review's 'bottom line' results: – what these are (numerically if appropriate) – how were the results expressed (NNT, odds ratio etc.) 	
<p>7. How precise are the results?</p> <p><i>HINT: Look at the confidence intervals, if given</i></p>	

C/ Will the results help locally?

<p>8. Can the results be applied to the local population?</p> <p><i>HINT: Consider whether</i></p> <ul style="list-style-type: none"> – the patients covered by the review could be sufficiently different to your population to cause concern – your local setting is likely to differ much from that of the review 	<p>Yes <input type="checkbox"/></p> <p>Can't tell <input type="checkbox"/></p> <p>No <input type="checkbox"/></p>
<p>9. Were all important outcomes considered?</p>	<p>Yes <input type="checkbox"/></p> <p>Can't tell <input type="checkbox"/></p> <p>No <input type="checkbox"/></p>
<p>10. Are the benefits worth the harms and costs?</p> <p>Even if this is not addressed by the review, what do you think?</p>	

Appendix 6

Quality criteria for diagnostic test studies

Quality assessment form for studies looking at diagnostic accuracy (Taken from the QUADAS checklist)

Title:

First Author:

Date:

<p>Question 1</p> <p>Was the spectrum of patients representative of the patients who will receive the test in practice?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 2</p> <p>Were the selection criteria clearly described?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 3</p> <p>Is the reference standard likely to correctly classify the target condition?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>

Question 4 Is the time period between reference standard and index test short enough to be reasonable?	Yes No Unclear <i>Comments</i>
--	--

Question 5 Did the whole sample or a random selection of the sample receive verification using a reference standard?	Yes No Unclear <i>Comments</i>
--	--

Question 6 Did patients receive the same regardless of the index test result?	Yes No Unclear <i>Comments</i>
---	--

Question 7 Was the reference standard independent of the index test? (i.e. the index test did not form part of the reference standard)	Yes No Unclear <i>Comments</i>
--	--

Question 8 Was the execution of the index test described in sufficient detail to permit replication of the test?	Yes No Unclear <i>Comments</i>
--	--

<p>Question 9</p> <p>Was the execution of the reference standard described in sufficient detail to permit its replication?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 10</p> <p>Were the index test results interpreted without knowledge of the results of the reference standard?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 11</p> <p>Were the reference standard results interpreted without knowledge of the results of the index test?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 12</p> <p>Were the same clinical data available when test results were interpreted as would be available when the test is used in practice?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
<p>Question 13</p> <p>Were uninterpretable/intermediate test results reported?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>

<p>Question 14</p> <p>Were withdrawals from the study explained?</p>	<p>Yes</p> <p>No</p> <p>Unclear</p> <p><i>Comments</i></p>
---	---

Appendix 7

Excluded studies and reasons for exclusions

- Aguero AL, Borria JJ, de Mola M, Asnaghi P, Cansler A, Edelstein S, *et al.* The audiometric evaluation of Buenos Aires schoolchildren. *Boletín de la Oficina Sanitaria Panamericana* 1995;**119**:292–8. [Non-comparative study.]
- Al Allaf AMY, Ali A, Muneer M. Deafness in children and the need for cochlear implants. [Arabic]. *Journal of the Bahrain Medical Society* 2003;**15**:219–22. [Non-comparative study.]
- Arslan E, Turrini M, Lupi G, Genovese E, Orzan E. Hearing threshold assessment with auditory brainstem response (ABR) and ElectroCochleoGraphy (ECochG) in uncooperative children. *Scand Audiol Suppl* 1997;**46**:32–7. [Looking at uncooperative children, general anaesthetic was applied.]
- Augustsson I, Nilson C, Engstrand I. The preventive value of audiometric screening of preschool and young school-children. *Int J Pediatr Otorhinolaryngol* 1990;**20**:51–62. [Non-comparative study.]
- Babb MJ, Hilsinger RL Jr, Korol HW, Wilcox RD. Modern acoustic reflectometry: accuracy in diagnosing otitis media with effusion. *Ear Nose Throat J* 2004;**83**:622–4. [Not looking at screening.]
- Bamford J, Davis A, Boyle J, Law J, Chapman S, Brown SS, *et al.* Preschool hearing, speech, language, and vision screening. *Qual Health Care* 1998;**7**:240–7. [Audit.]
- Baumann U, Schorn K. Early detection of pediatric hearing loss. Visual and automated procedures compared. *HNO* 2001;**49**:118–25. [Children too young.]
- Beppu R, Hattori T, Yanagita N. Comparison of TEOAE with Play audiometry for screening hearing problems in children. *Auris Nasus Larynx* 1997;**24**:367–71. [Children too young.]
- Brunner M, Pfeiffer B, Heinrich C, Proschel U. Evaluation of the new Heidelberg preschool screening for auditory perception and language processing. *Folia Phoniatr Logop* 2005;**57**:48–58. [Not looking at screening.]
- Cadman D, Walter SD, Chambers LW, Ferguson R, Szatmari P, Johnson N, *et al.* Predicting problems in school performance from preschool health, developmental and behavioural assessments. *CMAJ* 1988;**139**:31–6. [Not looking at hearing.]
- Cadman D, Chambers LW, Walter SD, Feldman W, Smith K, Ferguson R. The usefulness of the Denver Developmental Screening Test to predict kindergarten problems in a general community population. *Am J Public Health* 1984;**74**:1093–7. [Not looking at hearing.]
- Combs JT. Predictive value of the angle of acoustic reflectometry. *Pediatr Infect Dis J* 1991;**10**:214–16. [Not looking at screening/hearing.]
- Combs JT. Single vs. double acoustic reflectometry tracings. *Pediatr Infect Dis J* 1989;**8**:616–20. [Not looking at screening/hearing.]
- Dancer J, Burl NT, Waters S. Effects of unilateral hearing loss on teacher responses to the SIFTER. Screening Instrument for Targeting Educational Risk. *Am Ann Deaf* 1995;**140**:291–4. [Some of the subjects had previously diagnosed hearing problems.]
- Dempster JH, Mackenzie K. Clinical role of free-field voice tests in children. *Clin Otolaryngol Allied Sci* 1992;**17**:54–6. [Looking at children who had previously been referred to a hearing clinic.]
- Dempster JH, Mackenzie K. Tympanometry in the detection of hearing impairments associated with otitis media with effusion. *Clin Otolaryngol Allied Sci* 1991;**16**:157–9. [Children had already been referred.]
- Douniadakis DE, Nikolopoulos TP, Tsakanikos MD, Vassiliadis SV, Apostolopoulos NJ. Evaluation of acoustic reflectometry in detecting otitis media in children. *Br J Audiol* 1993;**27**:409–14. [Children had already been referred for hearing problems.]
- Driscoll C, Kei J, McPherson B. Hearing screening for children in community settings using transient evoked otoacoustic emissions. *Asia Pac J Speech Lang Hear* 2003;**8**:179–84. [Screening is not comparative.]
- Driscoll C, Kei J, McPherson B. Transient evoked otoacoustic emissions in 6-year-old school children: a normative study. *Scand Audiol Suppl* 2000;**29**:103–10. [Looking at disabled children.]
- Elliott M, Jones JC, Jones R, Pritchard VG, Robinson BE. An inter-district audit of the school entry medical examination in Cheshire. *Public Health* 1994;**108**:203–10. [Not a study, just an audit of medical examinations.]
- Emmer MB, Silman S. The prediction of hearing loss in persons with cerebral palsy using contralateral acoustic reflex threshold for broad-band noise. *Am J Audiol* 2003;**12**:91–5. [Children studied had cerebral palsy.]
- Finitzo T, Friel-Patti S, Chinn K, Brown O. Tympanometry and otoscopy prior to myringotomy: issues in diagnosis of otitis media. *Int J Pediatr Otorhinolaryngol* 1992;**24**:101–10. [Children had recurring otitis media.]
- Flanary VA, Flanary CJ, Colombo J, Kloss D. Mass hearing screening in kindergarten students. *Int J Pediatr Otorhinolaryngol* 1999;**50**:93–8. [Non-comparative study.]

- Garrubba V, Grandori F, Lamoretti M, Nicolai P, Zanetti D, Antonelli AR. Electric response audiometry in infants and preschool children. Long-term control of the results. *Acta Otolaryngol Suppl* 1991;**482**:36–43. [Children all under 4.]
- Gershel J, Kruger B, Giraudi-Perry D, Chobot J, Rosenberg M, Shapiro IM, *et al.* Accuracy of the Welch Allyn AudioScope and traditional hearing screening for children with known hearing loss. *J Pediatr* 1985;**106**:15–20. [Some children had previously diagnosed hearing problems.]
- Gray S, Yamauchi T. Preschool screening of speech, language and hearing – model program of early identification and intervention. *Clin Res* 1976;**24**:A175. [Non-comparative study.]
- Guo Y, Yao D. The application of otoacoustic emissions in paediatric hearing screening. *Chung-Kuo i Hsueh Ko Hsueh Yuan Hsueh Pao Acta Academiae Medicinae Sinicae* 1996;**18**:284–7 [Used high-risk infants only.]
- Haggard MP, Wood EJ, Carroll S. Speech, admittance and tone tests in school screening. Reconciling economics with pathology and disability perspectives. *Br J Audiol* 1984;**18**:133–53. [Outcomes not relevant.]
- Heath RW, *et al.* Hearing dysfunction in Hawaiian preschoolers: its relation to educational achievement and family characteristics. Internal report, January 1987. [Paper could not be retrieved.]
- Herer GR, Glatke TJ, Rafitis IA, Cummiskey C. Detection of hearing loss in young children and adults using otoacoustic emissions. *Folia Phoniatr Logop* 1996;**48**:117–21 [Non-comparative study.]
- Holmes AE, Jones-Muir KC, Kember FJ. Acoustic reflectometry versus tympanometry in pediatric middle ear screenings. *Lang Speech Hear Serv Sch* 1989;**20**:41–9. [Some children had previously diagnosed hearing problems.]
- Holtby I, Forster DP. Evaluation of pure tone audiometry and impedance screening in infant schoolchildren. *J Epidemiol Commun Health* 1992;**46**:21–5. [Children too old.]
- Jones J, Batchelor L, Gordon N, West M. The preschool medical: an evaluation of this examination and its role in child health surveillance. *Child Care Health Dev* 1989;**15**:425–34. [Non-comparative study.]
- Kaleida PH, Stool SE. Assessment of otoscopists' accuracy regarding middle-ear effusion. Oscopic validation. *Am J Dis Child* 1992;**146**:433–5. [Physician validation.]
- Kanasaku M, Suzuki S, Notoya M, *et al.* The screening level of pure tone audiometry for young children. *Audiol Jpn* 1977;**20**:702–8. [Non-comparative study.]
- Karzon RG. Validity and reliability of tympanometric measures for pediatric patients. *J Speech Hear Res* 1991;**34**:386–90. [Children had already been referred for hearing problems.]
- Kazanas SG, Maw AR. Tympanometry, stapedius reflex and hearing impairment in children with otitis media with effusion. *Acta Otolaryngol* 1994;**114**:410–14 [Children already had diagnosed hearing problems.]
- Koike KJ, Wetmore SJ. Interactive effects of the middle ear pathology and the associated hearing loss on transient-evoked otoacoustic emission measures. *Otolaryngol Head Neck Surg* 1999;**121**:238–44. [Children were deaf and blind and already had suspected ear problems.]
- Krueger WWO, Ferguson L. A comparison of screening methods in school-aged children. *Otolaryngol Head Neck Surg* 2002;**127**:516–19. [Children too old.]
- Lee DH, Yeo SW. Clinical diagnostic accuracy of otitis media with effusion in children, and significance of myringotomy: diagnostic or therapeutic? *J Korean Med Sci* 2004;**19**:739–43 [Children already had OME.]
- Lous J. Secretory otitis-media in schoolchildren – is screening for secretory otitis-media advisable? *Dan Med Bull* 1995;**42**:71–99. [Audit.]
- McKenzie E, Magian V, Stokes R. A study of the recommended pass/fail criteria for impedance audiometry in a school screening program. *J Otolaryngol* 1982;**11**:40–5. [Comparative results not valid for this review.]
- Mackie K, Dermody P. Use of a Monosyllabic Adaptive Speech Test (MAST) with young children. Research Note. *J Speech Hear Res* 1986;**29**:275–81. [Included children with learning difficulties.]
- Magnusson M, Rasmussen F, Sundelin C. Early identification of children with communication disabilities – evaluation of a screening programme in a Swedish county. *Acta Paediatr* 1996;**85**:1319–26. [Children too young.]
- Maki-Torkko E, Sorri M, Jarvelin MR. Conditions for paediatric hearing screening: a survey in 28 Finnish child welfare clinics. *Public Health* 1998;**112**:47–51 [A study looking at nurses and practitioners, not children.]
- Marriage J, King J, Briggs J, Lutman ME. The reliability of the SCAN test: results from a primary school population in the UK. *Br J Audiol* 2001;**35**:199–208. [Children too old.]
- Matkin ND. Analysis of a recorded test for the measurement of hearing in children. December 1969. [Paper could not be retrieved.]
- Matusiak M, Wierzbicka M, Szyfter W. [Prevalence of conductive hypoacusis in children aged 5–9 years old from rural area in Poland – prospective screening of healthy subjects] [Polish]. *Otolaryngol Polska* 2002;**56**:459–66. [Non-comparative study.]
- Maw AR, Tiwari RS. Children with glue ear: how do they present? *Clin Otolaryngol Allied Sci* 1988;**13**:171–7. [Wrong outcomes.]
- de Melker RA. Evaluation of the diagnostic value of pneumatic otoscopy in primary care using the results of

tympanometry as a reference standard. *Br J General Pract* 1993;**43**:22–4. [Children included were deaf.]

Merer DM, Gravel JS. Screening infants and young children for hearing loss: examination of the CAST procedure. *J Am Acad Audiol* 1997;**8**:233–42. [Some children had previously diagnosed hearing loss.]

National Institutes of Health. Consensus: early identification of hearing impairment in infants and young children. Summary. *Am J Otol* 1994;**1**:130–1. [Not a study.]

Ng J, Yun HL. Otoacoustic emissions (OAE) in paediatric hearing screening – the Singapore experience. *J Singapore Paediatr Soc* 1992;**34**:1–5. [Children included described as being 'at risk'.]

Okalidou A, Kampanaros M. Teacher perceptions of communication impairment at screening stage in preschool children living in Patras, Greece. *Int J Lang Commun Disord* 2001;**36**:489–502. [Looking at prevalence of communication problems, cross-sectional study that only gives one figure for the prevalence of hearing impairment in their study population.]

Piskorski P, Keefe DH, Simmons JL, Gorga MP. Prediction of conductive hearing loss based on acoustic ear-canal response using a multivariate clinical decision theory. *J Acoust Soc Am* 1999;**105**:1749–64. [Not looking at screening.]

Proschel U, Eysholdt U. Evoked otoacoustic emissions in children in relation to middle ear impedance. *Folia Phoniatr* 1993;**45**:288–94. [The children already had conductive hearing loss in at least one ear.]

Richardson MP, Williamson TJJ, Lenton SW, Tarlow MJ, Rudd PT. Otoacoustic emissions as a screening test for hearing impairment in children. *Arch Dis Child* 1995;**72**:294–7. [No breakdown of age.]

Rothman R, Owens T, Simel DL. Does this child have acute otitis media? [review]. *JAMA* 2003;**290**:1633–40. [Not looking at screening.]

Sagalovich BM, Shimanskaia EI. Age-related characteristics of dynamic indicators of acoustic impedance of the middle ear in children. *Vestn Otorinolaringol* 1992;(3):9–13. [Non-comparative.]

Scaldwell WA. Prevalence of otitis media in Cree and Ojibway school children in six Ontario communities. *Journal of American Indian Education* 1985;**25**:1–5. [Not screening.]

Schuster M, Kummer P, Hoppe U, Eysholdt U, Weber A, Rosanowski F. Guidelines and their practical application in congenital hearing loss. *Gesundheitswesen* 2003;**65**:566–71. [Children already had diagnosed hearing problems.]

Silman S, Silverman CA, Arick DS. Pure-tone assessment and screening of children with middle-ear effusion. *J Am Acad Audiol* 1994;**5**:173–82. [Children already had OME.]

Stewart MG, Ohlms LA, Friedman EM, Sulek M, Duncan NO III, Fernandez AD, *et al.* Is parental perception an accurate predictor of childhood hearing loss? A prospective study. *Otolaryngol Head Neck Surg* 1999;**120**:340–4. [Children already had OME.]

Sturmer RA, Green JA, Funk SG. Cognitive-development related to performance on preschool hearing screening-tests. *J Dev Behav Pediatr* 1983;**4**:94–8. [Not a comparison of hearing tests.]

Swedish Council on Technology Assessment in Health Care. Universal newborn hearing screening – early assessment briefs 2004. [Wrong population.]

Takata GS, Chan LS, Morphew T, Mangione-Smith R, Morton SC, Shekelle P. Evidence assessment of the accuracy of methods of diagnosing middle ear effusion in children with otitis media with effusion. *Pediatrics* 2003;**112**:1379–87. [Just looking at OME diagnosis.]

Taylor CL, Brooks RP. Screening for hearing loss and middle-ear disorders in children using TEOAEs. *Am J Audiol* 2000;**9**:50–5. [Children had been referred.]

Westerlund M, Sundelin C. Screening for developmental language disability in 3-year-old children. Experiences from a field study in a Swedish municipality. *Child Care Health Dev* 2000;**26**:91–110. [Children too young.]

Wood EJ, Lutman ME, Fernandes MA. Validation of a screening oto-admittance instrument. *Br J Audiol* 1982;**16**:273–5. [Wrong population.]

Yasuhara A, Hori A. A comparison of the three-dimensional auditory brainstem response and the conventional auditory brainstem response in children. *Brain Dev* 2002;**24**:750–7. [Not looking at hearing.]

Yockel NJ. A comparison of audiometry and audiometry with tympanometry to determine middle ear status in school-age children. *J Sch Nurs* 2002;**18**:287–92. [Non-comparative study.]

Zakzouk SM. Epidemiology and etiology of hearing impairment among infants and children in a developing country. Part I. *J Otolaryngol* 1997;**26**:335–44. [Non-comparative study.]

Zielhuis GA, Gerritsen AAM, Gorissen WHM, Dekker LJ, Rovers MM, van der Wilt GJ, *et al.* Hearing deficits at school age; the predictive value of otitis media in infants. *Int J Pediatr Otorhinolaryngol* 1998;**44**:227–34 [Not looking at screening.]

Appendix 8

Summary of quality of systematic reviews

TABLE 73 *Quality of systematic reviews*

	Barlow et al., 1998⁴³	NZHTA, 1998⁴⁴	Pirozzo et al., 2003⁴⁷
Was the aim stated clearly?	Yes	Yes	Yes
Were the appropriate sort of papers sought?	Yes	Yes	Can't tell
Are the important relevant studies included?	Yes	Yes	Can't tell
Was the quality of the studies appropriately assessed?	Yes	Yes	Yes
If the results are combined was it appropriate to do so?	NA	NA	NA
Can the results be applied to the local population?	Can't tell	Can't tell	Can't tell
Were all important outcomes considered?	Yes	Yes	Yes
Are the benefits worth the harms and costs?	Yes	Yes	Yes

Appendix 9

Quality of primary studies

TABLE 74 Quality of primary studies

	Q. 1	Q. 2	Q. 3	Q. 4	Q. 5	Q. 6	Q. 7	Q. 8	Q. 9	Q. 10	Q. 11	Q. 12	Q. 13	Q. 14	Score ^c
Ritchie and Merklein, 1972 ⁵⁷	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	10
FitzZaland and Zink, 1984 ⁵⁵	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	9
Gomes and Lichtig, 2005 ⁶⁹	Yes	No	Yes	Unclear	Yes	Yes	Yes	Unclear	Yes	No	No	Unclear	Unclear	Yes	7
Abou Haidar et al., 2005 ⁶⁸	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	Unclear	Unclear	Unclear	Unclear	No	8
Hamill, 1988 ⁵⁸	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Yes	11
Hammond et al., 1997 ⁶³	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	No	9
Hind et al., 1999 ⁶⁵	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	10
Holtby et al., 1997 ⁷⁰	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Unclear	Unclear	Yes	11
Lyons et al., 2004 ⁵³	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	9
Maragno and Teatini, 1983 ^{66a}	Yes	Unclear	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	NA
McCurdy et al., 1976 ⁷¹	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	No	9
Nienhuys et al., 1994 ⁶¹	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Unclear	Yes	9
Nozza et al., 1997 ⁷²	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Yes	11
Olusanya, 2001 ⁵¹	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	8
Orlando and Frank, 1987 ⁷³	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	Unclear	Unclear	Unclear	Unclear	Yes	12
Pang-Ching et al., 1995 ⁵²	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	9
Prescott et al., 1999 ⁵⁰	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Unclear	8
Rodriguez and Melguizo-Yopez, 1994 ^{56a}	Yes	Unclear	Unclear	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	NA
Rousch et al., 1992 ⁵⁹	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Unclear	Unclear	Yes	10
Rousch and Tait, 1985 ⁶²	Yes	Unclear	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	8
Sabo et al., 2000 ⁵⁴	Yes	Unclear	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	8
Schell, 1970 ^{64a}	Yes	Unclear	Unclear	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	NA
Skurr and Jones, 1981 ⁷⁵	Yes	Yes	Unclear	Yes	Unclear	Yes	Yes	No	No	Unclear	Unclear	Unclear	Unclear	No	5
Square et al., 1985 ⁶⁰	Yes	No	Unclear	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	8
Totals^b	20	11	17	18	12	20	19	16	16	4	5	0	0	16	

^a Non-English studies.
^b Total number of 'yes' responses to each criteria.
^c Total number of 'yes' scores for each study.

Questions

1. Was the spectrum of patients representative of the patients who will receive the test in practice?
2. Were the selection criteria clearly described?
3. Is the reference standard likely to correctly classify the target condition? (PTA has been taken as the most suitable reference standard.)
4. Is the time period between reference standard and index test short enough to be reasonable?
5. Did the whole sample or a random selection of the sample receive verification using a reference standard?
6. Did patients receive the same regardless of the index test result?

continued

TABLE 74 *Quality of primary studies (cont'd)*

7. Was the reference standard independent of the index test? (i.e. the index test did not form part of the reference standard)
8. Was the execution of the index test described in sufficient detail to permit replication of the test?
9. Was the execution of the reference standard described in sufficient detail to permit its replication?
10. Were the index test results interpreted without knowledge of the results of the reference standard?
11. Were the reference standard results interpreted without knowledge of the results of the index test?
12. Were the same clinical data available when test results were interpreted as would be available when the test is used in practice?
13. Were uninterpretable/intermediate test results reported?
14. Were any withdrawals from the study explained?

Appendix 10

Two by two tables for sensitivity and specificity where available

Ritchie and Merklein⁵⁷

TABLE 75 VASC screen (protocol 1) versus PTA

	Failed intervention	Passed intervention	
Passed control	5	116	Sensitivity 51%
Failed control	21	20	Specificity 96%

TABLE 76 VASC screen (protocol 2) versus PTA

	Failed intervention	Passed intervention	
Passed control	8	113	Sensitivity 59%
Failed control	24	17	Specificity 93%

FitzZaland and Zink⁵⁵

TABLE 77 Pure tone sweep versus combination of tests

	Failed intervention	Passed intervention	
Passed control	39	3334	Sensitivity 93%
Failed control	128	9	Specificity 99%

TABLE 78 Rinne audiometric test versus combination of tests

	Failed intervention	Passed intervention	
Passed control	11	3362	Sensitivity 91%
Failed control	124	13	Specificity 99.67%

TABLE 79 Tympanometry (negative pressure ≥ -150 mm) versus combination of tests

	Failed intervention	Passed intervention	
Passed control	298	3057	Sensitivity 93%
Failed control	127	10	Specificity 91%

TABLE 80 Tympanometry (negative pressure ≥ -175 mm) versus combination of tests

	Failed intervention	Passed intervention	
Passed control	183	3190	Sensitivity 91%
Failed control	127	10	Specificity 99%

TABLE 81 Tympanometry (negative pressure ≥ -200 mm) versus combination of tests

	Failed intervention	Passed intervention	
Passed control	73	3300	Sensitivity 91%
Failed control	125	12	Specificity 99%

TABLE 82 Tympanometry type B only

	Failed intervention	Passed intervention	
Passed control	0	3373	Sensitivity 40%
Failed control	55	82	Specificity 100%

Hammond and colleagues⁶³

TABLE 83 Questionnaire versus combination of tests

	Failed intervention	Passed intervention	
Passed control	259	227	Sensitivity 56%
Failed control	10	8	Specificity 52%

Hamill⁵⁸

TABLE 84 VASC versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	23	508	Sensitivity 87%
Failed control	39	6	Specificity 96%

Lyons and colleagues⁵³

TABLE 85 Tympanometry versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	171	1725	Sensitivity 85%
Failed control	94	16	Specificity 91%

TABLE 86 DPOAE (protocol 1) versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	241	1484	Sensitivity 97%
Failed control	273	8	Specificity 86%

TABLE 87 DPOAE (protocol 2) versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	289	1436	Sensitivity 97%
Failed control	273	8	Specificity 83%

TABLE 88 DPOAE (protocol 3) versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	440	1285	Sensitivity 98%
Failed control	277	4	Specificity 74%

TABLE 89 DPOAE (protocol 4) versus pure tone sweep test

	Failed intervention	Passed intervention	
Passed control	92	1633	Sensitivity 96%
Failed control	269	12	Specificity 95%

Maragno and Teatini⁶⁶

TABLE 90 SVEP test versus hearing assessment

	Failed intervention	Passed intervention	
Passed control	0	48	Sensitivity 100%
Failed control	31	2	Specificity 94%

McCurdy and colleagues⁷¹

TABLE 91 Tympanometry plus stapedius reflex versus PTA

	Failed intervention	Passed intervention	
Passed control	24	45	Sensitivity 71%
Failed control	57	23	Specificity 65%

Olusanya⁵¹

TABLE 92 Questionnaire versus pure tone audiometry

	Failed intervention	Passed intervention	
Passed control	15	291	Sensitivity 34%
Failed control	23	45	Specificity 95%

TABLE 93 Tympanometry versus PTA

	Failed intervention	Passed intervention	
Passed control	50	253	Sensitivity 50%
Failed control	25	25	Specificity 83%

TABLE 94 Otoscopy versus PTA

	Failed intervention	Passed intervention	
Passed control	116	193	Sensitivity 56%
Failed control	28	22	Specificity 62.4%

Sabo and colleagues⁵⁴

TABLE 95 Pure tone sweep test versus PTA

	Failed intervention	Passed intervention	
Passed control	106	429	Sensitivity 87%
Failed control	33	5	Specificity 80%

TABLE 96 TEOAE versus PTA

	Failed intervention	Passed intervention	
Passed control	48	487	Sensitivity 63%
Failed control	24	14	Specificity 91%

Square and colleagues⁶⁰

TABLE 97 Bone conduction versus impedance audiometry

	Failed intervention	Passed intervention	
Passed control	56	4	Sensitivity 26%
Failed control	14	39	Specificity 6.6%

Appendix 11

Economic search strategies

MEDLINE

1966 to August week 3 2005

Search date: 2 August 2005

Number of records: 74

Hearing loss\$.mp or exp Hearing loss/ or (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp or Hearing Disorders/ AND exp child, preschool/ or school entry.mp or exp Child Development/ or early detect\$.mp or infant school\$.mp or exp Schools, Nursery/ or kindergarten\$.mp or exp Child Day Care Centers/ or exp Nurseries/ or nursery school\$.mp AND screen\$.mp or exp Mass Screening/ AND (school entry adj3 (screen\$ or exam\$)).mp or (medical exam\$ adj2 school\$).mp AND economics.mp or exp ECONOMICS, NURSING/ or exp ECONOMICS, MEDICAL/ or exp ECONOMICS/ or exp ECONOMICS, HOSPITAL/ or exp ECONOMICS, PHARMACEUTICAL/ or (econom\$ or cost or costs or costly or costing or costed or price or prices or pricing or pharmaco-economic\$.tw or (expenditure\$ not energy).tw or (value adj1 money).tw or budget\$.tw or cost-effectiveness.mp or cost utili\$.mp or cost benefit.mp or exp Cost-Benefit Analysis/ or cost minimi\$.mp or exp Health Care Costs/ or economic evaluation\$.mp or exp 'Costs and Cost Analysis'/ or financ\$.mp or exp Resource Allocation/ or health resource allocation.mp or Health Resources/ or health resource utilization.mp or preference?.ab,ti,kw or qaly?.ab,ti,kw or quality adjusted.ab,ti,kw or (utility or utilities).ab,ti,kw

EMBASE

1980 to 2005 week 31

Search date: 2 August 2005

Number of records: 38

Hearing loss\$.mp or exp Hearing loss/ or (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp or Hearing Disorders/ AND school entry.mp or (pre adj school).mp or (nursery adj school\$.mp or exp Nursery School/ or kindergarten\$.mp or exp KINDERGARTEN/ or exp Day Care/ or infant school\$.mp early

detect\$.mp or exp Child Development/ AND screen\$.mp or exp MASS SCREENING/ or exp SCREENING/ or exp AUDITORY SCREENING/ or exp SCREENING TEST/ AND (school entry adj3 (screen\$ or exam\$)).mp or (medical exam\$ adj2 school\$).mp AND 'Cost Benefit Analysis'/ or 'Cost-effectiveness Analysis'/ or 'Cost Minimization Analysis'/ or 'Cost Utility Analysis'/ or Economic Evaluation/ or (cost or costs or costed or costly or costing).tw or (economic\$ or pharmaco-economic\$ or price\$ or pricing).tw or (technology adj assessment\$.tw

CINAHL

1982 to August 2005 week 5

Search date: 2 August 2005

Number of records: 32

Hearing loss\$.mp or exp Hearing loss/ or (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp or Hearing Disorders/ AND school entry.mp or (pre adj school).mp or (nursery adj school\$.mp or exp Schools, Nursery/ or kindergarten\$.mp or exp Day Care/ or infant school\$.mp or early detect\$.mp or exp Child Development/ AND (school entry adj3 (screen\$ or exam\$)).mp or (medical exam\$ adj2 school\$).mp AND exp Health Screening/ or screen\$.mp or exp Hearing Screening/ AND (cost or costs or costed or costly or costing).tw or (economic\$ or pharmaco-economic\$ or price\$ or pricing).tw or (technology adj assessment\$.tw or cost benefit analysis.mp or exp 'Cost Benefit Analysis'/ or cost-effectiveness.mp or cost minimization.mp or exp Health Care Costs/ or cost utility.mp or economic evaluation\$.mp or exp 'Economic Aspects of Illness'/ or exp ECONOMICS/ or exp ECONOMICS, PHARMACEUTICAL/ or health resource allocation.mp or exp Health Resource Allocation/ or health resource utilization.mp or exp Health Resource Utilization/

Cochrane Library (Wiley) NHS EED 2005 Issue 2

Search date: 28 July 2005

Number of records: 12

Exp hearing loss/ or hearing next loss or hearing next disorder* or hearing next difficult* or hearing next problem* or hearing next impair* or exp hearing disorders/ **AND** screening or exp mass screening/ **AND** school next entr* or early detect* or infant next school* or nursery next school* or kindergarten* or exp schools, nursery/ or exp Child Day Care Centers/ or exp Child, Preschool/ or exp Child Development/ **OR** school next entr* or medical near/3 exam* near/3 school*

ECONLIT

1969–2002 and 2003–2005

Search date: 28 July 2005

Number of records: 11

Hearing loss* or hearing disorder* or hearing difficult* or hearing problem* or hearing impair* or deafness or hypoacusis or hypacusis or hard of hearing or hard-of-hearing

OHE HEED

July 2005 issue

Search date: 26 July 2005

Number of records: 18

Hearing **AND** screen* **AND** child*

Total references (after de-duplication): 164

Appendix 12

Subsequent management intervention search strategies

MEDLINE

1966 to November week 3 2005

Search date: 17 November 2005

Number of records: 397

Hearing loss\$.mp or exp Hearing loss/ or (hearing adj (disorder\$ or difficult\$ or problem\$ or impair\$)).mp or Hearing Disorders/ or deaf\$.mp or otitis media with effusion.mp or exp Otitis Media with Effusion/ or OME.mp or glue ear.mp or (hard adj1 hearing).mp **AND** economics.mp or exp ECONOMICS, NURSING/ or exp ECONOMICS, MEDICAL/ or exp ECONOMICS/ or exp ECONOMICS, HOSPITAL/ or exp ECONOMICS, PHARMACEUTICAL/ or (econom\$ or cost or costs or costly or costing or costed or price or prices or pricing or pharmacoeconomic\$.tw or (expenditure\$ not energy).tw or (value adj1 money).tw or budget\$.tw or cost-effectiveness.mp or cost utili\$.mp or cost benefit.mp or exp Cost-Benefit Analysis/ or cost minimi\$.mp or exp Health Care Costs/ or economic evaluation\$.mp or exp 'Costs and Cost Analysis'/ or financ\$.mp or exp Resource Allocation/ or health resource allocation.mp or Health Resources/ or health resource utilization.mp or preference?.ab,ti,kw or qaly?.ab,ti,kw or quality adjusted.ab,ti,kw or (utility or utilities).ab,ti,kw **AND** hearing aid\$.mp or exp Hearing Aids/ or cochlear implant\$.mp or exp Cochlear Implants/ or exp 'Rehabilitation of Hearing Impaired'/ or hearing tactic\$.mp or autoinflation.mp or exp Middle Ear Ventilation/ or grommet\$.mp or tympanostomy.mp or myringotomy.mp or adenoidectomy.mp or exp ADENOIDECTOMY/ or exp Language Therapy/ or exp Speech Therapy/ or speech language therapy.mp or ear nose throat.mp or exp EDUCATION/ or education\$.mp or exp EDUCATION, SPECIAL/ or exp Teaching/ or teach\$.mp or exp Comprehensive Health Care/ or audiolog\$.mp or otolaryngology\$.mp or pediatric\$.mp or clinician\$.mp or exp NURSE CLINICIANS/ or exp Nursing Staff/ **AND** limit to ('newborn infant (birth to 1 month)' or 'infant (1 to 23 months)' or 'preschool child (2 to 5 years)' or 'child (6 to 12 years)')

EMBASE

1980 to 2006 week 3

Search date: 16 January 2006

Number of records: 190

'hearing loss'/exp or 'hearing loss' or 'hearing disorder'/exp or 'hearing disorder' or 'hearing difficulty'/exp or 'hearing difficulty' or 'hearing problem' or 'hearing impairment'/exp or 'hearing impairment' or deaf* or 'otitis media with effusion'/exp or 'otitis media with effusion' or ome or 'glue ear'/exp or 'glue ear' or 'hard *1 hearing' **AND** 'cost benefit analysis'/exp or 'cost benefit analysis' or 'cost-effectiveness analysis'/exp or 'cost-effectiveness analysis' or 'cost minimization analysis'/exp or 'cost minimization analysis' or 'economic evaluation'/exp or 'economic evaluation' or ('cost'/exp or 'cost') or costs or costed or costly or costing or economic* or pharmacoeconomic* or price or pricing or 'technology *3 assessment' or 'health resources'/exp or 'health resources' or 'quality of life'/exp or 'quality of life' **AND** 'hearing aid'/exp or 'hearing aid' or 'cochlear prosthesis'/exp or 'cochlear prosthesis' or 'auditory rehabilitation'/exp or 'auditory rehabilitation' or autoinflation or 'middle ear ventilation'/exp or 'middle ear ventilation' or 'tympanostomy tube'/exp or 'tympanostomy tube' or grommet* or 'myringotomy'/exp or 'myringotomy' or adenoidectomy'/exp or 'adenoidectomy' or 'speech therapy'/exp or 'speech therapy' or 'otorhinolaryngology'/exp or 'otorhinolaryngology' or 'ear nose throat surgery'/exp or 'ear nose throat surgery' or 'education'/exp or 'education' or 'teaching'/exp or 'teaching' or 'audiology'/exp or 'audiology' or pediatric* or nurs* **AND** [embase]/lim **AND** ([newborn]/lim or [infant]/lim or [preschool]/lim or [school]/lim)

CINAHL

1982 to December 2005 week 1

Search date: 1 December 2005

Number of records: 263

Hearing loss\$.mp or exp Hearing loss/ or (hearing adj (disorder\$ or difficult\$ or problem\$ or

impair\$)).mp or Hearing Disorders/ or deaf\$.mp or exp Deafness/ or otitis media with effusion.mp or exp Otitis Media with Effusion/ or OME.mp or glue ear.mp or (hard adj1 hearing).mp **AND** (cost or costs or costed or costly or costing).tw or (economic\$ or pharmacoeconomic\$ or price\$ or pricing).tw or (technology adj assessment\$).tw or cost benefit analysis.mp or exp 'Cost Benefit Analysis'/ or cost-effectiveness.mp or cost minimization.mp or exp Health Care Costs/ or cost utility.mp or economic evaluation\$.mp or exp 'Economic Aspects of Illness'/ or health resource allocation.sh,mp or health resource utilization.sh,mp or preference?.mp or exp Life Expectancy/ or exp 'Outcomes (Health Care)'/ or exp Quality of Life'/ or qaly?.mp or exp Health Status/ or quality adjusted.mp or (utility or utilities).mp **AND** hearing aid\$.mp or exp Hearing Aids/ or cochlear implant\$.mp or exp Cochlear Implant/ or Communication Skills/ or exp 'Rehabilitation of Hearing Impaired'/ or hearing tactic\$.mp or Conversation/ or Counseling/ or autoinflation.mp or exp Middle Ear Ventilation/ or grommet\$.mp or tympanostomy.mp or myringotomy.mp or adenoidectomy.mp or exp ADENOIDECTOMY/ or exp Education, Speech-Language Pathology/ or exp 'Rehabilitation, Speech and Language'/ or speech language therapy.mp or exp Speech Therapy/ or exp 'Education, Continuing (Credit)'/ or exp Surgery, Otorhinolagynologic/ or exp Specialties, Nursing/ or ear nose throat.mp or exp DEAF EDUCATION/ or exp PATIENT EDUCATION/ or exp PARENTING EDUCATION/ or education\$.mp or exp EDUCATION, SPECIAL/ or exp 'OUTCOMES OF EDUCATION'/ or exp EDUCATION, CONTINUING/ or exp EDUCATION, AUDIOLOGY/ or exp EDUCATION, SPEECH-LANGUAGE PATHOLOGY/ or exp EDUCATION/ or teach\$.mp or audiolog\$.mp or otolaryngolog\$.mp or exp PEDIATRICS/ or pediatric\$.mp or clinician\$.mp **AND** limit to (newborn infant <birth to 1 month> or infant <1 to 23 months> or preschool child <2 to 5 years> or child <6 to 12 years>)

Cochrane Library (Wiley internet version) NHS EED 2006 Issue 1

Search date: 31 January 2006

Number of records: 69

Hearing next (loss or disorder* or difficulty or problem or impairment) or deaf or otitis next media or OME or glue next ear or hard near/2 hearing **AND** hearing next aid* or cochlear next implant* or rehabilitation near/2 hearing or hearing next tactics or autoinflation or middle next ear next ventilation or grommet* or tympanostomy or myringotomy or adenoidectomy or language next therapy or speech next therapy or ear next nose or education or teaching or comprehensive next health or audiology or otolaryngology or paediatrician* or clinician* **AND** restrict to NHS EED database

ECONLIT

1969–2002 and 2003–2005/12

Search date: 11 January 2006

Number of records: 39

hearing or deaf* or otitis **AND** manage* or treat* or intervene* or otolaryngology* or audiolog* or speech or rehabilit* or nurs* or aid* or implant* or grommet* or autoinflation or tympanostomy or myringotomy or adenoid* or language

OHE HEED

January 2006 issue

Search date: 10 January 2006

Number of records: 189

hearing or deaf* or otitis **AND** manage* or treat* or intervene* or otolaryngology* or audiolog* or speech or rehabilit* or nurs* or aid* or implant* or grommet* or autoinflation or tympanostomy or myringotomy or adenoid* or language

Total references (after de-duplication): 960

Appendix 13

Inclusion/exclusion economic data form

Trial author and date:	Ref. no:		
	Yes	No	?
1. Based on primary data collection or systematic review?			
2. Children 4–6 years old, undergoing any of the following interventions?			
Pure tone audiometry			
Questionnaires			
Reflectometry			
Otoadmittance tests			
Speech audiometry			
Otoacoustic emissions			
Medical examinations (which entail a hearing screen)			
3. Including at least two of the following interventions?			
Pure tone audiometry			
Tympanometry			
Acoustic reflex			
Otoadmittance tests			
Auditory brainstem response			
Medical examinations (which entail a hearing screen)			
Speech perception tests			
Distraction test			
Behavioural test			
Questionnaires			
Otoacoustic emissions			
No screen			
4. Assessing any of the following outcomes?			
Year with no or mild/moderate disability due to hearing loss			
Year with moderate or severe disability due to hearing loss			
Quality-adjusted life-year gained			
Utility measure			
Health status measure			
5. Resource use and costs and utilities associated with screening programmes and subsequent management?			
6. Report resource use and cost separately?			
7. Report sufficient detail to extract costs and outcome data relevant to each alternative comparison of screening programmes?			

Appendix 14

Inclusion/exclusion subsequent interventions data form

Trial author and date:	Ref. no:		
	Yes	No	?
1. Based on primary data collection or systematic review?			
2. Children from birth to 12 years of age, undergoing any of the following interventions?			
Hearing aids			
Autoinflation			
Middle ear ventilation			
Myringotomy			
Adenoidectomy			
Speech and language therapy			
Hearing tactics (family, community, school)			
Referral to specialists			
Cochlear implantation			
3. Assessing any of the following outcomes?			
Year with no or mild/moderate disability due to hearing loss			
Year with moderate or severe disability due to hearing loss			
Quality-adjusted life-year gained			
Utility measure			
Health status measure			
4. Resource use and costs and utilities associated with subsequent management interventions?			
5. Report resource use and cost separately?			

Appendix 15

Economic data extraction form

Economic Data Extraction Form		
General Information		
Paper Reference No.	Date:	Reviewer ID:
Author/Year:		
Title:		
Sub Title:		
Journal:		
Source of funding:		
Notes/Comments:		
Study Characteristics		
Health Technology:		
Comparator:		
Type of Intervention	Economic Study Type	Perspective
Primary Prevention <input type="checkbox"/>	Cost-effectiveness Analysis <input type="checkbox"/>	NHS <input type="checkbox"/>
Secondary Prevention <input type="checkbox"/>	Cost-utility Analysis <input type="checkbox"/>	Societal <input type="checkbox"/>
Screening <input type="checkbox"/>	Cost-benefit Analysis <input type="checkbox"/>	Hospital <input type="checkbox"/>
Diagnosis <input type="checkbox"/>	Cost-consequence Analysis <input type="checkbox"/>	Not Stated <input type="checkbox"/>
Treatment <input type="checkbox"/>	Cost-Study <input type="checkbox"/>	Other (<i>Please Specify</i>) <input type="checkbox"/>
Rehabilitation <input type="checkbox"/>	Not Reported <input type="checkbox"/>	Setting:
Palliative Care <input type="checkbox"/>		
Other (<i>Please Specify</i>) <input type="checkbox"/>		
Not Reported <input type="checkbox"/>		
Hypothesis/Study Question:		
Study Population:		
Dates to which Data Relate	Modelling	
Effectiveness Evidence <input type="checkbox"/>	Was a model used?	
Resource Use <input type="checkbox"/>	Yes <input type="checkbox"/>	
Price Year <input type="checkbox"/>	No <input type="checkbox"/>	
	If <i>yes</i> state purpose and type:	

Source of Data		Source of Cost Data	
Source of Effectiveness Data		Actual Source	
Single Study	<input type="checkbox"/>	Literature Source	<input type="checkbox"/>
Synthesis of Prev. Pub.	<input type="checkbox"/>		
Link between Effectiveness and Costs			
Effectiveness data from a single study			
Study Sample:		Study design:	
Power calculation		RCT	Duration of follow-up:
Number subjects in intervention group		Non-RCT with concurrent controls	Loss to follow-up:
Number subjects in control group		Cohort study	Any blinding for assessment of outcomes:
Recruitment rate		Historical controls	Analysis of clinical study:
Number excluded from study		Before and after study	Treatment completers
Method of sample selection:		Case series	Intention to treat
		Other (specify)	Effectiveness results:
		Not reported	
		Number of centres	
Effectiveness data from a synthesis of previous studies (model)			
Study inclusion criteria:	Study designs included:		Number of primary studies included:

Study exclusion criteria reported:	RCT			Method of combination of primary studies:		
Sources searched reported:	Non-RCT with concurrent controls			Meta-analysis		
Criteria used to judge validity:	Cohort study			Narrative method		
Concealment of randomisation	Historical controls			Other (specify)		
Blind assessment	Before and after study			Results of the review:		
Low drop out rates	Case series					
Other (specify)	Other (specify)					
Not reported	Not reported					

Economic Evaluation	
Measure of Benefits used in the Economic Analysis	
No Measure of Benefit (CCA or CMA) <input type="checkbox"/>	
Direct costs: Health service	Estimation of Direct Costs Based On:
	A Guess <input type="checkbox"/>
	Actual Data <input type="checkbox"/>
	Derived using Modelling <input type="checkbox"/>
	Other <input type="checkbox"/>
	Not Reported <input type="checkbox"/>
Direct costs: Patient	Estimation of Patient Direct Costs Based On:
	A Guess <input type="checkbox"/>
	Actual Data <input type="checkbox"/>
	Derived using Modelling <input type="checkbox"/>
	Other <input type="checkbox"/>
	Not Reported <input type="checkbox"/>
Source of Direct Cost Data:	Discounting Undertaken?
Price Year:	Yes <input type="checkbox"/> Discount Rate
Currency:	

<i>Economic Evaluation (continued)</i>	
Indirect Costs	Estimation of Indirect Costs Based On:
	A Guess <input type="checkbox"/>
	Actual Data <input type="checkbox"/>
	Derived using Modelling <input type="checkbox"/>
	Other <input type="checkbox"/>
	Not Reported <input type="checkbox"/>
Source of Indirect Cost Data	Discounting Undertaken?
Price Year:	
	Yes <input type="checkbox"/> Discount Rate:
	No <input type="checkbox"/>
Currency:	Conversion Rates Used:
<i>Statistical/Sensitivity Analyses</i>	
Statistical Tests Carried Out?	Types of test used in Analysis of Costs:
Yes <input type="checkbox"/>	
No <input type="checkbox"/>	

Type of Sensitivity Analysis:		Areas of Uncertainty Tested:			
One-way Analysis	<input type="checkbox"/>				
Two-way Analysis	<input type="checkbox"/>				
Multi-way Analysis	<input type="checkbox"/>				
Threshold Analysis	<input type="checkbox"/>				
Analysis of Extremes	<input type="checkbox"/>				
Probabilistic Analysis	<input type="checkbox"/>				
Other	<input type="checkbox"/>				
Not Reported	<input type="checkbox"/>				
Not Carried Out	<input type="checkbox"/>				
Results					
Clinical Outcome/Benefit:					
Duration of Benefits:		Side Effects Considered?		Y	N
Cost results:					
Cost of Adverse Events Considered?				Y	N
How were the estimates of Costs and Benefits Combined?				Results of Synthesis of Costs and Benefits:	
Cost/Life Saved	<input type="checkbox"/>				
Cost/Life Gained	<input type="checkbox"/>				
Cost/QALY	<input type="checkbox"/>				

Net Benefit	<input type="checkbox"/>		
Incremental Net Benefit	<input type="checkbox"/>		
Other	<input type="checkbox"/>		
Not Combined	<input type="checkbox"/>		
Author's Conclusions:			
Reviewer's Conclusions:			
Overall assessment of study quality:			

Appendix 16

Description of included papers

TABLE 98 Description of included papers

Study characteristics	Comparators	Economic study type and population	Data
<p>Authors: Driscoll, et al., 2000⁸⁸</p> <p>Setting: Audiological clinic, Australia</p> <p>Perspective: Not stated</p> <p>Funding: University of Queensland</p>	NA	<p>Study: Study</p> <p>Health technology: TEOAE and PTA</p> <p>Type of intervention: Screening</p> <p>Population: Infants with a mean age of 2 months (range 1.5–2.5) and children with a mean age of 6.2 years (range 5.2–7.9)</p> <p>Sources: Actual study, synthesis of data from previous publications for effectiveness</p>	<p>Date: 2000</p> <p>Effectiveness: Rate of tests per hour</p> <p>Resource use: Audiologists, equipment</p> <p>Price year: Not clear</p> <p>Direct costs: Hourly wage of a full-time audiologist, cost of equipment, cost of annual equipment maintenance, cost per child screened</p> <p>Indirect costs: NR</p> <p>Currency: Australian dollars</p> <p>Link between effectiveness and costs: $\text{Cost per child} = S/R + (C + (M*L)/(N*L))$</p> <p>where S = salary of screener, R = rate of tests/hour, C = cost of initial equipment purchase, M = cost of annual maintenance, L = expected life of equipment in years, and N = number of cases per year</p> <p>The total annual cost (TC) of the programmes was determined using the following equation: $\text{TC} = \text{CPC} \times N$. The cost per hearing impaired child (CPHla) was calculated using the equation: $\text{CPHla} = \text{TC}/\#\text{HL}$ (where #HL is the number of children diagnosed with a unilateral or bilateral, sensorineural or mixed hearing impairment of at least a moderate degree ($3\text{FA} \geq 41$)). The cost per child with sensorineural/mixed/conductive hearing impairment was calculated based on the number of children diagnosed with any hearing impairment</p>
<p>Authors: Holtby and Forster, 1992⁸⁹</p> <p>Setting: Infant or primary schools, UK</p> <p>Perspective: NHS</p> <p>Funding: Not stated</p>	Impedance measurements against PTA	<p>Study: Effectiveness</p> <p>Health technology: Impedance measurements</p> <p>Type of intervention: Screening</p> <p>Population: 6-year-old schoolchildren</p> <p>Sources: Single study for the effectiveness data. The source for the cost data was not reported</p>	<p>Date: 1988</p> <p>Effectiveness: Sensitivity, specificity, repeatability, predictive value, screening rate</p> <p>Resource use: Nurses, audiometers</p> <p>Price year: NR</p> <p>Direct costs: Salary, cost of equipment, cost of annual maintenance</p> <p>Indirect costs: NR</p> <p>Currency: UK sterling</p> <p>Link between effectiveness and costs: $\text{Cost per child screened per instrument} = S/R + (C + (M*L)/(N*L))$</p> <p>where S = salary of screener, R = rate of tests, C = cost of equipment, M = cost of annual maintenance, L = expected life of equipment in years, and N = number of children screened per instrument per year</p>



Health Technology Assessment reports published to date

Volume 1, 1997

No. 1

Home parenteral nutrition: a systematic review.

By Richards DM, Deeks JJ, Sheldon TA, Shaffer JL.

No. 2

Diagnosis, management and screening of early localised prostate cancer.

A review by Selley S, Donovan J, Faulkner A, Coast J, Gillatt D.

No. 3

The diagnosis, management, treatment and costs of prostate cancer in England and Wales.

A review by Chamberlain J, Melia J, Moss S, Brown J.

No. 4

Screening for fragile X syndrome.

A review by Murray J, Cuckle H, Taylor G, Hewison J.

No. 5

A review of near patient testing in primary care.

By Hobbs FDR, Delaney BC, Fitzmaurice DA, Wilson S, Hyde CJ, Thorpe GH, *et al.*

No. 6

Systematic review of outpatient services for chronic pain control.

By McQuay HJ, Moore RA, Eccleston C, Morley S, de C Williams AC.

No. 7

Neonatal screening for inborn errors of metabolism: cost, yield and outcome.

A review by Pollitt RJ, Green A, McCabe CJ, Booth A, Cooper NJ, Leonard JV, *et al.*

No. 8

Preschool vision screening.

A review by Snowdon SK, Stewart-Brown SL.

No. 9

Implications of socio-cultural contexts for the ethics of clinical trials.

A review by Ashcroft RE, Chadwick DW, Clark SRL, Edwards RHT, Frith L, Hutton JL.

No. 10

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

By Davis A, Bamford J, Wilson I, Ramkalawan T, Forshaw M, Wright S.

No. 11

Newborn screening for inborn errors of metabolism: a systematic review.

By Seymour CA, Thomason MJ, Chalmers RA, Addison GM, Bain MD, Cockburn F, *et al.*

No. 12

Routine preoperative testing: a systematic review of the evidence.

By Munro J, Booth A, Nicholl J.

No. 13

Systematic review of the effectiveness of laxatives in the elderly.

By Petticrew M, Watt I, Sheldon T.

No. 14

When and how to assess fast-changing technologies: a comparative study of medical applications of four generic technologies.

A review by Mowatt G, Bower DJ, Brebner JA, Cairns JA, Grant AM, McKee L.

Volume 2, 1998

No. 1

Antenatal screening for Down's syndrome.

A review by Wald NJ, Kennard A, Hackshaw A, McGuire A.

No. 2

Screening for ovarian cancer: a systematic review.

By Bell R, Petticrew M, Luengo S, Sheldon TA.

No. 3

Consensus development methods, and their use in clinical guideline development.

A review by Murphy MK, Black NA, Lamping DL, McKee CM, Sanderson CFB, Askham J, *et al.*

No. 4

A cost-utility analysis of interferon beta for multiple sclerosis.

By Parkin D, McNamee P, Jacoby A, Miller P, Thomas S, Bates D.

No. 5

Effectiveness and efficiency of methods of dialysis therapy for end-stage renal disease: systematic reviews.

By MacLeod A, Grant A, Donaldson C, Khan I, Campbell M, Daly C, *et al.*

No. 6

Effectiveness of hip prostheses in primary total hip replacement: a critical review of evidence and an economic model.

By Faulkner A, Kennedy LG, Baxter K, Donovan J, Wilkinson M, Bevan G.

No. 7

Antimicrobial prophylaxis in colorectal surgery: a systematic review of randomised controlled trials.

By Song F, Glenny AM.

No. 8

Bone marrow and peripheral blood stem cell transplantation for malignancy.

A review by Johnson PWM, Simnett SJ, Sweetenham JW, Morgan GJ, Stewart LA.

No. 9

Screening for speech and language delay: a systematic review of the literature.

By Law J, Boyle J, Harris F, Harkness A, Nye C.

No. 10

Resource allocation for chronic stable angina: a systematic review of effectiveness, costs and cost-effectiveness of alternative interventions.

By Sculpher MJ, Petticrew M, Kelland JL, Elliott RA, Holdright DR, Buxton MJ.

No. 11

Detection, adherence and control of hypertension for the prevention of stroke: a systematic review.

By Ebrahim S.

No. 12

Postoperative analgesia and vomiting, with special reference to day-case surgery: a systematic review.

By McQuay HJ, Moore RA.

No. 13

Choosing between randomised and nonrandomised studies: a systematic review.

By Britton A, McKee M, Black N, McPherson K, Sanderson C, Bain C.

No. 14

Evaluating patient-based outcome measures for use in clinical trials.

A review by Fitzpatrick R, Davey C, Buxton MJ, Jones DR.

No. 15

Ethical issues in the design and conduct of randomised controlled trials.

A review by Edwards SJL, Lilford RJ, Braunholtz DA, Jackson JC, Hewison J, Thornton J.

No. 16

Qualitative research methods in health technology assessment: a review of the literature.

By Murphy E, Dingwall R, Greatbatch D, Parker S, Watson P.

No. 17

The costs and benefits of paramedic skills in pre-hospital trauma care.

By Nicholl J, Hughes S, Dixon S, Turner J, Yates D.

No. 18

Systematic review of endoscopic ultrasound in gastro-oesophageal cancer.

By Harris KM, Kelly S, Berry E, Hutton J, Roderick P, Cullingworth J, *et al.*

No. 19

Systematic reviews of trials and other studies.

By Sutton AJ, Abrams KR, Jones DR, Sheldon TA, Song F.

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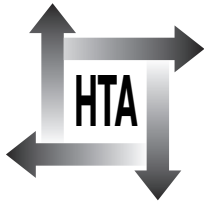
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