UK National Screening Committee

Screening for permanent hearing loss in children at school entry

External review against programme appraisal criteria for the UK National Screening Committee

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About the UK National Screening Committee (UK NSC)

The UK NSC advises ministers and the NHS in the 4 UK countries about all aspects of population screening and supports implementation of screening programmes.
Conditions are reviewed against evidence review criteria according to the UK NSC's evidence review process.

Read a complete list of UK NSC recommendations.

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Plain English summary

Hearing problems can affect the development of children's speech and communication skills. It can also affect their behaviour and ability to learn. There are different types of hearing loss. Hearing loss can be permanent or temporary.

About 1,300 children born in the UK each year have permanent hearing loss in one or both ears of moderate or worse severity.

Since 2006, most children are screened for hearing loss when they are born. But children with hearing loss can be missed or children can develop hearing loss when they are older. Some children are screened for hearing loss when they start school.

This document looks at screening children for permanent hearing loss when they start school. It includes new evidence published up to June 2018. In the past, the UK NSC recommended that screening children for hearing loss when they start school should continue whilst further research was being undertaken.

This document looks at some key questions:

1. how many UK children starting school have hearing loss?
2. how accurate are hearing screening tests in children starting school?
3. what are the results of hearing screening programmes for children starting school?

There is not enough evidence to change the current recommendation about screening for permanent hearing loss for children starting school. These areas are uncertain:

- how many UK children starting school with temporary hearing loss would be identified
- the accuracy of screening tests for permanent hearing loss in children starting school
- a lack of evidence suggesting an advantage to screening children at school entry age.

Executive summary

Purpose of the review

This document reviews the evidence on screening for permanent hearing loss in children at school entry against selected UK National Screening Committee (NSC) criteria.

Background

Hearing deficits can interfere with a child's speech and language development, communication, behaviour and ability to learn. Conductive hearing loss affects the passage of sound between the eardrum and inner ear and can be temporary or permanent. In sensorineural hearing loss there is permanent damage to the hair cells in the cochlea (the sensory hearing organ) or damage to the hearing neural pathways. The severity of hearing loss is measured in decibels (dB) with different categories for degree of hearing loss. Some screening tests for hearing impairment do not distinguish between permanent and temporary hearing impairment.

In the UK approximately 1 per 1,000 children (about 800 children per year) are born with a permanent hearing impairment of more than 40dB (moderate impairment) in both ears. An additional 0.6 per 1,000 (about 500 children per year) have a hearing impairment in one ear. Not all children will have a hearing impairment that can be identified at birth. The UK prevalence of permanent hearing impairment of more than 40dB at 3 years old is 1.07 per 1,000. For children aged 9 to 15 years this is 2.05 per 1,000.

School entry hearing screening was introduced in 1955 and remains in place in many parts of the UK. The number of children with hearing impairment identified by school entry hearing screening has decreased since newborn hearing screening was introduced in the early 2000s. However, cases can be missed or develop after newborn screening.

Focus of the review

This evidence summary reviews the prevalence and type of hearing loss in UK children at school entry age; the accuracy of hearing screening tests and the consequences of school entry hearing screening. It includes studies published up to June 2018. The key questions are:

1. what is the prevalence of hearing loss in children in the UK?
2. what is the accuracy of hearing screening tests, individually or in combination, used in children at school entry age?
3. what are the reported outcomes of school entry hearing screening programmes?

For questions 1 and 3 only peer reviewed studies published in English after March 2006 (after the full implementation of the Newborn Hearing Screening Programme) were eligible for consideration in the review. For question 2, peer reviewed studies published in English after May 2014 (the search date of a recent Health Technology Assessment) were eligible.

Recommendation under review

The UK NSC Child Health Sub-Group has previously recommended that screening for hearing loss in school age children should continue whilst further research was being undertaken.

Findings and gaps in the evidence of this review

Some data are available on the prevalence of permanent hearing impairment in UK children of school entry age. However, areas of uncertainty relate to:

- the prevalence of temporary hearing loss in children at school entry
- the accuracy of screening tests for hearing loss in children at school entry
- a lack of evidence indicating an advantage to screening children at school entry.

Recommendations on screening

The volume, quality and direction of new evidence is insufficient to change the current recommendation about screening for permanent hearing loss in children at school entry.

Limitations

This rapid review process was conducted over a condensed period of time and did not include grey literature sources. Studies not available in the English language, abstracts and poster presentations, were not included. Studies that were not published in peer-reviewed journals were not reviewed.

Evidence uncertainties

The body of evidence concerning the prevalence of hearing impairment in children in and around school entry age and the effectiveness of a screening programme in this population is small. Comparative studies exploring the outcomes in areas with school entry hearing screening programmes with those without screening programmes would help clarify the value of screening.

There is a lack of good quality evidence relating to the performance of screening tests in children of school entry age.

Introduction and approach

This evidence summary reviews screening for permanent hearing loss in children at school entry against selected UK National Screening Committee (NSC) criteria.

Background

Hearing deficits can interfere with a child's speech and language development, communication, behaviour and ability to learn[1]. The impact of hearing loss on a child's development depends on the severity of the hearing loss, whether one or both ears are affected and the age of the child at onset[1]. The impact on spoken language development and other educational outcomes will be greater in children born with hearing loss or who develop hearing loss soon after birth[1].

There are different types of hearing loss. Conductive hearing loss affects the passage of sound between the eardrum and inner ear. This can be temporary (eg due to infection, a build-up of fluid in the middle ear - otitis media with effusion (OME) - or the build-up of earwax), or it can be permanent[1,4]. In sensorineural hearing loss there is permanent damage to the hair cells in the cochlea (the sensory hearing organ) or damage to the hearing neural pathways[1].

The severity of hearing loss is measured in decibels (dB) with different categories for degree of hearing loss. There are several different categorisations for hearing loss. The World Health Organisation categories are[2]:

- slight/ mild: 26-40dB; trouble hearing and understanding soft-speech, speech from a distance or speech in a background of noise
- moderate: 31-60dB (children), 41-60dB (adults); difficulty hearing regular speech, even at close distances. May affect language development, interaction with peers and self-esteem
- severe: 61-80dB; may hear only very loud speech or loud environmental sounds, such as a siren or a door slamming. Most conversational speech is not heard
- profound : >81dB; may perceive loud sounds as vibrations. Speech and language may deteriorate.

The Newborn Hearing Screening Programme categories are based on the better hearing ear average at 0.5, 1, 2 and 4kHz and are[3]:

- mild: 21-39dB
- moderate: 40-69dB
- severe: 70-94dB
- profound: ≥95dB.

In the UK approximately 1 per 1,000 children (about 800 children per year) are born with a permanent bilateral (both ears) hearing impairment of more than 40dB. An additional 0.6 per 1,000 (about 500 children per year) have a unilateral (one ear) hearing impairment[4]. Not all children will have a hearing impairment that can be identified at birth. The UK prevalence of permanent hearing impairment of more than 40dB at 3 years old is 1.07 per 1,000. For children aged 9 to 15 years this is 2.05 per 1,000[4]. Many children will have a temporary hearing impairment at some point during their childhood and about 80% of children will experience an OME (build-up of fluid usually associated with an infection) before they are 6 years old[4].

Behavioural tests (audiometry) are usually used in school entry hearing screening programmes. These require understanding and co-operation from the child and test performance can be affected by the child's ability to perform a task on demand and maintain attention during the test[1]. Some screening tests for hearing impairment do not distinguish between permanent and temporary hearing impairment[4].

School entry hearing screening in the UK was introduced in 1955 and remains in place in many parts of the UK[1]. The introduction of a newborn hearing screening programme in England began in 2002 and was fully implemented by March 2006[3]. Participation in newborn hearing screening has been high since the programme started[3]. In 2016/17 the proportion of eligible babies who had completed screening by the target age was 99.2%[5]. A 2007 Health Technology Assessment (HTA)[6] reported that 1 in 8 services had stopped offering school entry hearing screening since 2005[4].

The number of children with a hearing impairment identified by school entry hearing screening has decreased since newborn hearing screening was introduced[4]. However, cases can be missed or develop after newborn screening[1]. The diagnosis of hearing loss in children between

newborn hearing and school entry, and in older children, is based on parental and professional awareness and follow-up of children who screened negative in newborn screening but were considered at risk[1].

There are conflicting views on the target group of children to be identified in school entry hearing screening. Some suggest that this should be children with a permanent hearing impairment that might benefit from prompt intervention[1]. Others suggest that any hearing loss, regardless of permanence or severity, should be identified so that any intervention can be recommended[1].

Current policy context and previous reviews

Following the introduction of newborn hearing screening, most cases of hearing impairment will have been identified before school entry. However, cases can be missed or develop later. Therefore the UK NSC Child Health Sub-Group recommended that screening for hearing loss in school age children should continue whilst further research was being undertaken[1].

Objectives

The current review aims to look at the prevalence and type of hearing loss in UK children at school entry age; the accuracy of hearing screening tests and the consequences of school entry hearing screening[1].

Table 1. Key questions for the evidence summary, and relationship to UK NSC screening criteria

	Criterion	Key questions	Studies included
	THE CONDITION		
1	The condition should be an important health problem as judged by its frequency and/or severity. The epidemiology, incidence, prevalence and natural history of the condition should be understood, including development from latent to declared disease and/or there should be robust evidence about the association between the risk or disease marker and serious or treatable disease.	1. What is the prevalence of hearing loss in children in the UK?	1
	THE TEST		
4	There should be a simple, safe, precise and validated screening test.	2. What is the accuracy of hearing screening tests, individually or in combination, used in children at school entry age?	1
	THE SCREENING PROGRAMME		
11	There should be evidence from high quality randomised controlled trials that the screening programme is effective in reducing mortality or morbidity. Where screening is aimed solely at providing information to allow the person being screened to make an "informed choice" (eg. Down's syndrome, cystic fibrosis carrier screening), there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened.	3. What are the reported outcomes of school entry hearing screening programmes?	2

The studies included in questions 2 and 3 came from the same HTA[4].

Methods

The current review was conducted by Solutions for Public Health (SPH), in keeping with the UK National Screening Committee evidence review process. Database searches were conducted on 20th June 2018 to identify studies relevant to the questions detailed in

Table 1.

Eligibility for inclusion in the review

The following review process was followed:

1. each abstract was reviewed against the inclusion/exclusion criteria by 1 reviewer. Where the applicability of the inclusion criteria was unclear, the article was included at this stage in order to ensure that all potentially relevant studies were captured.
2. full-text articles required for the full-text review stage were acquired.
3. each full-text article was reviewed against the inclusion/exclusion criteria by 1 reviewer, who determined whether the article was relevant to 1 or more of the review questions.
4. any queries at the abstract or full-text stage were resolved through discussion with a second reviewer.
5. the review was quality assured by a second senior reviewer in accordance with SPH's quality assurance process.

Eligibility criteria for each question are presented in Table 2 below. For questions 1 and 3 only peer reviewed studies published in English after March 2006 (after the full implementation of the Newborn Hearing Screening Programme) were eligible for consideration in the review. For question 2, peer reviewed studies published in English after May 2014 (the search date of a recent Health Technology Assessment) were eligible for consideration in the review.

A total of 1,646 unique references were identified and sifted by an information scientist by title and abstract for potential relevance to the review. An SPH reviewer assessed 74 titles and abstracts for further appraisal and possible inclusion in the final review.

Overall, 22 studies were identified as possibly relevant during title and abstract sifting and further assessed at full text (see Appendix 2 for study flow).

Table 2. Inclusion and exclusion criteria for the key questions

Key question	Inclusion criteria							Exclusion criteria
	Population	Target condition	Intervention	Reference Standard	Comparator	Outcome	Study type	
1. What is the prevalence of hearing loss in children in the UK?	Children at school entry age (4-6 years of age) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis	Hearing loss	N/a	N/a	N/a	Temporary or permanent conductive or sensorineural hearing loss of different degrees	Cross-sectional studies, cohort studies, systematic reviews of these	Non-UK studies
2. What is the accuracy of hearing screening tests, individually or in combination, used in children at school entry age?	Children at school entry age (4-6 years) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis	Hearing loss	Audiometry test (eg pure-tone screen (PTS) and HearCheck (HC) screener); transient or distortion product otoacoustic emissions or auditory evoked response tests	Pure-tone audiometry (PTA)	N/a	Diagnostic test accuracy: sensitivity, specificity, false positive rate, false negative rate, positive predictive value, negative predictive value	Consecutively enrolled or randomly assigned populations in systematic reviews, meta-analyses, cross-sectional studies, validation studies, prospective or	Case reports, case series, non-systematic reviews, case control studies, non-peer reviewed literature

							retrospective cohorts	
3. What are the reported outcomes of school entry hearing screening programmes?	Children at school entry age (4-6 years) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis	Hearing loss	School entry hearing screening programme	N/a	Any	Any	RCTs, prospective population based studies and systematic reviews of these	Non UK studies or studies in countries not analogous to the UK

Appraisal for quality/risk of bias tool

The following tools were used to assess the quality and risk of bias of each study included in the review:

- systematic reviews: Critical Appraisal Skills Programme (CASP) Systematic Review Checklist
- cohort studies: Critical Appraisal Skills Programme (CASP) Cohort Study Checklist

Databases/sources searched

A systematic search of 3 databases (Medline, Embase and Cochrane) was conducted on 20th June 2018 to identify studies relevant to the questions detailed in Table 1. The search strategy is presented in Appendix 1.

Question level synthesis

Criterion 1 — prevalence of hearing loss in children in the UK

The condition should be an important health problem as judged by its frequency and/or severity. The epidemiology, incidence, prevalence and natural history of the condition should be understood, including development from latent to declared disease and/or there should be robust evidence about the association between the risk or disease marker and serious or treatable disease.

Question 1 — What is the prevalence of hearing loss in children in the UK?

Sub-question — What is the prevalence of temporary (transient) conductive, permanent conductive and sensorineural hearing loss in children in the UK?

The population for this question excludes children who are already known to have a hearing impairment and children in high risk groups.

Previous studies have estimated that approximately 1 per 1,000 children in the UK are born with a permanent bilateral hearing impairment of more than 40dB. An additional 0.6 per 1,000 have a unilateral hearing impairment[4]. At 3 years old the prevalence of permanent hearing impairment of more than 40dB was 1.07 per 1,000 and at 9 to 15 years old this was 2.05 per 1,000[4].

Eligibility for inclusion in the review

- population — children at school entry age (4-6 years of age) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis
- intervention — N/a
- comparator — N/a
- outcomes — temporary or permanent conductive or sensorineural hearing loss of different degrees

- study design — cross-sectional studies, cohort studies, systematic reviews of these
- date and language — studies published in English after March 2006.

Description of the evidence

Database searches yielded 74 results, of which 9 were judged to be relevant to this question and 4 abstracts met the criteria for full text review. After review of the full texts, 1 study was included.

Reasons for excluding studies after review of the full text were:

- the study population did not match the population of interest to the review (2 studies)
- a paper duplicating the results from a cohort of children reported in a study that has been included (1 study).

Summary of findings

A study-level summary of data extracted from each included publication is presented in the appraisal of individual studies in Appendix 3. In Appendix 3 publications are stratified by question.

One large UK study (n=35,668), reported data about the prevalence of hearing impairment in children in the first year of primary school (Watkin and Baldwin 2011[7]). The study was conducted in North-East London (Waltham Forest), in an area where both newborn and school entry hearing screening was performed. The precise age of the children screened at school entry was not reported but in the UK children usually start primary school at the age of 4.

In the first year of primary school, 130 children had some form of permanent hearing impairment. This includes high risk groups such as children who developed a hearing impairment following an infection. The type of hearing impairment is provided in Table 3.

Table 3. Type of hearing impairment identified in 130 children in the first year of primary school

Hearing impairment	Number of children	Prevalence per 1,000 (95%CI)
Any bilateral or unilateral congenital, late onset or acquired permanent impairment of any degree	130	3.64 (3.02 to 4.27)
Moderate or worse bilateral permanent hearing impairment (≥40dB)	54	1.51 (1.11 to 1.92)
Mild bilateral permanent hearing impairment (20-39dB)	47	1.32 (0.94 to 1.69)
Unilateral permanent hearing impairment (≥20dB)	29	0.81 (0.52 to 1.11)

Of the 130 cases, 64 were identified through newborn screening (1.79 per 1,000; 95%CI 1.36 to 2.23) with 66 (1.86 per 1,000[*]) identified after newborn screening. This included 20 who moved into the area after newborn screening and 20 with late onset hearing impairment. The route leading to the identification of a permanent hearing impairment was reported for 57 of these 66 children (9 children who had moved into the area were excluded from this analysis due to incomplete histories):

- 44 were identified following concerns about their hearing
- 11 children were detected by school entry screening
- 2 children were identified by the health visitor distraction test.

The type of hearing impairment identified for the 11 children detected by school entry screening is provided in Table 4.

Table 4. Type of hearing impairment identified in 11 children detected by the school entry test

Hearing impairment	Number of children	Prevalence per 1,000 (95%CI)
Any severity	11	0.31 (0.13 to 0.49)
Moderate or worse bilateral permanent hearing impairment (≥40dB kHz)	3	0.08 (0.00 to 0.18)
Mild bilateral permanent hearing impairment (20-39 kHz)	4	0.11 (0.00 to 0.22)
Unilateral permanent hearing impairment	4	0.11 (0.00 to 0.22)

The study was appraised using the CASP checklist for cohort studies. There were no concerns about the conduct of the study or the reporting of the results. This was a large UK study including 35,668 children and the results are applicable to the current UK context in that the children were

[*] Calculated by SPH

born over a 10 year period in an area where newborn hearing screening had been introduced and school entry screening was also performed. However, it is uncertain whether the prevalence observed in this one area of North-East London would be generalisable to the UK as a whole.

Summary of Findings Relevant to Criterion 1: Criterion not met†

One study reported the prevalence of permanent hearing loss in UK children in the first year of primary school as 3.64 per 1,000 (95%CI 3.02 to 4.27). This figure was broken down by severity of the hearing impairment and whether it was bilateral or unilateral. The prevalence of permanent hearing loss of more than 40dB was 1.51 per 1,000 (95%CI 1.11 to 1.92). The proportion of hearing loss that was conductive or sensorineural was not reported. Details of how the hearing impairment was identified were reported and revealed that 11 of the 130 cases had been detected by school entry screening. The prevalence of permanent hearing loss detected by school entry screening was 0.31 per 1,000 (95%CI 0.13 to 0.49).

No UK studies reporting the prevalence of temporary hearing impairment were identified.

Comparison with previously reported prevalence of hearing loss in UK children is complicated by differences in the ages of the children for the prevalence figures reported.

Only 1 study was identified from 1 region of the UK. This study included a large sample, is of good quality and the results are applicable to the current UK screening context where newborn hearing screening is in place. However, no prevalence figure for temporary hearing impairment was identified and it is not clear if the prevalence of permanent hearing loss in this area of North-East London is generalisable to the rest of the UK. Due to this uncertainty, this criterion is not met.

† **Met** -for example, this should be applied in circumstances in which there is a sufficient volume of evidence of sufficient quality to judge an outcome or effect which is unlikely to be changed by further research or systematic review.
Not Met - for example, this should be applied in circumstances where there is insufficient evidence to clearly judge an outcome or effect or where there is sufficient evidence of poor performance.
Uncertain -for example, this should be applied in circumstances in which the constraints of an evidence summary prevent a reliable answer to the question. An example of this may be when the need for a systematic review and meta-analysis is identified by the rapid review.

Criterion 4 — accuracy of hearing screening tests used in children at school entry age

There should be a simple, safe, precise and validated screening test.

Question 2 — What is the accuracy of hearing screening tests, individually or in combination, used in children at school entry age?

Eligibility for inclusion in the review

- population — children at school entry age (4-6 years of age) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis
- intervention — audiometry test (eg pure-tone screen (PTS) and HearCheck (HC) screener); transient or distortion product otoacoustic emissions or auditory evoked response tests
- reference standard — pure-tone audiometry (PTA)
- outcomes — diagnostic test accuracy: sensitivity, specificity, false positive rate, false negative rate, positive predictive value (PPV), negative predictive value (NPV)
- study design — consecutively enrolled or randomly assigned populations in systematic reviews, meta-analyses, cross-sectional studies, validation studies, prospective or retrospective cohorts
- date and language — studies published in English after May 2014.

Description of the evidence

Database searches yielded 74 results, of which 44 were judged to be relevant to this question and 10 abstracts met the criteria for full text review. After review of the full texts, 1 systematic review, published within an HTA, was included.

Reasons for excluding studies after review of the full text were:

- the study population did not match the population of interest to the review (4 studies)
- a more recent systematic review was available (2 studies)
- the study was included in the HTA systematic review (1 study)

- the study did not report any test performance outcomes (1 study)
- a commentary/ discussion paper (1 study).

Summary of findings

A study-level summary of data extracted from each included publication is presented in the summary and appraisal of individual studies in Appendix 3. In Appendix 3 publications are stratified by question.

A 2016 HTA (Fortnum et al 2016)[4] included a systematic review on the diagnostic accuracy of screening tests used to identify hearing impairment at or around school entry. As this was an update of a systematic review performed within a 2007 HTA[6], the authors searched for studies published between January 2005 and July 2014. The search identified 10 studies including 2,566 children and covering questionnaires, audiometry, transient-evoked otoacoustic emissions and automated auditory brainstem response. In assessing the performance of screening tests Fortnum et al (2016)[4] considered the identification of any type of hearing impairment as the outcome of interest. The reference standard had to include pure tone audiometry. The results of the included studies are summarised in Table 5 below. There was no pooled analysis; all results in Table 5 are from individual studies[4].

In Table 5 sensitivity, specificity and prevalence were taken from Fortnum et al (2016)[4] and PPV and NPV were calculated for this review by SPH.

Table 5 demonstrates that the sensitivity and specificity scores varied considerably. A number of different questionnaire screening tests, audiometry screening tests and transient-evoked otoacoustic emissions screening tests were identified and these applied a variety of different cut-off values for a positive test. In addition to the variation between the studies included in the review the confidence intervals around the individual estimates of sensitivity and specificity were wide reducing confidence in the results.

All of the studies included in the systematic review were small and the prevalence of hearing impairment in the study populations ranged from 1.77% to 74%. This is considerably higher than the UK prevalence of permanent hearing loss reported in response to question 1 (3.64 per 1,000 ie 0.364%). The figures are not directly comparable because the

systematic review considered any type of hearing impairment and may therefore also include temporary hearing loss. However, the PPV scores calculated for these studies may be higher than would be found if the same sensitivity and specificity scores were applied to a UK population. For example, a sensitivity of 75% and specificity of 98% applied to a prevalence of 5.05% results in a PPV and NPV of 67% and 99%. However, if the prevalence is reduced to 0.364% the NPV is similar but the PPV decreases to 12%‡.

‡ Calculated by SPH

Table 5. Summary of the results from Fortnum et al (2016)[4]

Screening test	N	Prevalence	Screening test cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
Questionnaires							
CHQSC	317	5.05%	Not specified	56% (30 to 80)	60% (54 to 66)	7%	96%
CHQSC	154	9.74%	≥1 to ≥5	Range 0% to 67%	Range 55% to 100%	---	---
			≥1	67% (38 to 88)	55% (46 to 63)	14%	94%
Questionnaire (NS)	735	1.77%	≥1	100% (75 to 100)	75% (71 to 78)	7%	100%
Questionnaire (NS)	214	46.72%	≥6	44% (34 to 54)	87% (79 to 92)	75%	64%
Audiometry[§]							
Siemens HearCheck Navigator	821 (ears)	4.75%	Not specified	23% (11 to 39)	97% (96 to 98)	28%	96%
Home Audiometer	80	12.50%	>40dB (any frequency)	100% (69 to 100)	50% (38 to 62)	22%	100%
Software		11.25%	>40dB (0.5kHz excluded)	78% (40 to 97)	92% (83 to 97)	55%	97%
Smart Hearing	312	5.13%	>30dB at 1,2 or 4 kHz	38% (15 to 65)	93% (89 to 95)	23%	97%
Transient-evoked otoacoustic emissions[**]							
Madsen Celesta 503	317	5.05%	signal to noise ratio values (average 1.5 to 4kHz) of ≥3dB and whole-wave reproducibility of ≥50%	75% (48 to 93)	98% (96 to 99)	67%	99%
ILO 92 recorder	86	11.63%	Spectrum recorded ≥3dB above noise floor and halfway across frequency bands 2-3kHz and 3-4kHz	90% (55 to 100)	64% (53 to 75)	25%	98%
Otodynamics Echo Port ILO 288	135	74%	Response for 3 of 5 frequency range with TEOAE 5dB above noise floor	100% (3 to 100)	94% (89 to 97)	98%	100%
Automated auditory brainstem response[††]							
MB11 BERA-phone®	115	9.57%	Not specified	100% (72 to 100)	94% (88 to 98)	64%	100%

CHQSC – Chinese Hearing Questionnaire for School Children; CI - confidence intervals; dB – decibels; NPV – negative predictive value; NS – not specified; PPV – positive predictive value

[§] Behavioural tests in which a child must indicate if they have heard a sound
[**] Otoacoustic emissions are recorded by a small probe placed in the external ear canal
[††] A neurological test of auditory brainstem function in response to an auditory stimulus

The systematic review was appraised using the CASP checklist for systematic reviews. There were no areas of concern in the conduct of the review. The study authors assessed quality of the individual studies included in the review using the QUADAS tool. Overall, 3 studies were considered to be of moderate quality and 7 of good quality. The study authors identified a number of areas of selection bias and reasons why the studies may have limited applicability to a UK context:

- some studies included children younger than 4-6 years, reflecting the fact that school entry age varies
- 7 studies were conducted in countries with no established universal newborn hearing screening programme
- most studies included small self-selected samples recruited from a single locality and may not be representative of their population
- in 5 studies the reference standard was considered suboptimal
- 5 studies did not report the time period between the index test and reference standard
- blinding of the index test evaluators to the reference standard result was not reported in 3 studies and blinding of the reference standard evaluators to the index test result was not reported in 5 studies.

The 10 included studies were from China, Brazil, Greece, Japan, Kenya the Philippines and the USA.

Fortnum et al (2016)4 noted that several studies assessing the accuracy of the pure-tone screen had been identified in the 2007 HTA6. However, no new studies were identified for inclusion in the 2016 HTA. In the 2007 HTA the sensitivity of the pure-tone screen ranged from 82% to 100% and the specificity from 65% to 99%6.

Summary of Findings Relevant to Criterion 4: Criterion not met‡‡

One systematic review on the diagnostic accuracy of screening tests was identified, which included 10 small studies with a total of 2,566 children. There was a lack of consistency in the results of the included studies, limiting any conclusions that can be drawn about the accuracy of screening tests for children of school entry age. Whilst the systematic review was of good quality, there were some concerns about the quality of the included studies. The applicability of the results to the current UK context is questionable, eg it is uncertain if the prevalence of hearing impairment in the included studies is applicable to the UK and 7 of the 10 studies were conducted in countries where there is no universal newborn screening programme.

This criterion is not met.

‡‡ **Met** -for example, this should be applied in circumstances in which there is a sufficient volume of evidence of sufficient quality to judge an outcome or effect which is unlikely to be changed by further research or systematic review.
Not Met - for example, this should be applied in circumstances where there is insufficient evidence to clearly judge an outcome or effect or where there is sufficient evidence of poor performance.
Uncertain -for example, this should be applied in circumstances in which the constraints of an evidence summary prevent a reliable answer to the question. An example of this may be when the need for a systematic review and meta-analysis is identified by the rapid review.

Criterion 11 — outcomes and potential impact of school entry hearing screening programmes

There should be evidence from high quality randomised controlled trials that the screening programme is effective in reducing mortality or morbidity. Where screening is aimed solely at providing information to allow the person being screened to make an "informed choice" (eg. Down's syndrome, cystic fibrosis carrier screening), there must be evidence from high quality trials that the test accurately measures risk. The information that is provided about the test and its outcome must be of value and readily understood by the individual being screened.

Question 3 — What are the reported outcomes of school entry hearing screening programmes?

Sub-question — What is the impact of a potential false-negative on children and their families?

In this question, no restriction was placed on the type or definition of school entry hearing screening programme eligible for inclusion.

Eligibility for inclusion in the review

- population — children at school entry age (4-6 years of age) excluding those with known hearing impairments and high risk groups such as those with Down's syndrome, cytomegalovirus infection or meningitis
- intervention — school entry hearing screening programme
- comparator — any
- outcomes — any
- study design — RCTs, prospective population based studies and systematic reviews of these
- date and language — studies published in English after May 2014.

Description of the evidence

Database searches yielded 74 results, of which 22 were judged to be relevant to this question and 9 abstracts met the criteria for full text review. After review of the full texts, 1 systematic review and 1 cohort study were included both of which were published within the same HTA[4].

Reasons for excluding studies after review of the full text were:

- the study and did not include a comparator (3 studies)
- the study population did not match the population of interest to the review (3 studies)
- a more relevant paper reporting the same data was available and included (1 study)
- a commentary/ discussion paper (1 study).

Summary of findings

A study-level summary of data extracted from each included publication is presented in the summary and appraisal of individual studies in Appendix 3. In Appendix 3 publications are stratified by question.

One cohort study compared outcomes for children referred for suspected hearing impairment from a UK area that has a school entry screening programme (Nottingham) compared to an area that does not (Cambridge) (Fortnum et al 2016[4]). The newborn screening programme was fully implemented in both areas. The study reported all referrals made between September 2012 and June 2014, except those identified from newborn hearing screening. The Nottingham audiology service accepted referrals from parents and health professionals (including school entry hearing screening). The Cambridge audiology service received referrals from a variety of professionals from health, education and social services[4].

The results of this study are summarised in Table 6.

Table 6. Summary of Fortnum et al (2016)[4] cohort study

Outcome	Nottingham (screening)	Cambridge (no screening)	Comparison
Number of referrals for assessment (per 1,000 person years)	21.9 (n=1,702)	34.4 (n=1,108)	Rate ratio 0.64 (95%CI 0.59 to 0.69), p<0.001
Mean age at referral (years) for all children	4.70	4.66	Mean difference 0.04 (95%CI -0.04 to 0.11), p=0.37
Mean age at referral (years) for confirmed cases	4.97	4.51	Mean difference 0.47 (95%CI 0.24 to 0.70), p<0.001
Confirmed hearing impairment (per 1,000 person years)	2.51 (17% of children referred; n=195)	3.04 (11% of children referred; n=98)	Rate ratio 0.82 (95%CI 0.64 to 1.06), p=0.12
Level of hearing impairment: left ear average (median; IQR)	35.0dB (26.3 to 41.3)	31.3dB (22.5 to 38.8)	No comparison reported
Level of hearing impairment: right ear average (median; IQR)	32.5dB (22.5 to 40)	31.3dB (23.8 to 37.5)	No comparison reported
Type of hearing impairment			No comparison reported
Bilateral conductive:	70.8%	71.4%	
Unilateral conductive:	20.5%	20.4%	
Bilateral sensorineural:	0.5%	2.0%	
Unilateral sensorineural:	2.6%	1.0%	
Unilateral mixed:	0%	2.0%	
'Normal' binaural[§§]:	3.6%	2.0%	
Incomplete:	2.1%	1.0%	

CI – confidence intervals; dB – decibels; IQR – interquartile range

In Nottingham, 21.5% of referrals came from school hearing screening[4].

This study was appraised using the CASP checklist for cohort studies. This was a retrospective cohort study. This study design introduces the possibility of selection bias in the study population, from the patients included in the analysis or the classification of outcomes from patient records. In this study, hearing impairment was determined by whether a child was referred for further assessment, given a hearing aid or discharged. Although it was stated that both services assessed children's hearing according to UK national and local protocols, it is not clear that the same cut-off levels were used to determine hearing impairment. The proportion of children with mild, moderate or severe hearing impairment was not provided.

[§§] The study authors stated that for 'normal' binaural, absolute values of hearing loss may be >30dB but soundfield testing would indicate that to be 'normal'

The study authors acknowledged possible epidemiological and social differences between the 2 areas. For example, socioeconomic deprivation is higher in Nottingham than Cambridge. It was not possible to adjust the analysis for potential cofounding variables. Hearing impairment included both temporary and conductive hearing impairment. No follow-up of children was done to determine whether the impairment detected was permanent or temporary or any subsequent impact on child development. The cohort included all referrals except those identified from newborn hearing screening and is therefore likely to include children from high risk groups such as Down's syndrome, cytomegalovirus infection or meningitis. The results may not be generalisable to other areas of the UK.

One systematic review (Fortnum et al 2016[4]) searched for studies on the impact of a potential false-negative screening result on children and their families. A false negative screening result happens when a child who passed the screening test does in fact have a hearing impairment. The systematic review searched for papers published up to May 2014. No studies were identified.

The review was assessed using the CASP checklist for systematic reviews. There was a lack of details about any language restrictions applied in the search but otherwise there were no concerns.

Summary of Findings Relevant to Criterion 11: Criterion not met***

One study reported outcomes comparing an area with school entry screening to an area with no screening. There was no significant difference in the yield of confirmed cases of hearing impairment between an area where school entry screening was in place and an area where it was not. The number of referrals for assessment was higher in the area without school entry screening (rate ratio 0.64 95%CI 0.59 to 0.69, p<0.001), however, there was no significant difference in the mean age at referral. There were some concerns about the quality of the study and in the assessment of hearing impairment. The applicability of the results of this study to the UK as a whole is unclear.

No studies were identified assessing the potential impact of a false negative screening test.

Due to the limited evidence identified and the concerns about quality this criterion is not met.

*** **Met** -for example, this should be applied in circumstances in which there is a sufficient volume of evidence of sufficient quality to judge an outcome or effect which is unlikely to be changed by further research or systematic review.
Not Met - for example, this should be applied in circumstances where there is insufficient evidence to clearly judge an outcome or effect or where there is sufficient evidence of poor performance.
Uncertain -for example, this should be applied in circumstances in which the constraints of an evidence summary prevent a reliable answer to the question. An example of this may be when the need for a systematic review and meta-analysis is identified by the rapid review.

Review summary

Conclusions and implications for policy

This evidence summary reviews screening for permanent hearing loss in children at school entry against selected UK NSC criteria for appraising the viability, effectiveness and appropriateness of a screening programme.

The volume, quality and direction of new evidence is insufficient to change the current recommendation about screening for permanent hearing loss in children at school entry.

Some data are available on the prevalence of permanent hearing impairment in UK children of school entry age. However, areas of uncertainty relate to:

- the prevalence of temporary hearing loss in children at school entry
- the accuracy of screening tests for permanent hearing loss in children at school entry
- a lack of evidence indicating an advantage to screening children at school entry.

Limitations

A limitation for this review is the lack of good quality evidence relating to the performance of screening tests in children of school entry age and comparative studies exploring the outcomes of school entry hearing screening programmes.

This rapid review process was conducted over a condensed period of time (approximately 12 weeks). Searching was limited to 3 bibliographic databases and did not include grey literature sources. The review was guided by a protocol developed a priori. The literature search and first appraisal of search results were undertaken by 1 information scientist, and further appraisal and study selection by 1 reviewer. Any queries at both stages were resolved through discussion with a second reviewer. Studies not available in the English language, abstracts and poster presentations, were not included. Studies that were not published in peer-reviewed journals were not reviewed.

Appendix 1 — Search strategy

Electronic databases

The search strategy included searches of the databases shown in Table 7.

Table 7. Summary of electronic database searches and dates

Database	Platform	Searched on date	Date range of search
MEDLINE	Ovid SP	20th June 2018	2006 to Present (Q1,Q3) 2014 to Present (Q2)
Embase	Ovid SP	20th June 2018	2006 to Present (Q1,Q3) 2014 to Present (Q2)
The Cochrane Library	Wiley Online	20th June 2018	2006 to Present (Q1,Q3) 2014 to Present (Q2)

Search Terms

Search terms for MEDLINE are shown in Table 8. A similar search was conducted for Embase. Search terms for the Cochrane Library databases are shown in Table 9.

Table 8. Search strategy for MEDLINE

#	Search terms	Results
Question 1		
1	exp Hearing Loss/	64117
2	((loss or losing or lose) adj3 hearing).ti,ab.	49151
3	deaf*.ti,ab.	33952
4	(sensorineural adj3 loss).ti,ab.	11019
5	(conductive adj3 loss).ti,ab.	2592
6	((snhl or chl) and hearing).ti,ab.	1384
7	1 or 2 or 3 or 4 or 5 or 6	97101
8	prevalence/	254782
9	prevalence.ti,ab. or epidemiolog*.ti.	625652
10	Cross-Sectional Studies/	270187
11	(crosssectional or cross-sectional).ti,ab.	281921
12	8 or 9 or 10 or 11	990410
13	child/ or child, preschool/	1777686
14	(child* or schoolchild* or preschool* or pre-school* or girl* or boy* or pediatric* or paediatric*).ti,ab.	1502979
15	13 or 14	2334867

16	exp United Kingdom/	345572
17	(united kingdom or uk or britain or gb or england or wales or scotland or northern ireland or nhs*).ti,ab,in.	1415422
18	16 or 17	1602480
19	7 and 12 and 15 and 18	205
20	limit 19 to (english language and yr="2006 -Current")	121
Question 2		
1	exp Hearing Loss/	64117
2	((loss or losing or lose or impair*) adj3 hearing).ti,ab.	49151
3	deaf*.ti,ab.	33952
4	(sensorineural adj3 loss).ti,ab.	11019
5	(conductive adj3 loss).ti,ab.	2592
6	((snhl or chl) and hearing).ti,ab.	1384
7	1 or 2 or 3 or 4 or 5 or 6	97101
8	child/ or child, preschool/	1777686
9	(child* or schoolchild* or preschool* or pre-school* or girl* or boy* or pediatric* or paediatric*).ti,ab.	1502979
10	8 or 9	2334867
11	exp Hearing Tests/	44323
12	exp Hearing Loss/di [Diagnosis]	13487
13	((hearing or auditor* or acoustic* or otoacoustic*) adj3 (screen* or test* or diagnos*)).ti,ab.	13439
14	audiometr*.ti,ab.	12817
15	(pure tone adj2 (test* or screen*)).ti,ab.	474
16	hearcheck.ti,ab.	4
17	otoacoustic emission*.ti,ab.	4935
18	Mass Screening/	94378
19	(hearing and (test* or screen* or diagnos*)).ti.	3666
20	11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19	156424
21	7 and 10 and 20	10151
22	limit 21 to (english language and yr="2014 -Current")	1553
23	Developing Countries/	70350
24	(Africa or Caribbean or West Indies or South America or Latin America or Central America).hw,ti,ab,cp.	176474
25	(Afghanistan or Albania or Algeria or Angola or American Samoa or Armenia or Armenian or Azerbaijan or Bangladesh or Benin or Byelarus or Byelorussian or Belarus or Belorussian or Belorussia or Belize or Bhutan or Bolivia or Bosnia or Herzegovina or Hercegovina or Botswana or Brazil or Brasil or Bulgaria or Burkina Faso or Burkina Fasso or Upper Volta or Burundi or Urundi or Cambodia or Khmer Republic or Kampuchea or Cameroon or Cameroons or Cameron or Camerons or Cape Verde or Central African Republic or Chad or China or Colombia or Comoros or Comoro Islands or Comores or Mayotte or Congo or Zaire or Costa Rica or Cote d'Ivoire or Ivory Coast or Cuba or Djibouti or French Somaliland or Dominica or Dominican Republic or East Timor or East Timur or Timor Leste or Ecuador or Egypt or United Arab Republic or El Salvador or Eritrea or Ethiopia or Fiji or Gabon or Gabonese Republic or Gambia or Gaza or Georgia Republic or Georgian Republic or Ghana or Gold Coast or Grenada or Guatemala or Guinea or Guinea-Bisau or Guam or Guiana or Guyana or Haiti or Honduras or India or Maldives or Indonesia or Iran or Iraq or Jamaica or Jordan or Kazakhstan or Kazakh or Kenya or Kiribati or Korea or Kosovo or Kyrgyzstan or Kirghizia or Kyrgyz Republic or Kirghiz or Kirgizstan or Lao PDR or Laos or Lebanon or Lesotho or Basutoland or Liberia or Libya or Macedonia or Madagascar or Malagasy Republic or Malaysia or Malaya or Malay or Sabah or Sarawak or Malawi or	2341506

	Nyasaland or Mali or Marshall Islands or Mauritania or Mauritius or Agalega Islands or Mexico or Micronesia or Middle East or Moldova or Moldovia or Moldovian or Mongolia or Montenegro or Morocco or Ifni or Mozambique or Myanmar or Myanma or Burma or Namibia or Nepal or Netherlands Antilles or Nicaragua or Niger or Nigeria or Pakistan or Palau or Palestine or Panama or Papua New Guinea or Paraguay or Peru or Philippines or Philipines or Phillipines or Phillippines or Romania or Rumania or Roumania or Rwanda or Ruanda or Saint Lucia or St Lucia or Saint Vincent or St Vincent or Grenadines or Samoa or Samoan Islands or Navigator Island or Navigator Islands or Sao Tome or Senegal or Serbia or Sierra Leone or Sri Lanka or Ceylon or Solomon Islands or Somalia or Sudan or Suriname or Surinam or Swaziland or Syria or Principe or South Sudan or Tajikistan or Tadzhikistan or Tadjikistan or Tadzhik or Tanzania or Thailand or Timor-Leste or Togo or Togolese Republic or Tonga or Tunisia or Turkey or Turkmenistan or Turkmen or Tuvalu or Uganda or Ukraine or Uzbekistan or Uzbek or Vanuatu or New Hebrides or Vietnam or Viet Nam or West Bank or Yemen or Zambia or Zimbabwe or Rhodesia).hw,ti,ab,cp.	
26	((developing or less* developed or under developed or underdeveloped or middle income or low* income or underserved or under served or deprived or poor*) adj (countr* or nation? or state? or population? or world)).ti,ab.	84828
27	((developing or less* developed or under developed or underdeveloped or middle income or low* income) adj (economy or economies)).ti,ab.	435
28	(low* adj (gdp or gnp or gross domestic or gross national)).ti,ab.	218
29	(low adj3 middle adj3 countr*).ti,ab.	10767
30	(lmic or lmics or third world or lami countr*).ti,ab.	5612
31	transitional countr*.ti,ab.	144
32	23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31	2501509
33	22 not 32	1251
Question 3		
1	exp Hearing Loss/	64127
2	((loss or losing or lose or impair*) adj3 hearing).ti,ab.	49197
3	deaf*.ti,ab.	33963
4	(sensorineural adj3 loss).ti,ab.	11025
5	(conductive adj3 loss).ti,ab.	2596
6	((snhl or chl) and hearing).ti,ab.	1388
7	1 or 2 or 3 or 4 or 5 or 6	97159
8	child/ or child, preschool/	1778166
9	(child* or schoolchild* or preschool* or pre-school* or girl* or boy* or pediatric* or paediatric*).ti,ab.	1504305
10	8 or 9	2336333
11	exp Hearing Tests/	44326
12	exp Hearing Loss/di [Diagnosis]	13488
13	((hearing or auditor* or acoustic* or otoacoustic*) adj3 (screen* or test* or diagnos*)).ti,ab.	13450
14	audiometr*.ti,ab.	12825
15	(pure tone adj2 (test* or screen*)).ti,ab.	474
16	hearcheck.ti,ab.	4
17	otoacoustic emission*.ti,ab.	4936
18	Mass Screening/	94392
19	(hearing and (test* or screen* or diagnos*)).ti.	3669
20	11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19	156458
21	schools/ or schools, nursery/	34416
22	(school* or preschool* or preschool*).ti,ab.	270006

23	21 or 22	275980
24	7 and 10 and 20 and 23	950
25	limit 24 to (english language and yr="2006 -Current")	376
26	Developing Countries/	70367
27	(Africa or Caribbean or West Indies or South America or Latin America or Central America).hw,ti,ab,cp.	176615
28	(Afghanistan or Albania or Algeria or Angola or American Samoa or Armenia or Armenian or Azerbaijan or Bangladesh or Benin or Byelarus or Byelorussian or Belarus or Belorussian or Belorussia or Belize or Bhutan or Bolivia or Bosnia or Herzegovina or Hercegovina or Botswana or Brazil or Brasil or Bulgaria or Burkina Faso or Burkina Fasso or Upper Volta or Burundi or Urundi or Cambodia or Khmer Republic or Kampuchea or Cameroon or Cameroons or Cameron or Camerons or Cape Verde or Central African Republic or Chad or China or Colombia or Comoros or Comoro Islands or Comores or Mayotte or Congo or Zaire or Costa Rica or Cote d'Ivoire or Ivory Coast or Cuba or Djibouti or French Somaliland or Dominica or Dominican Republic or East Timor or East Timur or Timor Leste or Ecuador or Egypt or United Arab Republic or El Salvador or Eritrea or Ethiopia or Fiji or Gabon or Gabonese Republic or Gambia or Gaza or Georgia Republic or Georgian Republic or Ghana or Gold Coast or Grenada or Guatemala or Guinea or Guinea-Bisau or Guam or Guiana or Guyana or Haiti or Honduras or India or Maldives or Indonesia or Iran or Iraq or Jamaica or Jordan or Kazakhstan or Kazakh or Kenya or Kiribati or Korea or Kosovo or Kyrgyzstan or Kirghizia or Kyrgyz Republic or Kirghiz or Kirgizstan or Lao PDR or Laos or Lebanon or Lesotho or Basutoland or Liberia or Libya or Macedonia or Madagascar or Malagasy Republic or Malaysia or Malaya or Malay or Sabah or Sarawak or Malawi or Nyasaland or Mali or Marshall Islands or Mauritania or Mauritius or Agalega Islands or Mexico or Micronesia or Middle East or Moldova or Moldovia or Moldovian or Mongolia or Montenegro or Morocco or Ifni or Mozambique or Myanmar or Myanma or Burma or Namibia or Nepal or Netherlands Antilles or Nicaragua or Niger or Nigeria or Pakistan or Palau or Palestine or Panama or Papua New Guinea or Paraguay or Peru or Philippines or Philipines or Phillipines or Phillippines or Romania or Rumania or Roumania or Rwanda or Ruanda or Saint Lucia or St Lucia or Saint Vincent or St Vincent or Grenadines or Samoa or Samoan Islands or Navigator Island or Navigator Islands or Sao Tome or Senegal or Serbia or Sierra Leone or Sri Lanka or Ceylon or Solomon Islands or Somalia or Sudan or Suriname or Surinam or Swaziland or Syria or Principe or South Sudan or Tajikistan or Tadzhikistan or Tadjikistan or Tadzhik or Tanzania or Thailand or Timor-Leste or Togo or Togolese Republic or Tonga or Tunisia or Turkey or Turkmenistan or Turkmen or Tuvalu or Uganda or Ukraine or Uzbekistan or Uzbek or Vanuatu or New Hebrides or Vietnam or Viet Nam or West Bank or Yemen or Zambia or Zimbabwe or Rhodesia).hw,ti,ab,cp.	2343798
29	((developing or less* developed or under developed or underdeveloped or middle income or low* income or underserved or under served or deprived or poor*) adj (countr* or nation? or state? or population? or world)).ti,ab.	84920
30	((developing or less* developed or under developed or underdeveloped or middle income or low* income) adj (economy or economies)).ti,ab.	436
31	(low* adj (gdp or gnp or gross domestic or gross national)).ti,ab.	218
32	(low adj3 middle adj3 countr*).ti,ab.	10809
33	(lmic or lmics or third world or lami countr*).ti,ab.	5628

34	transitional countr*.ti,ab.	144
35	26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34	2503915
36	25 not 35	274

Table 9. Search strategy for the Cochrane Library Databases

#	Search terms
#1	((loss or losing or lose or impair*) near/3 hearing):ti,ab,kw (Word variations have been searched)
#2	screen* or diagnos* or test*:ti,ab,kw (Word variations have been searched)
#3	school* or preschool* or preschool*:ti,ab,kw (Word variations have been searched)
#4	1 and #2 and #3

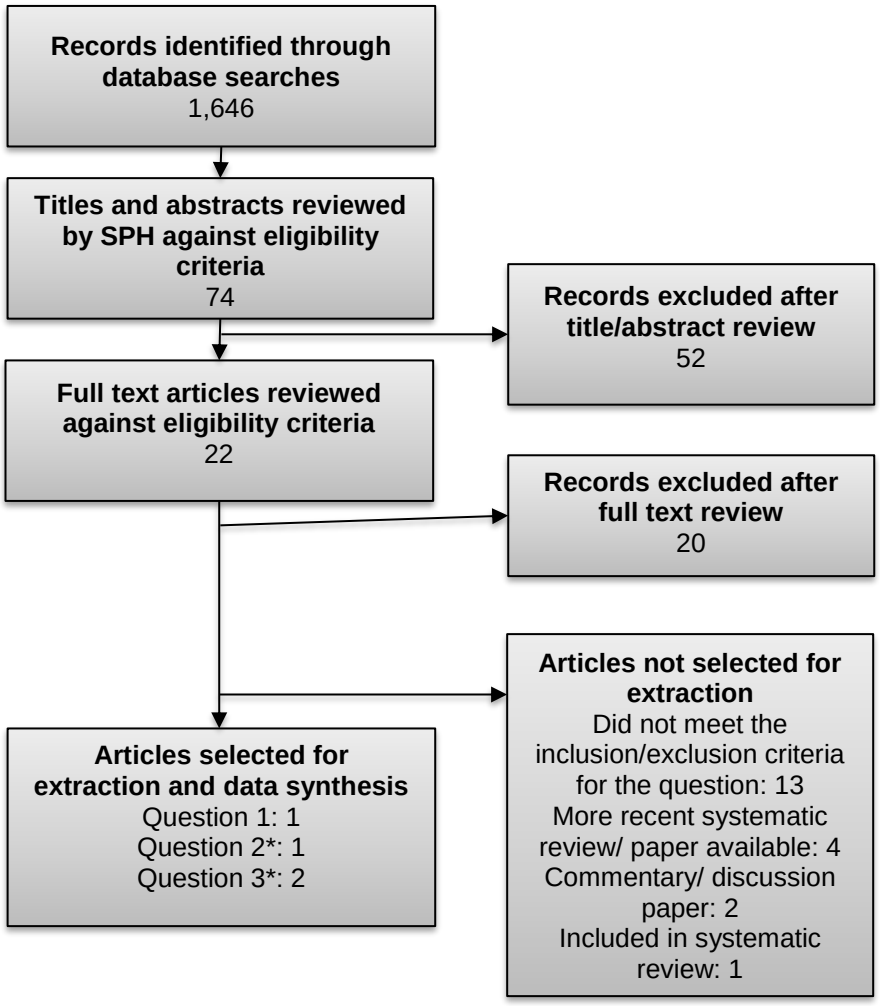
Duplicate references were removed.

Appendix 2 — Included and excluded studies

PRISMA flowchart

Figure 1 summarises the volume of publications included and excluded at each stage of the review. Twenty-two publications were ultimately judged to be relevant to 1 or more review questions and were considered for extraction. Publications that were included or excluded after the review of full-text articles are detailed below.

Figure 1. Summary of publications included and excluded at each stage of the review



*The studies included for questions 2 and 3 came from the same HTA[4].

Publications included after review of full-text articles

The 2 publications included after review of full texts are summarised in Table 10. Studies meeting the inclusion/ exclusion criteria for each individual question were included.

Table 10. Summary of publications included after review of full text articles, and the criteria each publication was identified as being relevant to

Study	The condition	The test	The screening programme	Comments
Watkin & Baldwin (2011)[7]	X			
Fortnum et al (2016)[4]		X	X	HTA

It was planned *a priori* that if a high number of studies met the inclusion/ exclusion criteria for each question, studies would be prioritised for extraction and data synthesis using the following approach:

1. systematic reviews and meta-analyses would be considered the highest quality of evidence if any were found
2. If multiple systematic reviews were identified , the most applicable and recent would be used
3. studies included in a systematic review (that was included) will not be separately reported
4. studies published after the search date of systematic reviews would also be included
5. Higher quality studies eg randomised controlled trials would be prioritised above lower quality studies eg uncontrolled studies
6. UK studies or studies in countries analogous to the UK (questions 1 and 3)

Publications not selected for extraction and data synthesis are clearly detailed in Table 11 below.

Publications excluded after review of full-text articles

Of the 22 publications included after the review of titles and abstracts, 20 were ultimately judged not to be relevant to this review. These publications, along with reasons for exclusion, are listed in Table 11.

Table 11. Publications excluded after review of full-text articles

Reference	Reason for exclusion
Pitt-Byrne T. Irish School Entry Screening referral trends and cohort comparison with preschool specialist referrals. International Journal of Audiology. 2018:1-9	Does not meet inclusion/exclusion criteria – no comparator
Kelly EA, Li B, Adams ME. Diagnostic Accuracy of Tuning Fork Tests for Hearing Loss: A Systematic Review. Otolaryngology - Head & Neck Surgery, 2018:194599818770405	Does not meet inclusion/exclusion criteria - population
Hall JW. Effective And Efficient Pre-School Hearing Screening: Essential For Successful Early Hearing Detection And Intervention (EHDI). JRHDI: Journal of Early Hearing Detection and Intervention. 2016;1(1):2-12	Commentary/ discussion paper
Bargen GA. Chirp-Evoked Auditory Brainstem Response in Children: A Review. American Journal of Audiology, 2015;24(4):573-83	Does not meet inclusion/exclusion criteria - population
Prieve BA, Schooling T, Venediktov R, Franceschini N. An Evidence-Based Systematic Review on the Diagnostic Accuracy of Hearing Screening Instruments for Preschool- and School-Age Children. American Journal of Audiology, 2015;24(2):250-67	More recent systematic review of diagnostic tests available
Serpanos YC, Senzer D, Renne B, Langer R, Hoffman R. The Efficacy of Routine Screening for High-Frequency Hearing Loss in Adults and Children. American Journal of Audiology, 2015;24(3):377-83	Does not meet inclusion/exclusion criteria - population
Dodd-Murphy J, Murphy W, Bess FH. Accuracy of school screenings in the identification of minimal sensorineural hearing loss. American Journal of Audiology, 2014;23(4):365-73	Does not meet inclusion/exclusion criteria – population
Lu J, Huang Z, Ma Y, Li Y, Mei L, Yao G, et al. Comparison between hearing screening-detected cases and sporadic cases of delayed-onset hearing loss in preschool-age children. International Journal of Audiology 2014;53(4):229-34	Does not meet inclusion/exclusion criteria – no comparator
Munoz K, Caballero A, White K. Effectiveness of questionnaires for screening hearing of school-age children: a comprehensive literature review. International Journal of Audiology, 2014;53(12):910-4	More recent systematic review of diagnostic tests available

Swanepoel de W, Eikelboom RH, Margolis RH. Tympanometry screening criteria in children ages 5-7 yr. Journal of the American Academy of Audiology, 2014;25(10):927-36	Does not meet inclusion/exclusion criteria – no test performance outcomes
Wu W, Lu J, Li Y, Kam AC, Fai Tong MC, Huang Z, et al. A new hearing screening system for preschool children. International Journal of Pediatric Otorhinolaryngology, 2014;78(2):290-5	Included in the 2016 HTA systematic review
Wood SA, Davis AC, Sutton GJ. Effectiveness of targeted surveillance to identify moderate to profound permanent childhood hearing impairment in babies with risk factors who pass newborn screening. International Journal of Audiology, 2013;52(6):394-9	Does not meet inclusion/exclusion criteria – population
Watkin P, Baldwin M. The longitudinal follow up of a universal neonatal hearing screen: the implications for confirming deafness in childhood. International Journal of Audiology, 2012;51(7):519-28	More relevant paper reporting same data available
Fitzpatrick EM, Crawford L, Ni A, Durieux-Smith A. A descriptive analysis of language and speech skills in 4- to 5-yr-old children with hearing loss. Ear & Hearing 2011 Sep-Oct;32(5):605-16	Does not meet inclusion/exclusion criteria – population
Bajaj Y, Sirimanna T, Albert DM, Qadir P, Jenkins L, Cortina-Borja M, et al. Causes of deafness in by consanguinity British Bangladeshi children: a prevalence twice that of the UK population cannot be accounted for alone. Clinical Otolaryngology, 2009;34(2):113-9	Does not meet inclusion/exclusion criteria – population
Bristow K, Fortnum H, Fonseca S, Bamford J. United Kingdom school-entry hearing screening: current practice. Archives of Disease in Childhood 2008 Mar;93(3):232-5	Commentary/ discussion paper
Yoong SY, Spencer NJ. A data collection system to audit post-newborn hearing surveillance programme: problems and possibilities. Child: Care, Health & Development. 2008 Sep;34(5):648-56	Does not meet inclusion/exclusion criteria – population
Bamford J, Fortnum H, Bristow K, Smith J, Vamvakas G. Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen. *Health Technol Assess* 2007;11(32)	More recent systematic review and publications using same data available
Serpanos YC, Jarmel F. Quantitative and qualitative follow-up outcomes from a preschool audiologic screening program: perspectives over a decade. American Journal of Audiology. 2007 Jun;16(1):4-12	Does not meet inclusion/exclusion criteria – no comparator
Sideris I, Glattke TJ. A comparison of two methods of hearing screening in the preschool population. Journal of Communication Disorders 2006 Nov-Dec;39(6):391-401	Does not meet inclusion/exclusion criteria – population

Appendix 3 — Summary and appraisal of individual studies

Data extraction and quality assessment for studies relevant to criterion 1

Key question 1: What is the prevalence of hearing loss in children in the UK?

Table 12. Watkin & Baldwin (2011)[7]

Publication	Watkin PM. Baldwin M. Identifying deafness in early childhood: requirements after the newborn hearing screen. Arch. Dis. Child 2011, 96: 62-66
Study details	Cohort study
Study objectives	Longitudinal follow-up of a cohort of UK children who received universal newborn screening to investigate the need for postnatal identification of hearing impairment
Inclusions	Congenital, late onset and acquired permanent childhood hearing impairment (bilateral or unilateral)
Exclusions	N/a
Population	35,668 children born between 1992 and 2002 in Waltham Forest London, followed-up until they had completed their first year of primary school. 33,860 received newborn hearing screening
Intervention	Universal newborn screening; health visitor distraction test as a universal screen until 1997 and afterwards targeting infants with risk factors; school entry hearing screen throughout the time period
Comparator	N/a
Outcomes	In the first year of primary school, 130 children had unilateral or bilateral congenital, late onset or acquired permanent hearing impairment of any degree (prevalence 3.64 per 1,000; 95%CI 3.02 to 4.27). Of these: • 54 had bilateral permanent hearing impairment ≥40dB kHz (prevalence 1.51 per 1,000; 95%CI 1.11 to 1.92) • 47 had bilateral permanent hearing impairment 20-39 kHz (prevalence 1.32 per 1,000; 95%CI 0.94 to 1.69) • 29 unilateral permanent hearing impairment ≥20dB kHz in worst hearing ear (prevalence 0.81 per 1,000 (95%CI 0.52 to 1.11) These figures include children with any permanent hearing impairment identified by any means between birth and school age and does not exclude high risk groups The study authors report that 64 of the 130 cases were identified by newborn screening (1.79 per 1,000; 95%CI 1.36 to 2.23), with 66 (1.86 per 1,000[10]) identified after newborn screening. This included: • 20 who moved into the area after newborn screening (0.56 per 1,000; 95%CI 0.32 to 0.81)

[10] Calculated by SPH

	• 20 with late onset hearing impairment (0.56 per 1,000 95%CI 0.32 to 0.81) Route leading to the identification of permanent hearing impairment was reported for 57 children identified after newborn screening. 9 children who had moved into the area with unverified histories were excluded from the analysis. Most (n=44) were identified following concerns. 2 children were identified by the health visitor distraction test. The remaining 11 children were identified by the school entry test: • All severity: 11 children (0.31 per 1,000; 95%CI 0.13 to 0.49) • Moderate or worse bilateral (≥40dB kHz): 3 children (0.08 per 1,000; 95%CI 0.00 to 0.18) • Mild bilateral (20-39 kHz): 4 children (0.11 per 1,000; 95%CI 0.00 to 0.22) • Unilateral: 4 children (0.11 per 1,000; 95%CI 0.00 to 0.22)
Quality appraisal	This study was appraised using the CASP checklist for cohort studies. There were no concerns about the sample size, recruitment, assessment or follow-up of the children. The results are applicable to the current UK context. Confidence intervals were provided for all calculations performed by the study authors.

Data extraction and quality assessment for studies relevant to criterion 4

Key question 2: What is the accuracy of hearing screening tests, individually or in combination, used in children at school entry age?

Table 13. Fortnum et al (2016)[4] (systematic review on screening tests)

Publication	Fortnum H. Ukoumunne OC. Hyde C. Taylor RS. Ozolins M. Errington S. Zhelev Z. Pritchard C. Benton C. Moody J. Cocking L. Watson J. Roberts S. A programme of studies including assessment of diagnostic accuracy of school hearing screening tests and a cost-effectiveness model of school entry hearing screening programmes. Health Technology Assessment 2016, 20(36)
Study details	Systematic review of screening tests within a health technology assessment
Study objectives	To assess the diagnostic accuracy of screening tests used to identify hearing impairment at or around school entry
Inclusions	10 studies including 2,566 children published between January 2005 and July 2014 in any language Tests had to be undertaken at a primary school or community setting
Exclusions	Studies with a wide age range that did not report different age categories separately
Population	Children aged 4 to 6 years. Studies that partially covered but slightly exceeded the age range of interest were included
Test	Pure-tone screen (also known as sweep pure-tone audiometry) Single-frequency pure-tone audiometry Transient-evoked otoacoustic emissions (TEOAE) Distortion product otoacoustic emission Questionnaires Otoadmittance tests Tympanometry Reflectometry Speech audiometry Automated auditory brainstem response (AABR)
Reference standard	Any reference standard that included pure-tone audiometry

Outcomes[11]

Questionnaires (4 studies)

Chinese Hearing Questionnaire for School Children (2 studies) with otoscopy, tympanometry and PTA as the reference standard

N	Prevalence (%)	Cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
317	5.05	Not specified	56% (30 to 80)	60% (54 to 66)	7%	96%
154	9.74	≥1 to ≥5	Range 0 to 67%	Range 55% to 100%	---	---
154	9.74	≥1	67% (38 to 88)	55% (46 to 63)	14%	94%

Questionnaire not named (2 studies). Reference standard was examination, otoscopy and PTA in 1 study (n=735) and otoscopy, tympanometry and PTA in the other study (n=214)

N	Prevalence (%)	Cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
735	1.77	≥1	100% (75 to 100)	75% (71 to 78)	7%	100%
214	46.72	≥6	44% (34 to 54)	87% (79 to 92)	75%	64%

No studies pre-specified the cut-off level for a positive test. Review authors reported the best test performance result

Audiometry (3 studies)
The audiometry devices used were the Siemens HearCheck Navigator (n=821 ears); Home Audiometer Software (n=80) and Smart Hearing (n=312)

Reference standard was PTA in 2 studies (n=821 ears and n=80) and otoscopy, tympanometry, PTA and distortion product otoacoustic emissions in 1 study (n=312)

N	Prevalence (%)	Cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
821 (ears)	4.75	dB not specified	23% (11 to 39)	97% (96 to 98)	28%	96%
80	12.5	>40dB any frequency	100% (69 to 100)	50% (38 to 62)	22%	100%
80	11.25	>40dB (0.5kHz excluded)	78% (40 to 97)	92% (83 to 97)	55%	97%
312	5.13	>30dB at 1,2 or 4kHz	38% (15 to 65)	93% (89 to 95)	23%	97%

TEOAE (3 studies)
The reference standard was otoscopy, tympanometry and PTA in 2 studies (n=317 and n=86) and PTA in 1 study (n=135)

[11] Fortnum et al (2016) provided details for sensitivity, specificity and prevalence. PPV and NPV were calculated for this review by SPH

N	Prevalence (%)	Cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
317	5.05	(a)	75% (48 to 93)	98% (96 to 99)	67%	99%
86	11.63	(b)	90% (55 to 100)	64% (53 to 75)	25%	98%
135	74	(c)	100% (3 to 100)	94% (89 to 97)	98%	100%

(a) signal to noise ratio values (an average of 1.5 to 4kHz) of at least 3dB and whole-wave reproducibility of at least 50%
(b) TEOAE spectrum recorded at least 3dB above the noise floor and halfway across the frequency bands of 2-3kHz and 3-4kHz
(c) TEOAE response obtained for 3 of 5 frequency range with TEOAE being 5dB above noise floor

AABR (1 study)
The reference standard was PTA and examination

N	Prevalence (%)	Cut-off	Sensitivity (95%CI)	Specificity (95%CI)	PPV	NPV
115	9.57	Not specified	100% (72 to 100)	94% (88 to 98)	64%	100%

The authors concluded that it was not possible to draw strong conclusions about the performance of individual tests for school entry hearing screening

Quality appraisal

The systematic review component of the HTA was assessed using the CASP checklist for systematic reviews. There were no areas of concern in the conduct of the review. The study authors assessed the individual studies included in the review and discussed a number of areas of potential bias.

No pooled analysis was performed due to significant heterogeneity between the studies.

The 10 included studies were from China, Brazil, Greece, Japan, Kenya the Philippines and the USA.

The studies included in the systematic review were assessed using the QUADAS tool. Overall, 3 studies were considered to be of moderate quality and 7 of good quality. Studies were consistently at low risk of bias in the application of the same reference standard to the whole sample or a random sample. However the study authors identified a number of areas of selection bias and reasons why the studies may have limited applicability to a UK context:

- some studies included children younger than 4-6 years, reflecting the fact that school entry age varies
- seven studies were conducted in countries with no established universal newborn hearing screening programme
- most studies included small self-selected samples recruited from a single locality and may not be representative of their population
- in 5 studies the reference standard was considered suboptimal
- 5 studies did not report the time period between the index test and reference standard
- blinding of the index test evaluators to the reference standard result was not reported in 3 studies and blinding of the reference standard evaluators to the index test result was not reported in 5 studies.

Data extraction and quality assessment for studies relevant to criterion 11

Key question 3: What are the reported outcomes of school entry hearing screening programmes?

Table 14. Fortnum et al (2016)[4] (cohort study)

Publication	Fortnum H. Ukoumunne OC. Hyde C. Taylor RS. Ozolins M. Errington S. Zhelev Z. Pritchard C. Benton C. Moody J. Cocking L. Watson J. Roberts S. A programme of studies including assessment of diagnostic accuracy of school hearing screening tests and a cost-effectiveness model of school entry hearing screening programmes. Health Technology Assessment 2016, 20(36)
Study details	Retrospective cohort study within a health technology assessment
Study objectives	To compare children referred for suspected hearing impairment from a UK area that has a school entry screening programme compared to a service that does not
Inclusions	All referrals to audiology services between September 2012 and June 2014
Exclusions	Children referred from newborn hearing screening
Population	Children aged 3 to 6 years who were referred to audiology services
Intervention	An area where school entry hearing screening is in place (Nottingham)
Comparator	An area with no school entry hearing screening since 1997 (Cambridge)
Outcomes	**Number of referrals for assessment** • Nottingham: 1,702 (21.9 per 1,000 person-years) • Cambridge: 1,108 (34.4 per 1,000 person-years) Significantly higher referral rate in Cambridge (rate ratio 0.64 95%CI 0.59 to 0.69, p<0.001) In Nottingham, 21.5% of referrals came from school hearing screening **Mean age at referral** No significant difference in mean age at referral between sites (4.70 years vs 4.66) (mean difference 0.04 95%CI -0.04 to 0.11, p=0.37) For confirmed cases, mean age at referral was significantly higher in Nottingham (4.97 years vs 4.51) (mean difference 0.47 95%CI 0.24 to 0.70, p<0.001) **Confirmed hearing impairment** Children were considered to have a hearing impairment if the outcome of their last appointment with audiology services was referral for further assessment or treatment (eg to ENT services) or hearing aid. Children discharged at their last appointment were considered to have no hearing impairment. • Nottingham: 195 (2.51 per 1,000 person-years; 17.0% of children referred (95%CI not reported)) • Cambridge: 98 (3.04 per 1,000 person-years; 10.6% (95%CI 8.7 to 12.8) of children referred) There was no significant difference between sites in confirmed hearing impairment yield (rate ratio 0.82 95%CI 0.64 to 1.06, p=0.12) In Nottingham, 30.8% of confirmed cases were from school hearing screening

In Nottingham, 25.5% (95%CI 19.8 to 31.2) of children referred from school entry hearing screening were confirmed to have a hearing impairment. 14.9% (95%CI 12.6 to 17.4) of children referred from other sources had a confirmed hearing impairment

Level of hearing impairment
Left ear average (0.5 to 4 kHz) median (IQR)

- Nottingham: 35.0dB (26.3 to 41.3)
- Cambridge: 31.3dB (22.5 to 38.8)

Right ear average (0.5 to 4 kHz) median (IQR)

- Nottingham: 32.5dB (22.5 to 40.0)
- Cambridge: 31.3dB (23.8 to 37.5)

Type of hearing impairment
Hearing impairment included temporary conductive and permanent sensorineural or conductive. The proportion of hearing loss that was temporary or permanent was not reported

	Nottingham	Cambridge
'Normal' binaural	3.6%	2.0%
Bilateral conductive	70.8%	71.4%
Unilateral conductive	20.5%	20.4%
Bilateral sensorineural	0.5%	2.0%
Unilateral sensorineural	2.6%	1.0%
Unilateral mixed	0%	2.0%
Incomplete	2.1%	1.0%

The authors stated that in the 'normal' binaural outcome, absolute values of hearing loss may be >30dB but soundfield testing would indicate that to be 'normal'

Quality appraisal

This study was appraised using the CASP checklist for cohort studies. The retrospective design of the study introduces the possibility of selection bias in the study population, from the patients included in the analysis or the classification of outcomes from patient records. Hearing impairment was determined by whether a child was referred on, given a hearing aid or discharged. The authors stated that both services assess children's hearing according to UK national and local protocols, however there is some uncertainty about whether the same cut-off levels were used to determine hearing impairment. Average level hearing for hearing impairment was reported, but the proportion of children with mild, moderate or severe hearing impairment was not provided.

The authors acknowledged possible epidemiological and social differences between the 2 areas. For example, socioeconomic deprivation is higher in Nottingham than Cambridge. It was not possible to adjust the analysis for potential cofounding variables.

Hearing impairment included both temporary and conductive hearing impairment. No follow-up of children was done to determine whether the impairment detected was permanent or temporary or any subsequent impact on child development. The cohort included all referrals except those identified from newborn hearing screening and is therefore likely to include

	children from high risk groups such as Down's syndrome, cytomegalovirus infection or meningitis. The results may not be generalisable to other areas of the UK.

CI – confidence intervals; IQR – inter-quartile range

Sub-question: What is the impact of a potential false-negative on children and their families?

Table 15. Fortnum et al (2016)[4] (systematic review on impact of screening)

Publication	Fortnum H. Ukoumunne OC. Hyde C. Taylor RS. Ozolins M. Errington S. Zhelev Z. Pritchard C. Benton C. Moody J. Cocking L. Watson J. Roberts S. A programme of studies including assessment of diagnostic accuracy of school hearing screening tests and a cost-effectiveness model of school entry hearing screening programmes. Health Technology Assessment 2016, 20(36)
Study details	Systematic review within a health technology assessment[12]
Study objectives	To assess the impact of a potential false-negative screening result
Inclusions	Studies published from database inception to May 2014 (language restrictions, if any, not reported)
Exclusions	None stated
Population	Children receiving screening hearing
Intervention	N/a
Comparator	N/a
Outcomes	No studies were identified reporting false-negative data for school entry hearing screening The review discussed several studies that reported numbers of children who passed newborn screening or testing as an infant but were later referred for audiological assessment. A similar study of college students was also discussed. The results of these studies are outside the scope of this review and are not reproduced here
Quality appraisal	This review was assessed using the CASP checklist for systematic reviews. No studies of interest were identified so only the conduct of the search could be assessed. There was a lack of detail provided about any language restrictions but otherwise there were no concerns.

[12] The HTA also discussed false negative results from a case-control study conducted as part of the HTA. These results are not included in this review as case-control study design is not one of the inclusion criteria for this question and the impact of a false negative within a case-control population may differ from the impact within a screening programme. The HTA also reported the results of a questionnaire sent to the parents of children referred to audiology services following school entry hearing screening. The questionnaire was sent to all parents, regardless of whether the child was diagnosed with a hearing impairment or not. No separate results were presented for children who had a false negative result therefore the questionnaire outcomes are not included in this review.

Appendix 4 – UK NSC reporting checklist for evidence summaries

All items on the UK NSC Reporting Checklist for Evidence Summaries have been addressed in this report. A summary of the checklist, along with the page or pages where each item can be found in this report, is presented in 16.

Table 16. UK NSC reporting checklist for evidence summaries

	Section	Item	Page no.
1.	TITLE AND SUMMARIES		
1.1	Title sheet	Identify the review as a UK NSC evidence summary.	Title page
1.2	Plain English summary	Plain English description of the executive summary.	5
1.3	Executive summary	Structured overview of the whole report. To include: the purpose/aim of the review; background; previous recommendations; findings and gaps in the evidence; recommendations on the screening that can or cannot be made on the basis of the review.	6
2.	INTRODUCTION AND APPROACH		
2.1	Background and objectives	Background – Current policy context and rationale for the current review – for example, reference to details of previous reviews, basis for current recommendation, recommendations made, gaps identified, drivers for new reviews	9
		Objectives – What are the questions the current evidence summary intends to answer? – statement of the key questions for the current evidence summary, criteria they address, and number of studies included per question, description of the overall results of the literature search.	11
		Method – briefly outline the rapid review methods used.	13

2.2	Eligibility for inclusion in the review	State all criteria for inclusion and exclusion of studies to the review clearly (PICO, dates, language, study type, publication type, publication status etc.) To be decided *a priori*.	14
2.3	Appraisal for quality/risk of bias tool	Details of tool/checklist used to assess quality, e.g. QUADAS 2, CASP, SIGN, AMSTAR.	16
3.	SEARCH STRATEGY AND STUDY SELECTION (FOR EACH KEY QUESTION)		
3.1	Databases/ sources searched	Give details of all databases searched (including platform/interface and coverage dates) and date of final search.	16
3.2	Search strategy and results	Present the full search strategy for at least one database (usually a version of Medline), including limits and search filters if used. Provide details of the total number of (results from each database searched), number of duplicates removed, and the final number of unique records to consider for inclusion.	Appendix 1
3.3	Study selection	State the process for selecting studies – inclusion and exclusion criteria, number of studies screened by title/abstract and full text, number of reviewers, any cross checking carried out.	17, 33
4.	STUDY LEVEL REPORTING OF RESULTS (FOR EACH KEY QUESTION)		
4.1	Study level reporting, results and risk of bias assessment	For each study, produce a table that includes the full citation and a summary of the data relevant to the question (for example, study size, PICO, follow-up period, outcomes reported, statistical analyses etc.). Provide a simple summary of key measures, effect estimates and confidence intervals for each study where available. For each study, present the results of any assessment of quality/risk of bias.	Appendix 3
4.2	Additional analyses	Describe additional analyses (for example, sensitivity, specificity, PPV, etc.) carried out by the reviewer.	22
5.	QUESTION LEVEL SYNTHESIS		

5.1	Description of the evidence	For each question, give numbers of studies screened, assessed for eligibility, and included in the review, with summary reasons for exclusion.	18, 21, 27
5.2	Combining and presenting the findings	Provide a balanced discussion of the body of evidence which avoids over reliance on one study or set of studies. Consideration of four components should inform the reviewer's judgement on whether the criterion is 'met', 'not met' or 'uncertain': quantity; quality; applicability and consistency.	18, 22, 28
5.3	Summary of findings	Provide a description of the evidence reviewed and included for each question, with reference to their eligibility for inclusion. Summarise the main findings including the quality/risk of bias issues for each question. Have the criteria addressed been 'met', 'not met' or 'uncertain'?	20, 26, 31
6.	REVIEW SUMMARY		
6.1	Conclusions and implications for policy	Do findings indicate whether screening should be recommended? Is further work warranted? Are there gaps in the evidence highlighted by the review?	32
6.2	Limitations	Discuss limitations of the available evidence and of the review methodology if relevant.	32

References

[1] UK National Screening Committee. Briefing note: screening for permanent hearing loss in children at school entry, April 2018
[2] World Health Organization. Childhood hearing loss: strategies for prevention and care. 2016. Available from http://apps.who.int/iris/bitstream/handle/10665/204632/9789241510325_eng.pdf;jsessionid=FF98152CA498111F49A789882CC95EB0?sequence=1 (Accessed August 2018)
[3] Wood SA. Sutton GJ. Davis AC. Performance and characteristics of the Newborn Hearing Screening Programme in England: The first seven years. International Journal of Audiology 2015, 54: 353-358
[4] Fortnum H. Ukoumunne OC. Hyde C. Taylor RS. Ozolins M. Errington S. Zhelev Z. Pritchard C. Benton C. Moody J. Cocking L. Watson J. Roberts S. A programme of studies including assessment of diagnostic accuracy of school hearing screening tests and a cost-effectiveness model of school entry hearing screening programmes. Health Technology Assessment 2016, 20(36)
[5] Bruderer A. Wood S. Data report to NHSP Advisory Group. Public Health England, October 2017
[6] Bamford J. Fortnum H. Bristow K. Smith J. Vamvakas G. Davies L. Taylor R. Watkin P. Fonseca S. Davis A. Hind S. Current practice, accuracy, effectiveness and cost-effectiveness of the school entry hearing screen. Health Technology Assessment 2007, 11(32)
[7] Watkin PM. Baldwin M. Identifying deafness in early childhood: requirements after the newborn hearing screen. Arch. Dis. Child 2011, 96: 62-66