Models for screening and surveillance of hearing in early childhood: Identification and review of evidence and efficiency

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An Evidence Check review brokered by the Sax Institute for NSW Kids and Families

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CONTENTS

LIST OF ABBREVIATIONS

AAA	American Academy of Audiology
AABR	Automated Auditory Brainstem Responses
CFHT	Child and Family Health Team
CMV	Cytomegalovirus
DPOAE	Distortion Product Otoacoustic Emissions
HKC	Healthy Kids Check
JCIH	Joint Committee on Infant Hearing
NHMRC	National Health and Medical Research Council
NICU	Neonatal Intensive Care Unit
OAE	Otoacoustic Emissions
PICO	Population Intervention Comparison Outcome
RCT	Randomised Controlled Trial
REA	Rapid evidence assessment
SCOUT	State-wide Comparisons of Outcomes Study
SWIS-H	State-wide Infant Screening – Hearing Program
TEOAE	Transient Evoked Otoacoustic Emissions
UNHS	Universal Newborn Hearing Screening
VRA	Visual Reinforcement Audiometry

MAIN MESSAGES

This review examined programs and service models that deliver population-based screening and surveillance of hearing for children aged 0-5 years. A rapid review methodology of literature published since 2005 was employed to investigate the relative effectiveness and efficiency of hearing screening in identifying postnatal hearing loss before school-age.

1. Approaches that are promising

   - Good quality research supports the value of targeted surveillance of infants who pass their newborn hearing screen and have a strong risk factor for postnatal hearing loss. Systematic follow-up systems should be instigated for children with congenital infection (especially CMV), craniofacial abnormalities, Down syndrome, and syndromes known to be associated with hearing loss.

     This is the only approach to post-neonatal surveillance for detection of hearing loss in young children that has been at least partly evaluated on a population level. Challenges primarily involve ensuring adequate population coverage. This requires systematically ascertaining the risk factors, then setting in place the mechanisms to track the children, provide the service at agreed ages, record, and minimise loss to follow-up, and be able to report on the success of the program.

     Infrastructural requirements would include (a) systematic support for professionals and parents to assist with accurate ascertainment of risk factor status and attendance at diagnostic appointments, and (b) adequate data systems.

   - Based on promising evidence, future directions are likely to include increased molecular and genetic testing alongside newborn hearing screening.

2. Approaches that are not supported

   For infants and young children up to school entry, recent evidence does not support:

   - Screening for transient middle ear difficulties (such as otitis media, glue ear)
   - Universal screening using automated technology in preschool, day care or health care settings
   - Use of questionnaires to detect hearing difficulties.

EXECUTIVE SUMMARY

Universal newborn hearing screening (UNHS), which has now been implemented in most of the developed world, has demonstrated effectiveness as a tool to detect the vast majority of congenital permanent moderate or greater hearing losses within the first few months of life. Furthermore, UNHS can lead to the detection of mild and unilateral hearing loss, which are not the target conditions of UNHS screening programs. However, some permanent hearing losses in childhood cannot be detected via UNHS. These postnatal hearing losses include acquired or progressive losses, as well as losses that were present but missed at screening (true false negatives, believed to be very uncommon). Children with such losses can be affected with significant speech and language delays and poorer education achievement if they do not receive the appropriate intervention.

The vast majority of postnatal hearing loss in the early childhood years is mild and temporary, due to otitis media with effusion dulling sound as it passes through the middle ear towards the cochlea. This condition affects almost all young children at some point (so that any screening program would need to be undertaken repeatedly and frequently) and around 5% of the population at any given time. Even though of concern to many parents and professionals, it does not meet population criteria for screening because it is transient, the hearing loss is usually mild, it has developmental outcomes similar to those without the condition, and treatment confers only temporary benefit but has the potential for lasting harm.

Therefore, systems for identification of permanent postnatal hearing loss were the focus of this review.

A review of the literature published since 2005 indicated that there were no papers with high levels of evidence (such as systematic reviews, meta-analyses, quality guidelines, or randomised controlled trials) reporting on systems or tools for detection of postnatal hearing loss. Consequently, a number of papers with lower levels of evidence (total 18) that met the inclusion criteria for this review were examined, using a rapid review methodology.

Each of these 18 papers was evaluated independently by two assessors against a number of criteria, namely (i) strength of the evidence presented (including assessment of the quality of the paper, risk of bias, quantity of evidence including statistical power and number of supporting papers, and NHMRC evidence ranking); (ii) overall strength; (iii) consistency (a judgement on the replicability of the results); (iv) cost effectiveness (efficiency); (v) generalisability (whether results could be generalised to the NSW population); and (vi) applicability (relevance to the NSW context). An overall ranking of the evidence presented by each paper was also determined.

A number of approaches to detection of postnatal hearing loss were identified through this review. They were:

- Targeted surveillance of infants with risk factors for postnatal hearing loss
- Screening using a variety of techniques including:
    - pure-tone audiometry
    - otoacoustic emissions technology

- questionnaires
- tympanometry
- behavioural audiological assessment
- imitanciometry
- visual inspection of the ear, and
- molecular testing.

The papers reporting on these approaches were heterogeneous in relation to the selection of participants, methodology, and outcomes of interest, precluding comparison across studies. Consequently, the papers were grouped based on either the approach taken to screening, or the setting in which the screening tool was implemented.

The results of the review indicated that targeted surveillance for children with a risk factor for postnatal hearing loss was supported by the available literature, and was classified overall as a “Promising” approach. This should be limited to a small number of risk factors, almost all of which are clearly evident soon after birth, i.e. congenital infection (especially CMV), craniofacial abnormalities, Down syndrome, and syndromes known to be associated with hearing loss. The studies reporting on this approach were of good quality and low bias, and were conducted in settings that would be largely generalisable to the NSW context. Challenges with this approach include the necessity to use tightly-defined criteria for the risk factors deemed to be appropriate for surveillance, and ensuring adequate support and data systems to maximise follow-up after identification of a risk factor.

Surveillance utilising a questionnaire approach was reported in one study for both infants and school entrants. Challenges relate to insufficient population coverage (despite efforts to improve this), poor practices around using the questionnaires tools, and minimal information as to accuracy (screening sensitivity and specificity) which is likely to be low. This approach was ranked as “Not supported”.

Hearing screening in primary care settings is viewed as opportunistic in some jurisdictions as it can be done in an existing setting, with an existing workforce, and can be incorporated into routine primary health care. The published literature in this field identified a number of challenges with this approach to hearing screening, including difficulties with compliance amongst young children to the testing requirements, lack of follow-up of children who required repeat screening or diagnostic assessment. Furthermore, these approaches tended to detect large numbers of children with transient mild conductive hearing loss rather than permanent hearing loss. The papers reporting on this approach had a low level of evidence. Although of low bias and fair to good quality, they did not provide sufficient data with which to make judgements about sensitivity and specificity of the screening techniques. Use of videoconferencing as a method of delivery of hearing screening in primary care was also examined. Although some of the findings reported were generalisable and applicable to the NSW context, given the existing evidence base hearing screening of young children in the primary care setting was ranked as “Not supported”.

Preschool and day-care settings provide an alternate environment in which to integrate hearing screening services for young children. Otoacoustic emissions, pure tone audiometry, tympanometry, imitanciometry and questionnaires have all been reported on as methods of screening in these settings. Amongst underserved populations, two approaches were classified as “Promising”; TEOAE screening in preschool/day-care, and a protocol involving

inspection of the ear and repeating OAE screening over a 2-4 week period. However when taking into account generalisability and applicability to the broader NSW population, and considerable operational challenges including difficulties with screen compliance and high loss to follow-up rates, screening in the general preschool setting was ranked as “Not supported” based on available literature.

Advances in molecular testing, particularly in screening simultaneously for multiple genetic mutations known to cause deafness and presence of CMV DNA within a single panel, has resulted in some research examining the potential role of genetic screening in identifying those at risk of permanent hearing loss that is not detectable by UNHS. Major trials are currently under way internationally examining this approach. This approach is likely to generalise and be applicable in NSW. Thus, while the current evidence remains insufficient to recommend its adoption, molecular testing via genetic screening of newborns was rated as a “Promising” adjunct to UNHS.

In conclusion, this Rapid Review supports several recommendations, as below.

1. Implement systems for targeted surveillance of young children who pass their newborn hearing screen but have one of the four following risk factors for postnatal hearing loss:

    - Down’s syndrome
    - Other syndromes known to be associated with a hearing loss
    - Craniofacial anomaly, and
    - Congenital infection.

    Successful implementation of such surveillance will depend on a number of factors, including sufficient population coverage, clear definitions of risk factors, systems to maximise diagnostic appointment uptake, and systems to track and follow children through early childhood.

2. Watch for forthcoming evidence regarding:

    - Costs and benefits of metabolic and CMV screening
    - School entry hearing screening (trial undertaken in England).

3. Audiologic services should continue to be widely and readily available for diagnostic testing of children with possible hearing loss referred via usual case-finding approaches, including:

    - Parent/professional concerns about hearing loss or development (especially in language and related abilities)
    - Illnesses or events during childhood known to cause hearing loss, e.g. meningitis, head injuries.

BACKGROUND

NSW Kids and Families is responsible for strategic, long-term planning for the health services for children and young people across NSW. The Child and Family Health Team (CFHT) delivers *NSW Kids and Families'* early childhood health and development program, which includes screening/surveillance as a core approach to maximising health outcomes for this population.

The NSW State-wide Infant Screening – Hearing (SWIS-H) Program aims to identify all babies born in NSW with significant permanent bilateral hearing loss by 3 months of age. A major objective of the program is to ensure children with permanent hearing loss are able to access appropriate intervention by 6 months of age. Currently, NSW does not have a formalised system to detect hearing loss in children beyond the newborn period.

This rapid review, brokered by the Sax Institute and conducted by the Centre for Community Child Health at the Murdoch Childrens Research Institute, focuses on children aged 0-5 years, in particular to determine the effectiveness and efficiency of hearing screening and surveillance programs and service models beyond newborn hearing screening.

INTRODUCTION

A string of advances over recent decades has profoundly altered the landscape of childhood hearing impairment.

The prevalence of sensorineural hearing loss has continued to fall as infective causes of acquired hearing loss, such as rubella and bacterial meningitis, become ever rarer due to immunisation and antibiotics. These gains are partly offset by increasing survival of very premature infants, although even here steadily improving care continues to reap benefits.

With national implementation of universal newborn hearing screening (UNHS), the mean age at which congenital bilateral and unilateral hearing losses are detected has plummeted since 2000 from over 2 years to usually within the first 3 months of life. Most states now achieve the benchmark coverage of at least 96% of infants screened. Evidence indicates that this detects at least 90% of moderate or greater congenital hearing losses, along with many cases of milder bilateral and unilateral loss (1, 2). With an underlying prevalence of around 1.1/1000 live births, 90% detection means that only around 1 in 10,000 babies may have undetected moderate or greater congenital hearing loss.

Furthermore, technological advances with cochlear implantation, advanced hearing aids and systems such as FM loops have revolutionised affected children's access to useable sound.

Collectively, therefore, the life chances of children born today with moderate or greater hearing losses are vastly better than those of their counterparts born just 20 years ago. This was confirmed by the State-wide Comparisons of Outcomes (SCOUT) study. This compared population outcomes in the early school years for children born in New South Wales in 2003-5 following the advent of SWIS-H with those of children born in the same period in Victoria, when neonatal intensive care (NICU) and risk factor screening were offered. Mean language scores rose from below (Victoria) to within (NSW) the normal range with the introduction of newborn hearing screening (3).

At the other end of the severity scale, anecdotally there is growing concern about the possible impacts of milder losses. With diagnostic techniques and amplification technologies continually improving and Australian Hearing's outstanding service provision arrangements, diagnosis and treatment of hearing losses are now within reach for all children. Thus, Australian Hearing's fastest-growing treatment group in the last five years has been children with mild hearing losses. These large numbers of hearing aids raise questions as to what additional benefits versus costs might accrue from possibly bringing fitting forward by screening. No one would argue that having a hearing loss of any degree is beneficial.

This concern is offset by a number of considerations. Firstly, the vast majority of mild losses are due to otitis media with effusion (glue ear). Almost all children have at least one episode of otitis media with effusion, and many have repeated episodes. Although transient, around half of the affected children have a mild hearing loss while the effusion is present. However, large population-based studies (such as the Pittsburgh and Nijmegen studies) demonstrated the benign nature of such losses with no long-term developmental, behavioural and academic outcomes demonstrated (4, 5). Further, randomised trials showed that tympanostomy tubes not only have no lasting developmental benefits for otherwise-normal children, but may even be harmful with much higher rates of subsequent lasting tympanic membrane abnormalities (6).

Whereas transient mild conductive losses due to otitis media with effusion are common, permanent sensorineural mild losses are not. The Melbourne-based Hearing in Schools Study showed slight and mild sensorineural loss to affect less than 1% of primary school children (7). While such losses did reduce phonological discrimination, this did not translate into obvious adverse downstream effects on standardised assessments of language, academic achievement, social-emotional wellbeing, or quality of life (7). A large study found that adults rarely accept hearing aids until hearing losses exceed around 35 dB HL in the better ear – close to the cut point for moderate hearing loss – suggesting that the costs and hassles outweigh the subjective benefits of aiding at milder levels, at least for adults (8).

These findings jointly suggest that screening for milder hearing loss may not be a fruitful or cost-effective activity.

The Problem

It is timely to reconsider the place for post-neonatal early-years screening for hearing loss for a variety of reasons:

1. The prevalence of known permanent hearing loss approximately doubles between birth and school entry, mostly towards the mild end of the spectrum. These additional cases most likely represent:

    - losses that were always present but not detected by UNHS
    - hearing losses that developed *de novo* during the preschool years
    - progressive losses, i.e. already present but too mild to detect at birth.

    For any of these, there may have been a prolonged period during which the hearing loss was present but not identified. This may not be a palatable situation.

2. Improved technologies based on otoacoustic emissions (OAEs) and hand-held automated auditory brainstem responses (AABR) make it feasible to rapidly test hearing in large numbers of children in primary care and educational settings, but the costs and benefits of doing so remain uncertain.

3. Screening for known ‘deafness genes’ and pre/perinatal cytomegalovirus infection is becoming a viable complement or alternative to direct screening of hearing itself. It can be undertaken on samples routinely collected at birth, obviating the challenges of testing the child.

In summary, as mechanisms for detection and treatment become more accessible, the balance may be swayed towards offering additional screening during the infant and preschool years. On the other hand, when UNHS and follow-up are fully operational, most postnatal hearing loss detected by screening is likely to be mild; almost all will be transient; and there is little evidence that at the population level (the sphere within which screening is typically offered) mild hearing loss is harmful in terms of broad language, academic and quality of life outcomes. Thus, it is timely to carefully consider recent evidence examining the costs, benefits and potential harms before making any changes to current hearing screening practices in young children.

The Aim

This rapid review addresses 3 key questions:

1. What programs and service models deliver population-based screening and surveillance of hearing for children aged 0-5 years (i.e. beyond the neonatal period)?

2. What is the relative effectiveness and efficiency of these programs?

3. Is there evidence to support a hearing program that provide screening and surveillance beyond the neonatal period, and are there any ‘key considerations’ for the NSW setting in particular?

METHOD

This literature review utilised a rapid evidence assessment (REA) methodology. The REA is a research methodology that uses similar methods and principles to a systematic review but makes concessions to the breadth and depth of the process, in order to be completed within a short timeframe. Rigorous methods for locating, appraising and synthesising the evidence related to a specific topic are utilised by the REA; however, the methodology places a number of limitations in the search criteria and in how the evidence is assessed. For example, REAs often limit the selection of studies to a specific time frame (e.g., last 10 years), and limit selection of studies to published peer-reviewed, English-language studies (therefore excluding unpublished pilot studies, difficult-to-obtain material and/or non-English language studies). The REA can help inform policy and decision makers more efficiently by synthesising and ranking the evidence in a relatively short space of time, although it is not necessarily as exhaustive as a well-constructed systematic review or meta-analysis.

Defining the Research Question

The components of the question for this review were defined in terms of the population, the screening and surveillance models, and the outcomes (refer to Appendix 1). Operational definitions were established for key concepts, and specific inclusion and exclusion criteria were defined for studies for this review.

The population was defined as infants and children between the age of 0 and 5 years but excluding newborn infants and school-age children. Screening was defined in line with the definition specified in the NHMRC review 2002:

> **Screening test:** "Any measurement aimed at identifying individuals who could potentially benefit from intervention. This includes symptoms, signs, lab tests, or risk scores for the detection of existing or future disease."(9)

> **Screening program:** "In a screening program, a test, or a series of tests, is performed on a population that has neither the signs nor symptoms of the disease being sought but whose members have some characteristic that identifies them as being at risk from that disease, the outcome of which can be improved by early detection and treatment. Screening actually consists of all the steps in a program from the identification of the population at risk to the diagnosis of the disease or its precursor in certain individuals to the treatment of those individuals."(10)

> **Population surveillance:** Population surveillance focuses on groups or entire populations, and enables observation of changes and trends at a public health level. This is sometimes referred to as monitoring (11).

The outcomes of screening and surveillance models were defined as follows:

> **Effectiveness:** the extent to which the model or program improved the desired outcomes when applied to the population.

> **Efficiency:** the cost-effectiveness of the model or program, inclusive of any harms, benefits and costs of the program to individuals and society.

Search Strategy

The following databases were used to identify relevant literature related to this topic: Ovid MEDLINE, CINAHL (EBSCO), PsychINFO, Cochrane library, and PubMed. An example of the search strategy conducted in the Ovid Medline database can be found in Appendix 2.

The quality of studies was assessed using the levels of evidence described by the National Health and Medical Research Council (NHMRC) (Appendix 3). Given the low yield of relevant papers, we did not exclude any on the basis of study design.

Search Terms

The search terms specific to this question that were included in searching the Title/s, Abstract/s, MeSH terms, and Keywords lists were:

- *hearing disorders, persons with hearing impairments, hearing loss, otitis media, otitis media with effusion, glue ear, conductive hearing loss, hearing tests, tympanometry, pure tone screen, acoustic reflex, acoustic reflectometry, otoacoustic emissions screen, pneumatic otoscopy*
- *hearing screening, mass screening, population surveillance, public health surveillance*

Paper Selection

Studies were evaluated according to the following inclusion and exclusion criteria:

Included:
1. Nationally or locally published peer-viewed research studies 2. Human infants and children between 0-5 years 3. English language
Excluded:
1. Non-English 2. Published prior to 2005 3. Study group - age >5 years, or neonatal screening 4. Validation study, animal study, review paper, technical report, stand-alone methods paper 5. Developing country 6. Not population-based screening or surveillance, or outcome data did not report on screening model 7. Screening test for specific disease or disorder 8. Papers where a full text version is not readily available

Information Management

Papers identified via filtering and key word searches were imported into EPPI-Reviewer 4 software. Further refinement was required to ensure that only high quality and relevant publications were included for data extraction. A screening process was adopted to code for eligibility using content from the title and abstract; the screening form is presented in Appendix 4. All records were screened according the eligibility criteria and decisions to “include” or “exclude” were double-checked by a second reviewer for quality control purposes. Full text

versions of all studies identified as meeting eligibility requirements were obtained and uploaded to the software. The full text reports were then screened for inclusion by two independent reviewers. In the case of discrepancies, discussions were held and a consensus reached. Papers meeting the inclusion criteria were subject to data extraction.

The following information, where possible, was extracted for studies that met the inclusion criteria:

- Sample characteristics
- Objective of the screening model or program
- Hearing related issue/condition tested
- Hearing assessment method used
- Instrumental requirements
- Operational and administration parameters
- Workforce and capacity issues
- Evaluation data.

Evaluation of the Evidence

Five key components contributed to the overall evaluation of the evidence; these are listed and defined below, including the overall rankings applicable for each component.

Strength of the Evidence

Quality and risk of bias

This refers to how well the studies were conducted; how the participants were selected, allocated to groups, managed and followed-up; and how the study outcomes were defined, measured, analysed and reported.

An assessment of the quality and bias of each individual study was conducted by two independent reviewers using, where possible, standard means to evaluate quality and bias. Appendix 5 gives an example of the criteria used to evaluate non-RCT observational studies - Systems to Rate the Strength of Scientific Evidence. Appendix 6 gives an example of the criteria used for ranking screening tools and screening programs. An overall grading was applied to each individual study of “Good”, “Fair”, or “Poor”. In instances where there was discrepancy between reviewers, discussions were held and a consensus was reached.

Quantity of evidence

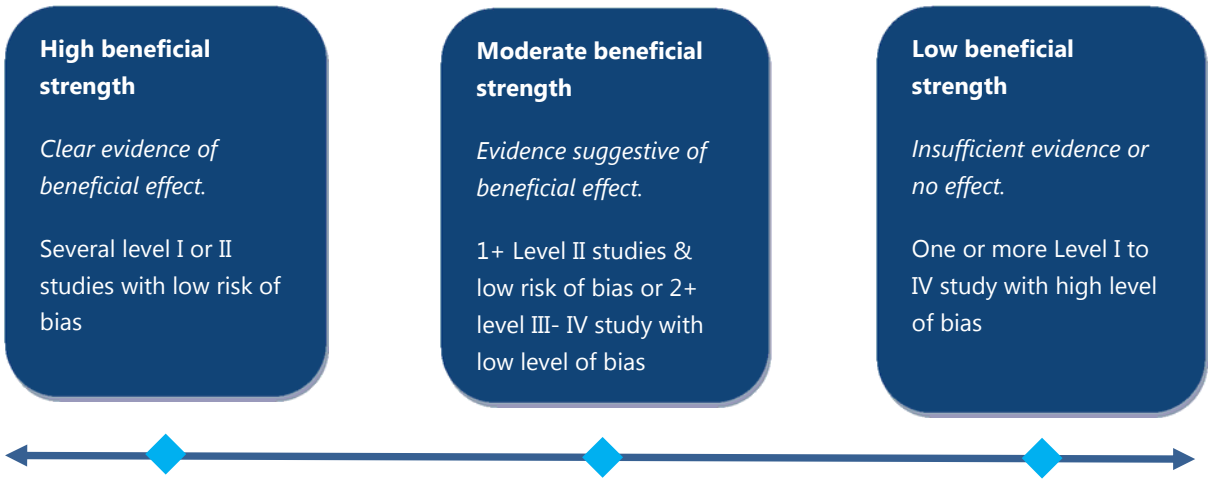
This refers to the number of studies included as the evidence base for each ranking. The quantity assessment also took into consideration the statistical power of the studies – i.e., whether there were enough participants to draw firm conclusions. Small underpowered studies that are otherwise sound may be included if their findings were generally similar but, in order to be considered high quantity of evidence, at least some of the studies cited as evidence must have been large enough to detect the size and direction of the effect.

Level of evidence

This reflects the best study types for the research question, assessed using the NHMRC hierarchy of evidence (Appendix 3). The most appropriate study design to answer this review's questions is level II evidence. Studies at levels III and IV are progressively less robust.

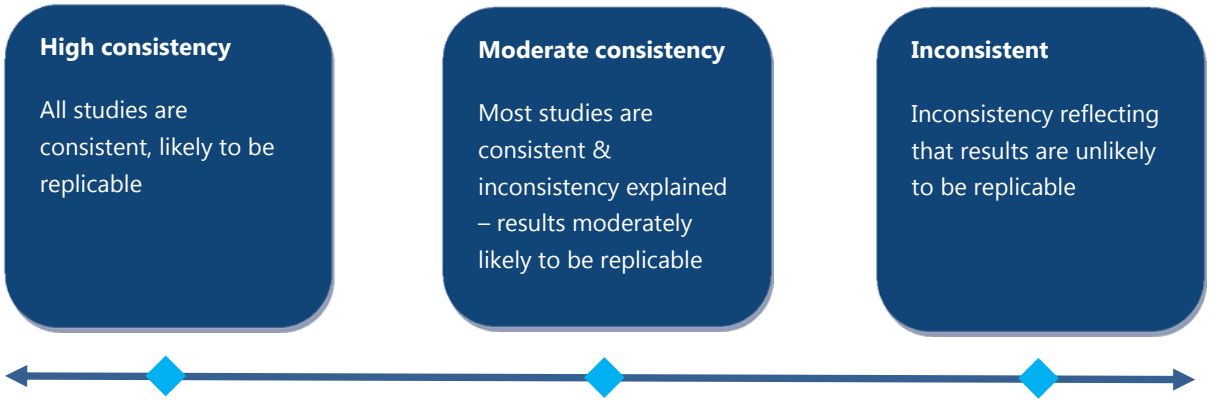
Overall strength

In consideration of the accumulated evidence for related papers a judgement was reached about the strength of the evidence base, taking into account the quality and risk of bias, quantity of evidence and level of evidence. This was determined by two independent raters and consensus reached in the event of any rating discrepancy.



Consistency

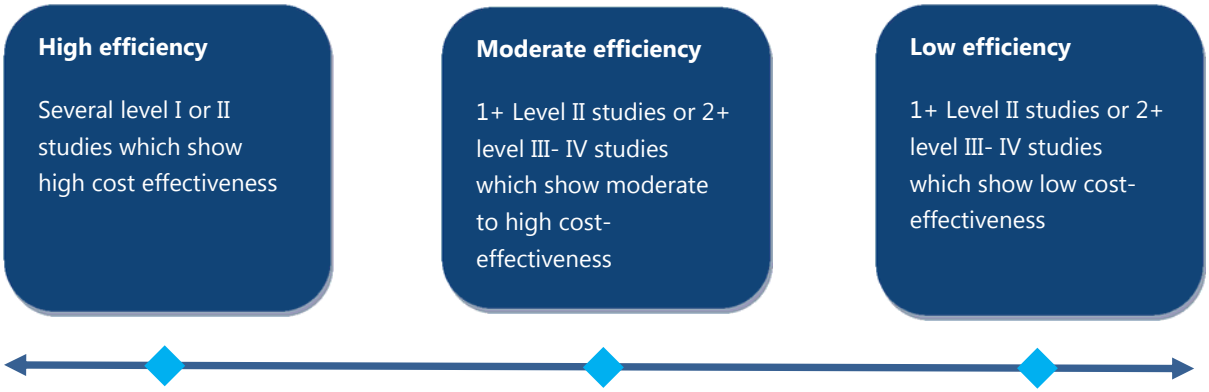
A judgement was made as to whether the findings were consistent across the included studies (including across a range of study populations and study designs).



Cost Effectiveness (Efficiency)

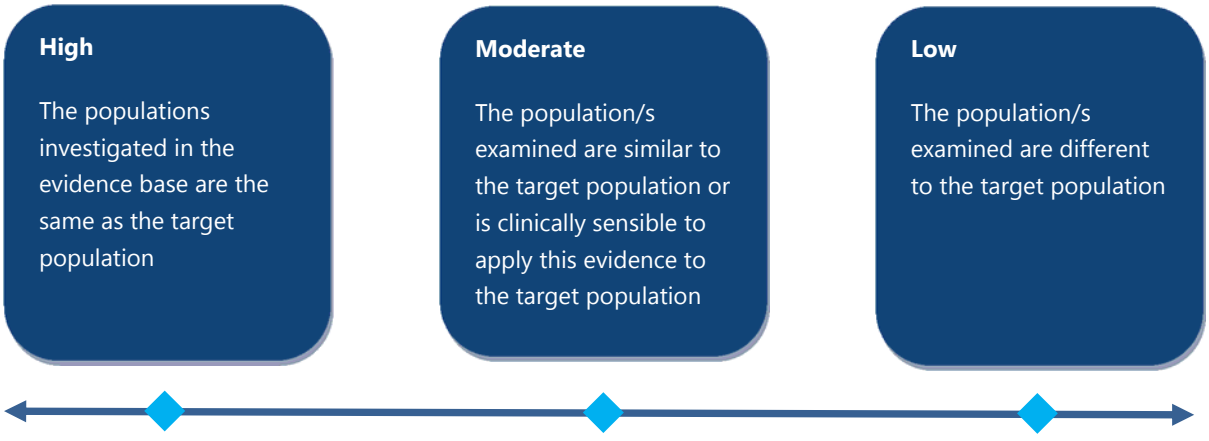
Efficiency is a measure of the economy with which an intervention of known efficacy and effectiveness is carried out. This is frequently used synonymously with cost-effectiveness, which in most cases is appropriate. As well as the actual costs of a screening program, a measure of efficiency or cost-effectiveness must consider the costs of any potential harms of

the intervention versus the benefits, and the opportunity costs of other interventions that are foregone in favour of the program in question.



Generalisability

Generalisability refers to how well participants and settings can be generalised to the NSW population. Population issues that might influence the relative importance of recommendations include gender, age or ethnicity, baseline risk, or the level of care (e.g. community or hospital). This is particularly important for evidence from randomised controlled trials (RCTs), as the setting and entry requirements for such trials are generally narrowly based and therefore may not be representative of all the patients to whom the recommendation may be applied in practice.



Applicability

A judgement on the applicability is determined by whether the evidence base is relevant to the NSW context, or to specific local settings (rural, cities). Factors that may reduce the direct application of study findings to the Australian or more local settings include organisational factors (e.g. availability of trained staff, clinic time, specialised equipment, tests or other resources) and cultural factors (e.g. attitudes to health issues, including those that may affect compliance with the recommendation).

Ranking the Evidence

An overall ranking of the evidence is determined by considering the criteria above – strength of evidence, consistency, cost effectiveness (efficiency), generalisability, and applicability. The total body of evidence was ranked into 4 categories “Supported”; “Promising”; “Unknown”; and “Not supported”. Agreement was sought between two independent raters.

Supported	Promising	Unknown	Not supported
Clear, consistent evidence of benefit	Evidence suggestive of benefit but more evidence needed	Insufficient evidence. More research needed	Evidence of no effect or negative effect

RESULTS

The search strategy identified 1,151 unique references, which were screened for eligibility for data extraction. The PRISMA flow chart in Appendix 7 illustrates the screening process and reference numbers. There were no systematic reviews, meta-analyses, quality guidelines, or randomised controlled trials. Given this low yield, all other studies that met the inclusion criteria were included for data extraction regardless of the evidence level. In total 18 papers were included, with 11 studies originating from the USA, two each from Australia, the UK, and China, and one from Brazil.

Summary

The majority of the studies identified by the search strategy were fairly evenly spread between medical settings (private/public hospitals, health clinics/health centres) and preschool/day-care centres. Although the focus was on children aged between 0 and 5 years of age, several studies included the target population as well as older children up to the age of 6 years (in five studies) and 19 years (one study). Most programs were designed to flag for risk of postnatal hearing loss rather than test specifically for a single hearing issue. To this end, most studies/programs also used multiple testing instruments. Tympanometry was used in 7 studies; pure tone screening was used in 4 studies; transient evoked otoacoustic emissions (TEOAEs), behavioural assessment (visual reinforcement audiometry (VRA) or play audiometry) and various versions of a risk factor questionnaire were utilised in 3 studies; otoacoustic emissions (OAE) screening (unspecified type) was reported in 2 studies; and automated auditory brain stem response (AABR), distortion product otoacoustic emissions (DPOAE), visual inspection, imitanciometry, and molecular testing were each reported once. Most of the studies used more than a singular screening measure.

The inclusive nature of this review meant that the outcomes of interest were highly variable and thus not easily comparable. That said, it was important to evaluate the evidence-base within this domain across varying objectives to adequately address the research questions and provide pertinent recommendations. A summary of the evidence can be found in Appendix 8 and details pertaining to the evaluation of individual studies can be found in Appendix 9.

Targeted Hearing Surveillance

The utility of targeted surveillance programs using a risk factor registry was explored by 3 studies, 2 of which are linked. The linked studies reported on the same program and data-set, which included all children born in Queensland, Australia, between September 2004 and December 2009. These children all received a bilateral pass on newborn hearing screening but had at least 1 risk factor as determined by a modified version of the registry used by the Joint Committee on Infant Hearing (JCIH) (12), and were referred for targeted surveillance.

The first of these studies identified a number of limitations with the targeted surveillance program (13). The lost contact rate was high (32.4%), there were delays in first surveillance assessment with only 46% seen within protocol timeframes, and extensive diagnostic assessments were completed on children with normal hearing (overall yield 0.76%).

The objective of the second study was to investigate which risk factors were most likely to predict postnatal hearing loss (14). Family history and craniofacial anomalies were the only risks identified as being predictive of the occurrence of hearing loss in children. Statistical limitations prevented any conclusive evidence about syndrome and prolonged ventilation, but preliminary results are in favour for monitoring these two risk factors. In contrast, low birth weight was not predictive of hearing loss. There was insufficient evidence to support monitoring of children who had suffered severe asphyxia, congenital infection, bacterial meningitis, professional concern, and/or hyperbilirubinemia.

The third study, conducted in the United Kingdom, examined the effectiveness of targeted surveillance for children who pass the newborn hearing screen and have risk factors for later hearing loss (15). It included children born between April 2006 and September 2009 (n=2,307,880). The overall prevalence of permanent hearing loss amongst children in the study sample who had a risk factor for hearing loss and passed the newborn hearing screening was very low at 1.49/1000. The results indicated that Down's syndrome, other syndromes known to be associated with a hearing loss, craniofacial anomaly, and congenital infection should be retained in the targeted surveillance program. Furthermore, based on the protocols for newborn hearing screening in the UK, NICU admission with a refer result in both ears at newborn otoacoustic emissions screening but a pass in both ears at newborn automated auditory brainstem response screening was also recommended for retention as a risk factor. Note that this does not apply in New South Wales, where only AABR is offered as a neonatal screen. In contrast, targeting the remaining risk factors - family history, aminoglycoside administration, bacterial meningitis, prolonged ventilation, jaundice at exchange transfusion level, NICU >48 hours, and neurodegenerative or neurodevelopmental disorders - were shown to be ineffective as a method of targeted surveillance and have been discontinued. Similar to the results reported in the Australian-based targeted surveillance program, uptake by those offered an appointment was low (55.3%).

The outcomes drawn from these studies were based on three good-quality papers with low level of bias; therefore, evidence was judged to be high. There was a reasonable amount of consistency between the studies, despite some variability about which risk factors were effective at predicting postnatal hearing loss. Given that the overall conclusions from these studies differed somewhat, we perhaps place greater weight on the risk factors shown to be predictive in the UK study (Down's syndrome, craniofacial abnormalities and congenital infection) by virtue of its much larger numbers. Targeted surveillance in Australia met the characteristics required for a screening test and screening program, whereas based on the data provided the UK study met the criteria for a screening program only (full details of evaluation of the evidence are provided in appendix 9). The studies were conducted in Australia and the UK, making the data broadly generalisable and applicable to the NSW context.

As such, the use of targeted surveillance as a means of detecting hearing loss during the early years is "Promising", notwithstanding the significant challenges with loss to follow-up reported in both studies. If these more tightly-defined criteria were adopted, it is likely that far fewer children than currently would be targeted but that both follow-up and yield would improve.

Questionnaire Surveillance

A UK study used an existing data collection system to audit the data and assess local service performance with the view to improving the quality of health care by improving practice (16). The audit of data, which are entered into the child health system for children eligible for universal infant and school entry hearing screening, was extracted for coverage, referral, and yield. The data were from an infant cohort born 2003-2005 and a school-entry cohort born 1998-2000. The audit demonstrated that neither the infant nor the school entry hearing questionnaire surveillance reached the agreed standard of 80% coverage of the cohorts. There was no improvement in overall coverage over time, the referral rate was 1.2 to 2.6% in the infant cohort and 4.2 to 6.6% in the school entry cohort, and sensorineural hearing loss was confirmed in 0% of the infant cohort and 3 to 5% in the school entry cohort. The authors also identified a number of key areas of poor practice. For example, infant hearing surveillance questionnaires were administered much earlier than the recommended 7–9 months of age; school entry hearing tests were often conducted in unsuitable conditions, such as environments with high background noise levels, resulting in supra-threshold test results requiring retesting in similar conditions and thus increasing likelihood of unnecessary referral for further testing. The authors also discussed workforce challenges including the increased workload for school nurses, and recruitment and retention difficulties for health visitors, school nurses and audiologists. All contributed to delays in completion of the hearing screen and poor adherence to good practice of screening children at appropriate ages (16).

Despite being a good quality paper the level of evidence was fairly low, based on the NHMRC evidence hierarchy, as was the quantity of evidence - a single paper (17). The criteria for a screening test or program was “Not supported”, particularly for the school-entry test. The overall ranking of evidence for the use of questionnaires as tools for screening for hearing loss in infants and young children is “Not supported”.

Outcomes by Program Setting

The remainder of the surveillance programs/studies are presented in terms of the setting to provide a clearer point of reference for comparison.

Health Care Settings

The main objective of studies conducted within a primary care setting focused on the feasibility of integrating hearing screening protocols into services routinely provided by primary care providers. There was also some emphasis on and data available about hearing screening failure rates in primary care settings and referral practices following an abnormal screen.

Hearing screening failure rates

The screening programs and methods identified in the papers reporting on health care settings indicated high refer rates from screening, ranging from 3.8 to 10%. In a state such as NSW with an annual birth cohort approaching 100,000 births, this would equate to between 3,500 and 10,000 referrals for diagnostic audiology annually (if screening were offered at only one specific age between infancy and 5 years).

Incidence of postnatal hearing loss

The overall yield of possible or confirmed permanent sensorineural hearing loss from the screening programs ranged from 0.35% to 5.1% (16, 18, 19). In addition Bhatia et al. (18) reported 6% of participants had a transient conductive hearing loss.

Operational matters

Several operational issues were identified in studies examining hearing screening protocols within primary care settings. A study in the USA, which utilised a convenience sample of children undergoing hearing screening during a well-child visit to determine referral rates and practices, identified significant procedural concerns. First, age at testing influenced the number of children who completed the hearing screen protocol – pure tone screening was used most often (95%) with the rest (5%) screened using play audiometry. Almost half of the 3 year olds did not complete screening, compared with 7% of 4 year olds, 3% of the 5 year olds, and < 1% of children 6 years or older (20). Second, of those children who could not be assessed there was no further action taken in most cases (73%) and the rest were either scheduled for a recheck or referred for further evaluation (20). Significantly, of the children that did not pass the screen, less than half (41%) were either referred or rechecked (20).

Another US-based study examined the capacity of primary care providers to identify children in need of audiologic follow-up in a periodic infant-toddler hearing screening program implemented during well-child visits (18). In terms of operational success the authors report that 75% of screens finished in 1 to 10 minutes, staff found it useful when applied to their daily practice, and did not feel that it significantly affected clinic flow (18). The method implemented by this study involved the combined use of office-based otoacoustic emission screening with tympanometry as well as a risk factor questionnaire, which is in line with current American Academy of Audiology guidelines recommending multistep screening protocols to reduce referral rates. The referral rate based on this multistep protocol yielded a total of 2.6% of screened children with results suggesting a permanent sensorineural hearing loss and 6% with results suggesting a transient conductive hearing loss. Around one-fifth of patients were unable to successfully complete the OAE screen due to either noise disturbance, patient uncooperativeness, or equipment malfunction, despite the questionnaires having been completed. The tympanogram was able to be interpreted 86% of the time it was attempted and the audiologist agreed with the physician's interpretation in 89% of cases.

The two linked studies reporting on this issue were of good quality and low bias, but only met the criteria for low level of evidence (20, 21). That said, the issues raised by these two studies – operational challenges in screening young children in primary care settings and inconsistent or absence of follow-up by primary health care providers - were significant, and represent considerable challenges with regard to screening young children in this setting. The Bhatia et al. (18) study was both at a low level of evidence and only "fair" with regard to quality and bias rating, and it also raised significant issues of efficacy of hearing screening in this setting. In each regard an additional drawback was that the screening protocol did not meet the criteria for an acceptable screening test, primarily due to data not adequately reported to make this judgment, such as results of diagnostic testing. Therefore, even though this approach could be generalisable and applicable in NSW, the use of a hearing screening protocol in the primary care setting was ranked "Not supported".

One study examined the feasibility of using videoconferencing via Skype as a means of service delivery for speech, language, and hearing screening (22). The study was conducted in urban community health clinics, which service a relatively high proportion of minority, low socioeconomic areas. Several hearing assessment methods were utilised by this service model: tympanometry, behavioural assessment (VRA or play audiometry), brief parental interview (not hearing specific), and distortion product otoacoustic emissions (DPOAE). Service was provided such that the clinic facilitator performed the required test and the audiologist provided supervision and real time interpretation via videoconferencing. Screening reliability was determined by comparing pass/refer rates between those conducted via videoconferencing compared with those conducted on-site at the clinics. Reported reliability for pure tone and DPOAE screening was 100% and only 84% for tympanometry screenings. Attempts were made to evaluate by means of a survey participant satisfaction with videoconferencing service delivery, but the response rate was low (39.9%). Families that did complete the survey strongly agreed that the sound and picture quality was good, generally felt very comfortable using the computer, had a high level of satisfaction with the use of videoconferencing as a method of service delivery, and preferred videoconferencing over having a separate appointment at another facility with an audiologist. Seventy-five percent of families who had a child not pass one or more components of the screening, scheduled and kept that appointment; although this this figure is far from ideal, it is significantly higher than the proportion identified via targeted surveillance.

The strength of evidence of videoconferencing in the primary care setting is fair-to-poor given the low level of evidence, poor quality and bias check, and the inadequacy in meeting the criteria required for an acceptable screening test. Information regarding cost-effectiveness would be necessary to sufficiently evaluate this model in comparison to others, as well as high-quality evaluation of the various screening approaches reported. Furthermore, there was not enough information about recruitment and sample characteristics to determine generalisability or applicability to the NSW context. Improving access to health care for the underserved is an important goal; however, given the limitations identified in this study, and the other studies within similar settings discussed above, the overall ranking of evidence for screening via videoconferencing is judged as “Not supported”.

Workforce & capacity

Staff undertaking the hearing screening protocol in primary care settings were described as one or more of the following: clinic facilitator, medical assistants, research assistants, clinic staff, and/or physicians. Several of the reviewed studies briefly described the process used to train staff. This typically involved at least a half day training session, which may have included printed materials, demonstrations, and hands-on practice (18-20). In one US-based study, practices were also provided with research assistants and guidelines were distributed to each practice; however, no mechanism was created to ensure compliance or alter decisions made by individual physicians (20). For each of these programs audiologists were available for ongoing monitoring and consultation (18-20). Staff involved in the Bhatia et al. (18) study reported that that the training session helped them to prepare families and they felt very comfortable performing the screen. No other studies reported on the training quality or clinician attitude toward, or acceptance of, the program.

Preschool/Day-care Setting

The objectives of studies/programs implemented in preschool/day-care settings varied substantially and are synthesised below.

Hearing screening referral rates by method

Several studies aimed to compare outcomes between different hearing screening instruments/protocols. Two studies examined outcomes between TEOAE and pure tone audiometry (23, 24). Sideris et al. (23) reported no difference in the rate of referral for follow-up between these two methods (21.5% for pure tone, 21% for TEOAE). Yin et al. (24) reported similar findings; 126 of 142 preschool children (93.3%) passed both the TEOAE and the pure tone screen. No child passed the TEOAE and then had a refer on pure tone testing, yielding zero false-negative results; on the other hand, 8 children had a refer on TEOAE but passed pure tone screening. The reported sensitivity of TEOAE screening was 1.0 (95% CI, 0.054-1.00), specificity was 0.94 (0.88-0.97), positive predictive value of a refer was 0.11 (95% CI 0.005-0.49), negative predictive value was 1.00 (0.96-1.00) (24).

Although there was disparity between the ratings of the quality and bias check (24) – “Good” and (23) – “Fair”) the results were quite consistent, except that the Sideris et al. (23) study reported no information about sensitivity and specificity. That said, most of the criteria for an acceptable screening program was reached on evaluation of the Yin et al. study (24). The applicability to the NSW population was considered high but the generalisability was low as the study was conducted within a large, urban, metropolitan school district and located in underserved communities, comprising a relatively large proportion of children who required special education services and with over 70% of Latino/Hispanic descent. Data were lacking in regard to final yield of permanent hearing loss and of cost effectiveness. In spite of some low ratings noted above and clearly the lack of quantity of evidence, hearing screening using TEOAEs within a preschool setting performed by school nurses could be ranked as “Promising” in underserved communities if definitive outcomes were reported. Because such outcomes are not available, it was finally ranked as ‘Not Supported’.

A study conducted in Brazil aimed to evaluate the effectiveness of two screening methods - imitanciometry screening (described as tympanometry and ipsilateral and contralateral acoustic reflexes) and a brief questionnaire, developed to identify children at risk for conductive hearing loss, and compare this data with complete audiologic evaluation (25). The brief questionnaire reportedly had 97.7% sensitivity, 48.5% specificity, and accuracy of 67.6%. Using combined screening tests (imitanciometry and risk questionnaire) administered serially showed better specificity, accuracy and odds ratio when compared with screening separately or when administering tests in parallel.

The study evaluating imitanciometry screening was unique in terms of the tools used and the study location (Brazil), which limits the generalisability of the data. The applicability was considered low as familiarity with imitanciometry is likely much lower than other hearing screening tools. The level of evidence was low and the overall quality and bias check was evaluated as fair. Furthermore the test itself did not rate well in regards to acceptability of what constitutes a screening test. Thus the overall ranking of this screening protocol was deemed “Not supported”.

The only other study directly comparing methods in the preschool setting was based in the USA (26). The aim in this study was to compare the signal to noise ratio between distortion product otoacoustic emissions (DPOAE) and transient evoked otoacoustic emissions (TEOAE) and whether any difference would influence the pass/refer rates; the study also examined the agreement between the emission methods and the tympanometric findings. The authors report that DPOAEs tend to have higher signal to noise ratio than TEOAEs, but there was no significant difference between the pass/refer rates for each method at any frequency. Each OAE method pass/refer rates conflicted with the tympanometric pass/refer rates in 9/33 children (approximately 27% of the time) (26).

The evidence level for use of DPOAE or TEOAE screening in the preschool setting was low in terms of NHMRC standards, the overall quality and bias check was rated as poor, and the acceptability of the screening tool was also low. The generalisability is difficult to determine given the small sample size and limited detail provided as to demographic characteristics, so the applicability becomes almost irrelevant, although presumably since it was conducted in the USA that it would be a similar context to a NSW setting. This hearing screening protocol using TEOAE or DPOAE in a preschool setting was ranked as “Not supported”.

Preschool hearing screening versus sporadic identification

A study in China aimed to compare the age at diagnosis and ages of intervention for cases of delayed-onset hearing loss identified sporadically or via a preschool hearing screening program. The average age of 26 children at the time of diagnosis in the screening group (52.81 ± 13.23 months) was significantly earlier than in the 33 cases identified in the sporadic group (62.03 ± 12.86 months; $p < 0.05$). The age at intervention of children with bilateral moderate to severe hearing loss in the screening group (50.40 ± 10.76 months) was also earlier than in the sporadic group (62.73 ± 13.77 months; $p < 0.05$) (27).

The overall level of evidence was low and the quality and bias rating was judged to be fair. There was inadequate information to accurately assess the acceptability of the screening tool and, whilst the screening program met most of the criteria, there was no information about its cost effectiveness. In view of the issues discussed this program was ranked as “Not supported”.

Repeated OAE and visual otological inspection

A study from the USA aimed to screen underserved children ≤3 years of age for hearing loss using otoacoustic emissions (OAE) technology and to systematically document multi-step screening and diagnostic outcomes. The hearing screen protocol comprised a visual inspection of the ear and up to three OAE screenings over a 2-4 week period (28). The protocol was designed in this way to significantly limit false positive findings by specifying that children be screened multiple times before receiving a diagnostic evaluation, thus minimising potential over-referral to health care providers for transient conditions (such as temporary congestion due to head colds for example). The sample consisted of 4,519 children and 6% of these required medical or audiological follow-up. One hundred and seven children were identified as having hearing loss or disorder of the outer, middle, or inner ear requiring treatment or monitoring. Five of the 107 children had bilateral and 2 had permanent unilateral hearing loss; of these 4 had passed the newborn screening, 2 were not screened, and one did not follow-up on referral.

The NHMRC level of evidence was rated was low and the quality and bias was deemed good. The acceptability of the screening protocol was ultimately difficult to determine as information regarding the sensitivity and specificity was not sufficiently reported. The study was conducted in the USA in an underserved population comprising a high proportion of Hispanic children, so the generalisability to the NSW context as a whole is low, as is the applicability. The generalisability and applicability may be moderate if applied to specific settings within NSW, such as areas with low socioeconomic status or in rural locations, where organisational factors and attitudes to health issues may be more comparable. Based on the evidence overall, this hearing screening protocol was deemed "Not supported".

Operational considerations

Several of the reviewed preschool/day-care setting screening programs reported high levels of failure to follow-up after a failed hearing screen result. For example, follow-up was not conducted or unreported in 48% of children having undergone an audiologic screen at preschool, day-care, or at a Head Start program (29) in New York State. Similarly, 47.5% did not attend a referral for complete audiologic evaluation following a failed screen in a sample of preschool children in Brazil (25).

Non-compliance was an issue in several studies, with age being a significant influence, even up to the age of 4 years (23, 24). Inability of the child to cooperate was also reportedly higher for hearing screening conducted via pure tone screening (62.8%) compared with TEOAE (9.5%) (23).

Testing time was reported for TEOAE and the mean testing time was 43 seconds (range: 10 to 180 seconds) (24).

Estimated cost of TEOAEs screening for a cohort of 1,000 preschool children were as follows: TEOAE machine $5,000, Supplies (probe tips and extra probes) $530, wages $12,250, total $18,030 – total cost per child $18.03 (24).

Workforce & capacity

Staff performing the hearing screening protocol in preschool settings were either school nurses or doctors, audiologists, audiology graduate students, lay screeners, or a combination of these. The two studies that used school nurses or doctors provided staff with 1-2 hours of "hands-on" training by a factory representative in the use of the equipment, which in both instances was Otodynamics Echo Port ILO 288 (24, 30). Eiserman et al. (28) had the hearing screening performed by "lay screeners" and provided them with a six-hour training session as well as access to audiological technical support.

Molecular Testing

The utility of molecular testing for the detection of causes of delayed hearing loss which is missed by current audiometric screening at birth was examined by a study in the USA. There were 3,681 infants examined; 35 (0.95%) had a positive SoundGene™ panel, 16 had mitochondrial mutations, 9 had Pendred mutations, 5 were cytomegalovirus (CMV) DNA positive, 2 had connexin mutations, and 3 had a combination of different mutations. The data demonstrated that infants with an abnormal SoundGene™ panel were at increased risk for hearing loss compared to neonates without mutations. Three (8.6%) of the 35 infants had

persistent hearing loss compared to 5 (0.21%) of 2,398 subjects with no report of mutation. Of the total number of infants examined, 8 (0.22%) had persistent hearing loss, 5 (62.5%) had abnormal newborn audiometric screens, 2 (25%) had an abnormal SoundGene™ panel (1 was CMV positive, 1 had a mitochondrial mutation), and 1 (12.5%) had no identifiable risk factors (31).

The level of evidence was low, but the paper was of good quality and low bias. It was difficult to assess the acceptability of the screening tool as there was not enough information supplied about the process. In regards to generalisability and applicability, this is likely to be high. As such, this was deemed "Promising" as an adjunct to hearing screening programs.

Grey Literature

Although a systematic search of the grey literature was beyond the scope of this rapid review we thought it pertinent to mention a number of programs that either implement a hearing screening protocol beyond the newborn period and/or endorse such a program.

Queensland, Australia: Healthy Kids Check

The Healthy Kids Check (HKC) is an Australian Government initiative to assess 4-year-old children for physical developmental concerns. The HKC is administered by GPs and there are 6 mandatory screening items, including hearing. A recent examination to determine the frequency of health problems as identified by the HCK found that hearing was one of the three most identified developmental problems, along with speech and language and anatomical concerns (32). These data were published from an audit of medical records from 2 Queensland general practices (n=557). Although these data appear particularly relevant to the NSW context, it was not possible to make a judgement on the overall quality of this program as it relates to hearing screening. There were significant missing data as well as inadequate information about sample characteristics, what the screen (if any) actually entailed (i.e. method, time, training, cost-effectiveness) or about diagnostic outcomes amongst children who had a positive screen result. This program was ranked "Unknown" on overall rankings of evidence.

American Academy of Audiology

The American Academy of Audiology (AAA) advocates for the early detection of hearing loss in early childhood and school-aged populations based on the premise that under-identification and lack of appropriate management of hearing loss has significant educational, cognitive, and social implications for an individual child as well as broad economic effects more generally (33). The AAA provides a summary of hearing screening recommendations, which includes advice and guidelines for pure tone screening, tympanometry screening, rescreening, and OAE. They also recommend against acoustic reflex testing, reflectometry, and hearing screening using speech materials. Although the document is substantial it is not adequately underpinned by evidence. As such the recommendations in this document were judged as "Not supported".

New Zealand

The Well Child/Tamariki Ora Programme in New Zealand includes a B4 School check where, amongst other checks, screening audiometry and tympanometry (if required) are used to

identify hearing impairment and/or otitis media effusion. This check is done as soon as possible after the fourth birthday. Repeat screening is provided to those children who require follow-up based on the result on their earlier screening. The possible outcomes on the screen are Not Tested, Pass, Rescreen and Refer. Published research on the B4 School Check appears limited to reporting on the pilot of the check, which indicated that the majority of referrals for hearing were for glue ear (34).

A summary of the overall ranking of each study is illustrated in Appendix 10.

DISCUSSION

This Rapid Review is timely because optimising the developmental and psychosocial outcomes of childhood hearing loss remains of great concern to many.

With the introduction of UNHS, almost all congenital permanent moderate or greater hearing losses are now detected within the first few months of life, along with many cases of milder and unilateral losses. UNHS has demonstrated effectiveness both as a tool and as a program and is now the standard of care throughout the developed world. Beyond UNHS are systems for detecting postnatal or other hearing losses in young children, and the evidence for such was the focus of this review.

While the transient losses of otitis media with effusion remain of concern to many, it is the view of these authors – informed by the literature – that this condition does not meet criteria for the introduction of a screening program. Therefore, the target of this review was the success of post-neonatal screening and surveillance for permanent hearing losses.

Below, we discuss the various tools and programs identified by this rapid review, consider their effectiveness, and make recommendations for each approach.

Targeted Surveillance

The Joint Committee on Infant Hearing (JCIH) first proposed using targeted surveillance as the primary method for monitoring postnatal hearing loss using risk factors in a position statement in 1973. This has since been amended several times in terms of the risk factors included and the recommended frequency of hearing assessments. The JCIH (12) Position Statement identified 11 risk factors associated with hearing loss, with eight of these marked as greater concern for delayed-onset hearing loss. The Position Statement also stipulated that any child with any one of these risk factors should receive at least one diagnostic audiological evaluation by 24 to 30 months of age (12), and for some high-concern risk factors (family history of permanent childhood hearing loss, CMV infection, extracorporeal membrane oxygenation, syndromes associated with progressive hearing loss, neurodegenerative disorders, trauma, postnatal infections associated with sensorineural hearing loss, chemotherapy, and caregiver concern) this surveillance should commence earlier and occur more frequently.

However, a systematic review conducted in 2012 investigating whether there was evidence-based support for targeted surveillance programs using a risk-factor registry to detect postnatal hearing loss (35) was inconclusive, with studies available at the time being based on small sample sizes and from single sites.

The 3 studies included in this review relating to targeted surveillance add to the literature and comprise good quality papers with large sample sizes. The results indicate that targeted hearing surveillance using risk factors is a promising method. Based on the UK audit of well over 2 million babies, it appears that this targeted surveillance should be limited to children with (i) Down Syndrome, (ii) other syndromes known to be associated with hearing loss (e.g. Treacher Collins syndrome, Pendred syndrome, CHARGE syndrome), (iii) craniofacial anomalies, and (iv) congenital infections (particularly CMV).

Several limitations were noted, such as low uptake of referral appointments, delays in surveillance assessment, and loss to follow-up. These were consistent with previous studies that have also identified significant barriers to using this method – i.e. lack of parental cooperation in providing risk factors and attending surveillance appointments (36), difficulties in behaviourally identifying hearing loss in young children (37), and significant proportions of children who develop a hearing loss postnatally not having any obvious risk factor (38, 39).

It is likely that restricting the targeted surveillance to these four readily-identifiable and highly predictive risk factors would reduce the numbers being followed (and therefore costs) and enhance both retention and yield. Significant improvements to ensure follow-up within medical care systems and current practice would be required for this to be effective.

Questionnaire Surveillance

A substantial audit indicated that questionnaire surveillance both in infancy and around school entry is inaccurate and has substantial challenges as a service delivery model. Given the limitations identified by the audit, it could not be supported.

Hearing Screening in Health and Preschool/Day-care Settings

A number of studies examined the feasibility of integrating hearing screening protocols within routine services such as well child visits. As these were studied in the US, the setting was the paediatrician's office, for which there is not an equivalent in Australia. There is no doubt that this can and does identify hearing losses, but also that it does not meet criteria for a screening program. Abnormalities detected are almost invariably the transient conductive losses of otitis media with effusion. Further, there were major operational concerns including high failure rates for children under 5 years (which, if all were referred, would be costly and could overload audiology services) coupled with high rates of non-completed screens in the younger children and loss to follow-up (which would counteract any value of the screen). For all these reasons, hearing screening programs integrated in the primary care setting were not supported.

Broadly, the same issues were encountered for screening in preschool and day-care settings.

At time of writing, Dr Heather Fortnum of Nottingham University is drawing to a close a large UK Health Technology Assessment randomised trial examining the cost-effectiveness of screening for permanent hearing loss in children at school entry (40). Outcomes, expected in 2015, will be very relevant to this rapid review, particularly as English children enter school at an age when Australian children are typically still in preschool.

Additional Considerations

Genetic testing used in conjunction with newborn hearing screening has been proposed to identify children who are at high risk of developing a postnatal hearing loss by virtue of a genetic mutation. The three most common genetic mutations associated with late onset hearing loss are GJB2 (i.e. connexin) deafness, mitochondrial A1555G mutation, and SLC26A4 mutation (i.e. Pendred syndrome) (41).

A number of cases of postnatally developed hearing loss are attributable to genetic mutations (42). Several options are being investigated as a screening tool for detecting genetic mutations, including the development of a diagnostic microarray chip (43), a DNA chip (44),

and a genetic-screening card (45). The costs of these are likely to fall rapidly over the next few years. This approach was classified as promising in this review based on a good quality, low bias paper (31). As more research occurs, this should be reconsidered.

Similarly, a combined approach of newborn hearing screening and CMV screening has been proposed to detect children with congenital CMV who may be at risk of developing a postnatal hearing loss (46). Several methods have been suggested as a screening tool for CMV, including blood (47, 48), saliva (49) or urine samples (50). However, despite the significant amount of research directed toward CMV screening, to date there are no formal policies regarding CMV screening in newborns. To establish recommendations, large-scale population studies are required to establish the efficacy of screening for CMV in newborns (51), and such trials are currently underway both within Australia and overseas.

RECOMMENDATIONS

1. Implement systems for targeted surveillance of young children who pass their newborn hearing screen but have one of the four following risk factors for postnatal hearing loss:

   - Down's syndrome
   - Other syndromes known to be associated with a hearing loss
   - Craniofacial anomaly, and
   - Congenital infection.

   This will require agreement on:

   - Population coverage
   - Clear and well-defined definitions of these risk factors
   - An appropriate workforce to implement detection of risk factors
   - Systems to maximise uptake of follow-up diagnostic assessments
   - Adequate IT and data systems to track and follow these children throughout early childhood
   - How to incorporate surveillance for children who subsequent to the neonatal period experience illnesses or events known to cause hearing loss (see Recommendation 3 below).

2. Watch for forthcoming evidence regarding:
   - Costs and benefits of metabolic and CMV screening
   - Fortmum et al.'s school entry hearing screening trial in England.

3. Audiologic services should continue to be widely and readily available for diagnostic testing of children with possible hearing loss based on a case-finding approach, including:
   - Parent/professional concerns about hearing loss
   - Parent/professional concerns about children's development, especially in language and related abilities
   - Illnesses or events during childhood known to cause hearing loss, e.g. meningitis, head injuries.

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APPENDICES

Appendix 1: Defining the Question - PICO Framework

PICO

The question was formulated within a Population Intervention Comparison Outcome (PICO) framework. Application of a PICO framework helps to structure, contain and set the scope for the research question. Inclusion of intervention (screening models) and comparison components is dependent on the question asked, and may not be appropriate for all question types.

What are the programs and service models that deliver population-based screening and surveillance of hearing for children aged 0-5 years?

PICO format: In children between 0-5 years of age what is the relative effectiveness and efficiency of hearing screening in identifying postnatal hearing loss before school-age.

P Patient, Problem, Population	**I Intervention (*screening*)**	**C Comparison (*optional*)**	**O Outcome (*when defining "more effective" is not acceptable unless it describes how the intervention is more effective*)**
AGE 0-5 GENDER (no specification) DIAGNOSES (none excluded)	Effectiveness: the extent to which the model or program improved the desired outcomes when applied to the population. Efficiency: the cost-effectiveness of the model or program, inclusive of any harms, benefits and costs of the program to individuals and society.		Programs and service models that deliver population-based screening and surveillance

Appendix 2: Information Retrieval

The following is an example of the search strategy conducted in Ovid Medline

Step	Search Terms	No. of Records
S1	exp hearing disorders/	36179
S2	persons with hearing impairments/	1370
S3	exp hearing loss/	30149
S4	otitis media/	5761
S5	otitis media with effusion/	2683
S6	glue ear.mp.	110
S7	Hearing Loss, Conductive/	1782
S8	1 or 2 or 3 or 4 or 5 or 6 or 7	44034
S9	exp hearing tests/	17644
S10	tympanometry.mp.	1008
S11	pure tone screen$.mp.	22
S12	acoustic reflex.mp.	242
S13	acoustic reflectometry.mp.	64
S14	otoacoustic emissions screen$.mp.	10
S15	pneumatic otoscop*.mp.	146
S16	9 or 10 or 11 or 12 or 13 or 14 or 15	18083
S17	8 or 16	51789
S18	hearing screen$.mp. and exp *hearing disorders/	1096
S19	mass screening/ and (exp *hearing disorders/ or hearing screen*.mp.)	571
S20	population surveillance/ or public health surveillance/	38883
S21	17 and (18 or 19 or 20)	1575
S22	(developing countr$ or third world or underdeveloped countr$ or under developed countr$).mp.	56065
S23	exp africa/	126634
S24	22 or 23	170302
S25	21 not 24	1143
S26	limit 25 to ("infant (1 to 23 months)" or "preschool child (2 to 5 years)")	479
S27	(child* or infant* or juvenil* or minor* or preschool* or pre-school* or nursery or pediatric* or paediatric*).af.	1499559
S28	25 and 27	910
S29	26 or 28	910
S30	limit 29 to (english language and yr="2005 - 2014")	441

Appendix 3: Levels of Evidence Hierarchy

NHMRC Evidence Hierarchy: designations of 'levels of evidence' for Screening Intervention

Level	Screening Intervention
Level I	A systematic review of level II studies
Level II	RCT
Level III-1	Evidence obtained from well-designed pseudo-randomised controlled trials (alternative allocation or some other method)
Level III-2	A comparative study with concurrent controls: • Non-randomised, experimental trial • Cohort study • Case-control study
Level III-3	A comparative study without concurrent controls: • Historical control study • Two or more single arm study
Level IV	Evidence obtained from case studies
Level V	*The current tables exclude expert opinion and consensus from an expert committee as they do not arise from scientific investigation*

Appendix 4: Screening Form

Used to code the eligibility of references acquired through search paradigms.

Screen on Title & Abstract

1. EXCLUDE Language: *Exclude if non-English*
2. EXCLUDE Date: *Exclude if published prior to 2004*
3. EXCLUDE Age: *Exclude if age of participants >5, or if neonatal screening*
4. EXCLUDE Study Type: *Exclude if validation study, animal study, review paper, technical report, stand-alone methods paper*
5. EXCLUDE Demographic location: *Exclude if developing country*
6. EXCLUDE Study Group: *Exclude if specific disease/disorder*
7. EXCLUDE Outcome: *Exclude if not population screening or surveillance or outcome data does not report on the screening model or is inappropriate*
8. EXCLUDE Unavailable: *Exclude if full-text version is not readily available*
9. INCLUDE based on title & abstract: *Cannot be excluded so is marked as INCLUDE. Will require retrieval of full paper*

Appendix 5: Quality & Bias Checklist for Observational Studies

Checklist for appraising the quality of Observational Studies*

Completed		
Yes	No	
		1. Study Question
		Clearly focused and appropriate question
		2. Study Population
		Description of study population
		Sample size justification
		3. Comparability of Subjects
		Specific inclusion/exclusion criteria for all groups
		Criteria applied equally to all groups
		Comparability of groups at baseline with regard to disease status and prognostic factors
		Study groups comparable to non-participants with regard to confounding factors
		Use of concurrent controls
		Comparability of follow-up among groups at each assessment
		4. Exposure or Intervention
		Clear definition of exposure
		Measurement method standard, valid and reliable
		Exposure measured equally in all study groups
		5. Outcome measures
		Primary/secondary outcomes clearly defined
		Outcomes assessed blind to exposure or intervention
		Method of outcome assessment standard, valid and reliable
		Length of follow-up adequate for question
		6. **Statistical Analysis**
		Statistical tests appropriate
		Multiple comparisons taken into consideration
		Modelling and multivariate techniques appropriate
		Power calculation provided
		Assessment of confounding
		Dose-response assessment if appropriate
		7. **Results**
		Measure of effect for outcomes and appropriate measure of precision
		Adequacy of follow-up for each study group
		8. **Discussion**
		Conclusions supported by results with possible biases and limitations taken into consideration

*where not all criteria were relevant for each study they were assessed on the domains that applied and an overall rating ascertained.

Appendix 6: Criteria for the Appraisal of Screening Tests and Screening Programs

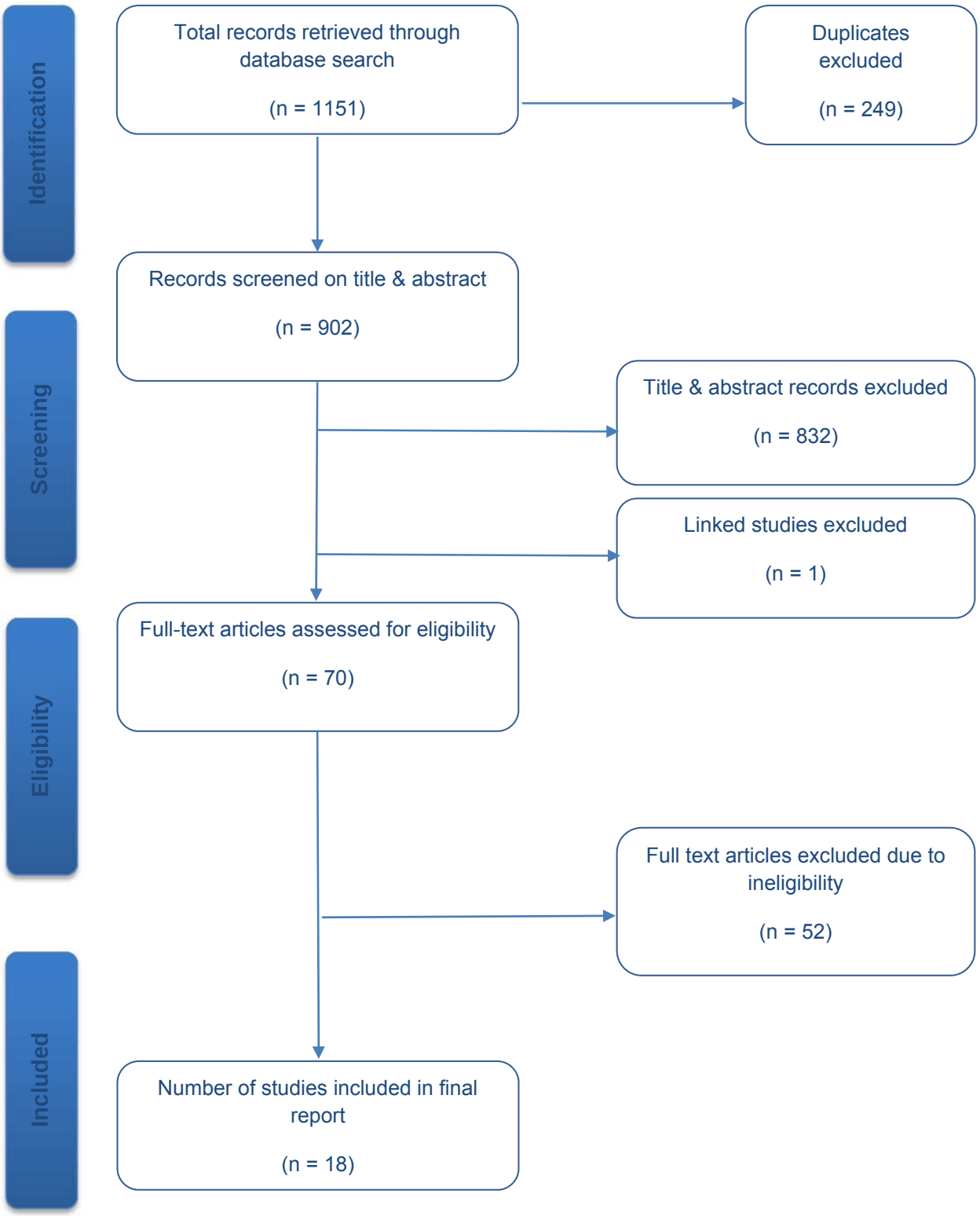
Criteria for a Screening Test

	Yes/No/Unknown
Simple, quick & easy to interpret	
Acceptable to public	
Accurate	
Repeatable	
Sensitive	
Specific	

Criteria for a Screening Program

	Yes/No/Unknown
Important health problem	
Accepted treatment	
Facilities for diagnosis and treatment	
Latent or early symptomatic stage	
Suitable test or examination	
Test acceptable to the population	
Natural history adequately understood	
Agreed policy on whom to treat	
The cost of case-finding balanced with expenditure on medical care as a whole	

Appendix 7: PRISMA Flow Chart Representing the Number of Records Retrieved at Each Stage of the Rapid Review Assessment Process



Appendix 8: Evidence Summary

Summary of Evidence: Targeted Surveillance

Study	Sample Characteristics	Study Details	Condition	Method/tool	Workforce
Beswick (2012)	**Origin of study** • Australia **Study setting** • targeted surveillance [Info] *tertiary hospitals, non-tertiary hospitals, private audiology/early intervention clinics* **Age group** • 3 to 12 months [Info] *Children are seen for their first targeted surveillance appointment between 3 and 12 months of age, depending on the risk factor. Family history - 6 mths, then every 6 mths until 2 yr old, discharge assessment at 3yr Infection - 3 mths, f/u appt at 6 mths, then every 6 mths until 2 yr. Remaining risk factors - a single appt at 9–12 mths* **Sample characteristics of study group** • % male [Info] *52.8%* • n= [Info] *7320 (2.82% of 261,328) children were referred for targeted surveillance in Queensland*	**Description of study type** • screening intervention	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Tympanometry [Info] *Dependent on risk factor: family history infection syndrome prolonged ventilation bacterial meningitis low birth weight severe asphyxia craniofacial anomalies hyperbilirubinemia professional concern* • Transient evoked otoacoustic emissions (TEOAEs) [Info] *Dependent on risk factor: family history infection syndrome prolonged ventilation bacterial meningitis low birth weight severe asphyxia craniofacial anomalies hyperbilirubinemia professional concern* • Behav. assessment (VRA or play audiometry [Info] *Dependent on risk factor: family history infection syndrome prolonged ventilation bacterial meningitis low birth weight severe asphyxia craniofacial anomalies hyperbilirubinemia professional concern* • Auditory brainstem response (ABR) [Info] *Dependent on risk factor: infection* Instrumental requirements • Add details [Info] *risk factor registry based on both the JCIH risk factor list and the risk factor registry utilised in the United Kingdom*	**Workforce & capacity issues** • Staff [Info] *risk registry requires staff across Queensland to input data at the site level into centralised database UNHS - uses enrolled or registered nurses or midwives trained in aABR Newborn screen parents interviewed by nurse/midwife*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Workforce
Beswick (2013)	**Origin of study** • Australia **Study setting** • targeted surveillance **Age group** • 3 to 12 months **Sample characteristics of study group** • % male [Info] *1113 (52.8%)* • Birth weight & GA [Info] *,34 wk for 1003 (47.6%) children,$34 wk for 1096 (52.0%) children, and not stated for eight (0.4%) children* • Race/ethnicity [Info] *Indigenous status was neither Aboriginal nor Torres Strait Islander (TSI) for 1975 (93.7%) children, Aboriginal and/or TSI for 92 (4.4%) children, and not stated for 40 (1.9%) children* • n= [Info] *2107* • Risk factors [Info] *Risk factors for individual child range 1-5 1457 children; 69.2% one risk factor only*	**Description of study type** • screening intervention [Info] *population-based retrospective cohort study*	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	NA/Not Required	NA/Not Required

Study	Sample Characteristics	Study Details	Condition	Method/tool	Workforce
Wood (2013)	**Origin of study** • UK **Study setting** • targeted surveillance **Sample characteristics of study group** • n= [Info] *69,050 children eligible for targeted surveillance*	NA/Not Required	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Instrumental requirements** [Info] *Screening is carried out using standard techniques and protocols and with equipment that has been approved for use within NHSP (England). There are separate protocols for well babies and NICU/SCBU babies. Detailed protocols and pathways are available at http://hearing.screening.nhs.uk.*	NA/Not Required

Summary of Evidence: Questionnaire Surveillance

Study	Sample Characteristics	Study Details	Condition	Method/tool	Workforce
Yoong (2008)	**Origin of study** • UK **Study setting** • Data Audit **Age group** • Add age range [Info] *7 to 9 months & School-aged children*	A comparative study without concurrent controls • Historical control study [Info] *review of screening program*	NA/Not Required	**Hearing assessment method** • Questionnaire [Info] *Hearing Surveillance Questionnaire* Instrumental requirements • Add details [Info] *SystmOne*	**Workforce & capacity issues** • Staff [Info] *steering group was identified for their specific professional role in facilitating, monitoring and evaluation of the data collection process This steering group has overall responsibilities for a hearing surveillance programme that has to be consistent with the recommendations of the NHSP*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Workforce
					executive committee guidance document as well as the Hall report

Summary of Evidence: Health Care Setting

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
Bhatia (2013)	**Origin of study** • USA **Study setting** • Federally qualified health centers **Age group** • 0-3 years **Sample characteristics of study group** • Race/ethnicity [Info] *Participating clinics served a predominately Hispanic population who had either Medicaid or were uninsured (underserved population)* • n=	**Description of study type** • Other (write in) [Info] *prospective chart review*	**Hearing-related issue/condition tested** • Risk flag for sensorineural hearing loss	**Hearing assessment method** • Tympanometry • Otoacoustic emissions screening • Risk Factors Questionnaire	**Operational & administration parameters** • Add detail [Info] *A private nonprofit audiology and early intervention center partnered with the clinics to provide audiology support, equipment, training, protocol development, data collection, diagnostic testing, and case management.*	**Workforce & capacity issues** • Staff • Training [Info] *half-day training session on Infant-toddler hearing screening (ITHS) with ongoing monitoring and consultation from audiology staff.*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
	[Info] *2397*					
Ciccia (2011)	**Origin of study** • USA **Study setting** • health clinic **Age group** • Up to age 6 years **Sample characteristics of study group** • n=*411* • demographics [Info] *The organizations involved in the study had a large patient base of minority, low-income families.*	**Description of study type** • screening intervention	NA/Not Required	**Hearing assessment method** • Tympanometry [Info] *1st year - facilitator relayed results to audiologist for realtime interpretation. 2nd year facilitator used the PC-based tympanometer with the clinician site audiologist accessing the results using remote desktop computing (NTRconnect) for realtime interpretation.* • Behav. assessment (VRA or play audiometry [Info] *1st year - clinic facilitator used a portable audiometer & performed the screening with videoconferencing supervision by the audiologist. 2nd year - clinic facilitator used a PC-based audiometer & performed the screening via videoconferencing with the results automatically provided for realtime interpretation. A routine audiometry response (hand raise) was employed when the child was able or conditioned play audiometry was used.* • Brief parental interview [Info] *parental concerns, relevant medical, family and developmental*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *screening performed by clinic facilitator with supervision provided via videoconferencing*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
				information • Distortion product otoacoustic emissions (DPOAE) [Info] *year 1 - clinic facilitator performed the DPOAE with the results provided to the audiologist via videoconferencing for realtime interpretation. Year 2 - clinic facilitator used a PC-based DPOAE screener & the clinician site audiologist accessed the results using remote desktop computing for realtime interpretation.* **Instrumental requirements** [Info] *Computer kiosks were set up at the two clinics, each equipped with Dell laptops with 43 cm screens, web cameras (Microsoft Life Cam VX-3000) and Skype 3.8 for Windows. Equipment used for the audiology screening was: (1) acoustic impedance audiometer (Earscan); (2) automatic handheld otoacoustic emissions instrument (OtoRead); (3) otoscope (Welch Allyn); (4) diagnostic audiometer (MedRx Avant A2D); (5) standard typmanometer (Maico Ero-Scan Pro DP).*		
Foust (2013)	**Origin of study** • USA	**Description of study type**	**Hearing-related issue/condition**	**Hearing assessment method** • Tympanometry	**Operational & administration**	**Workforce & capacity issues**

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
	Study setting • health clinic [Info] *primary care setting* • school-based clinic [Info] *primary care setting* **Age group** • Add age range [Info] *0-5 years* **Sample characteristics of study group** • % male [Info] *40* • Race/ethnicity [Info] *Hispanic 351 (41) White 99 (12) Pacific Islander 66 (8) Other 33 (4) Ethnicity not stated, n (%) 297 (35)* • n= [Info] *846 children, 619 (73%) were served in the 2 school-based clinics 227 (27%) were served in the community clinic* • demographics [Info] *693 (82%) - families incomes at or below the federal poverty*	• screening intervention	**tested** • Risk flag for postnatal hearing loss	• Otoacoustic emissions screening • Visual inspection **Instrumental requirements** [Info] *handheld Biologic AuDX distortion product (DP) OAE instruments The cost of equipment was ~$3700 per unit. Disposable pediatric foam probe covers (~$1 per child)*	**parameters** [Info] *average time required for OAE screening based on the first 350 children screened, was 4 mins per child*	• Staff [Info] *4 medical assistants (MAs)* • Training [Info] *pediatric audiologist provided training to 4 medical assistants (MAs) Standardized training materials (video, printed manuals and materials) from the Early Childhood Hearing Outreach Initiative were used during a 4-hour training session, along with live demonstrations and hands-on practice Audiologist was also available to provide ongoing technical support and consultation as needed*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
	level;					
Halloran (2005)	**Origin of study** • USA **Study setting** • health clinic **Age group** • Add age range [Info] *3 to 19 years* **Sample characteristics of study group** • Race/ethnicity [Info] *< 3% (n=29) of children race/ethnicity other than African American or white (excluded from analyses)* • n= [Info] *convenience sample of 1,061* • demographics [Info] *< 1% (n=11) lacked insurance (excluded from analyses)* • exclusions [Info] *Children currently under the care of an*	**Description of study type** • screening intervention	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Pure tone screening • Tympanometry • Behav. assessment (VRA or play audiometry • Risk Factors Questionnaire	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *Practices provided with RAs* • Training [Info] *Prior to study - guidelines distributed to the practices during a training session conducted by the PI & assisting audiologist with the clinic staff and/or physicians at each practice.*

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
	audiologist					
Halloran (2009)	**Origin of study** • USA **Study setting** • health clinic	comparative study with concurrent controls • Cohort study [Info] *prospective cohort study*	NA/Not Required	NA/Not Required	NA/Not Required	NA/Not Required
Lim (2013)	**Origin of study** • USA **Study setting** • hospital [Info] *Pediatrix - provides the staff, administrative support and management necessary for high-quality newborn hearing screen services to hospital partners* **Sample characteristics of study group** • % male [Info] *52%*	**Description of study type** • screening intervention [Info] *median birth weight 3,255 g (2,440-3,880) median gestational age 39 weeks (36-40)*	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Molecular testing **Instrumental requirements** • Add details [Info] *SmartScreener-Plus 2 automated auditory brainstem response screening system*	NA/Not Required	NA/Not Required

Study	Sample Characteristics	Study Details	Condition	Method/tool	Operational Matters	Workforce Issues
	• Birth weight & GA [Info] *median birth weight was 3,255 g (2,440-3,880g, 10th 90th percentile)* • Race/ethnicity • exclusions [Info] *major congenital anomalies*					

Summary of Evidence: Preschool/Day-care Setting

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
Chen (2013)	**Origin of study** • China **Study setting** • preschool/day-care centre **Age group** • 3 to 6 years [Info] *4.86 +-1.67 years* **Sample characteristics of study group** • % male [Info] *14,865 boys and 13,681 girls* • n= [Info] *28,546 of 29,775 eligible* • exclusions [Info] *significant clinical symptoms such as mental retardation, craniofacial anomalies, and cerebral palsy*	**Description of study type** • Other (write in) [Info] *cross-sectional, representative cluster sample survey*	NA/Not Required	**Hearing assessment method** • Transient evoked otoacoustic emissions (TEOAEs) **Instrumental requirements** • Add details [Info] *Otodynamics ILO88 Echoport Otoacoustic Emission Test System and Quickscreen software*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *school nurses & doctors Audiologist provided consultation* • Training [Info] *approx 2 hours of ' hands-on ' training by the factory rep on the use of the Otodynamics ILO88 Echoport Otoacoustic Emission Test System and the Quickscreen software*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
Dille (2007)	**Origin of study** • USA **Study setting** • preschool/day-care centre **Age group** • 0-5 years • Add age range [Info] *4mths to 4yrs, 4mths* **Sample characteristics of study group** • % male [Info] *18 boys, 15 girls* • n= [Info] *38 enrolled* • exclusions [Info] *only if refused testing*	A comparative study without concurrent controls • Two or more single arm study	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss [Info] *pass/refer rates*	**Hearing assessment method** • Tympanometry • Transient evoked otoacoustic emissions (TEOAEs) • Distortion product otoacoustic emissions (DPOAE) Instrumental requirements • Add details [Info] *Tympanograms obtained using a GSI 37 autotymp screener Avail. probe tone freq - 220 Hz TEOAE & DPOAE - same probe assembly: an Otodynamics SGD-type pediatric probe. Otodynamics ILO88 DP + TEOAE version 5.6 software was used. Quickscreen used to obtain TEOAE response. The Otodynamics ILO92 software was used to obtain the DPOAE*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *2 experienced audiologists (TG and MD) & 1 audiology graduate student (BE) performed all of the testing*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
				response		
Eiserman (2008)	**Origin of study** • USA **Study setting** • preschool/day-care centre **Age group** • 0-3 years **Sample characteristics of study group** • Race/ethnicity [Info] *Ethnicity Hispanic 2437 (54%) Caucasian 1331 (29%) American Indian 271 (6%) African American 158 (4%) Bi-racial 98 (2%) Asian 21 (1%) Unknown 203 (4%)* • n= [Info] *4,519*	NA/Not Required	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Otoacoustic emissions screening [Info] *protocol was designed to significantly limit false positive findings by specifying that children not passing the initial OAE screening be screened up to two more times before receiving an evaluation over a 2-4 week period* • Visual inspection Instrumental requirements • Add details [Info] *Bio-logic AuDX distortion product otoacoustic emissions instruments*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *lay screeners* • Training [Info] *Standardized procedures and manuals were used to train all screeners in performing OAE screening and adhering to a follow-up protocol*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
Lü (2014)	**Origin of study** • China **Study setting** • preschool/day-care centre **Age group** • 3 to 6 years **Sample characteristics of study group** • % male [Info] *screening - 14 boys, 12 girls sporadic - 16 boys, 17 girls* • n= [Info] *screening group n=26 sporadic n=33*	NA/Not Required	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	NA/Not Required	NA/Not Required	NA/Not Required
Samelli (2012)	**Origin of study** • Brazil **Study setting** • preschool/day-care	A comparative study without concurrent controls • Two or more single arm study	**Hearing-related issue/condition tested** • Risk flag for conductive	**Hearing assessment method** • Pure tone screening • imitanciometry [Info] *tympanometry and ipsilateral acoustic reflexes scan*	NA/Not Required	NA/Not Required

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
	centre **Age group** • 3 to 6 years **Sample characteristics of study group** • n= [Info] *507*		hearing loss	• Questionnaire Instrumental requirements • Add details [Info] *Otoflex 100 Questionnaire acoustic booth*		
Serpanos (2007)	**Origin of study** • USA **Study setting** • preschool/day-care centre **Age group** • Add age range [Info] *3 to 5 years* **Sample characteristics of study group** • % male [Info] *male (n=645), female (n = 607) gender*	**Description of study type** • screening intervention	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Pure tone screening • Tympanometry Instrumental requirements • Add details [Info] *single test station consisted of one piece of equipment (portable audiometer or tympanometer) set on a table and two surrounding chairs tympanometer (Grason-Stadler 1737) - 1 test station Beltone 119, Beltone Scout: TDH 50 earphones, MX51*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *graduate-level audiology or speech-language pathology students under the supervision of an ASHA-certified audiologist licensed by NY state*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
	unspecified (n = 64) • n= [Info] *34,979 children*			*cushions; Grason-Stadler 1717: TDH 39 earphones, MX51 cushions - other 3 to 4 test stations*		
Sideris (2006)	**Origin of study** • USA **Study setting** • preschool/day-care centre **Age group** • Add age range [Info] *2 years 1 month to 5 years 10 months* **Sample characteristics of study group** • n= [Info] *200*	A comparative study without concurrent controls • Two or more single arm study	**Hearing-related issue/condition tested** • Risk flag for postnatal hearing loss	**Hearing assessment method** • Pure tone screening • Tympanometry • Transient evoked otoacoustic emissions (TEOAEs) • Behav. assessment (VRA or play audiometry **Instrumental requirements** • Add details [Info] *Beltone 120 portable audiometer Otodynamics ILO88 Echoport Otoacoustic Emission Test System and the Quickscreen software Grason Stadler GSI 33 Middle Ear Analyzer*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *Experienced, certified, pediatric audiologists*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
Yin (2009)	**Origin of study** • USA **Study setting** • preschool/day-care centre **Age group** • Add age range [Info] *2 to 6 years* **Sample characteristics of study group** • Race/ethnicity [Info] *Over 70% of the children were of Latino/Hispanic descent Roughly equal numbers of African American and Asian/Pacific Islanders rounding out the rest* • n= [Info] *convenience sample - approx 1200 attending 12 preschools* • demographics [Info] *Approx 10% of children received special*	A comparative study without concurrent controls • Historical control study	NA/Not Required	**Hearing assessment method** • Pure tone screening [Info] *Preschool children first received TEOAE screening Within 3 months, children were rescreened using pure tone audiometry by 2 school audiologists* • Transient evoked otoacoustic emissions (TEOAEs) **Instrumental requirements** • Add details [Info] *Otodynamics Echo Port ILO 288*	NA/Not Required	**Workforce & capacity issues** • Staff [Info] *school nurse, nurse coordinator, and pediatrician audiologist at Children's Hospital Los Angeles provided further consultation school audiologists* • Training [Info] *1 hour of "hands-on" training by the factory representative on the use of the Otodynamics Echo Port ILO 288*

Study	Sample Characteristics	Study Details	Condition	Method/Tool	Operational Matters	Workforce Issues
	education services					

Appendix 9: Evaluation of the Evidence: Targeted Surveillance

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
Beswick (2012)	**Origin of study** • Australia **Study setting** • targeted surveillance [Info] *tertiary hospitals, non-tertiary hospitals, private audiology/early intervention clinics*	**Description of study type** • screening intervention	**Level of evidence** • NHMRC - cannot be classified **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • yes **Sensitive** • yes **Specific** • yes	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes **Latent or early symptomatic stage** • no **Suitable test or examination** • yes **Test acceptable to the population** • yes **Natural history adequately understood** • yes **Agreed policy on whom to treat** • yes **The cost of case-finding balanced with expenditure on medical care as a whole** • no [Info] *unlikely*
Beswick (2013)	**Origin of study** • Australia **Study setting** • targeted surveillance	**Description of study type** • screening intervention [Info] *population-based retrospective cohort study*	**Level of evidence** • NHMRC - cannot be classified **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • yes	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Sensitive • no **Specific** • no	**Latent or early symptomatic stage** • no **Suitable test or examination** • yes **Test acceptable to the population** • yes **Natural history adequately understood** • yes **Agreed policy on whom to treat** • yes **The cost of case-finding balanced with expenditure on medical care as a whole** • no
Wood (2013)	**Origin of study** • UK **Study setting** • targeted surveillance	NA/Not Required	**Level of evidence** • NHMRC - cannot be classified **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • no **Acceptable to public** • no **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes **Latent or early symptomatic stage** • yes **Suitable test or examination** • yes **Test acceptable to the population** • yes **Natural history adequately understood** • yes

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
					Agreed policy on whom to treat • yes **The cost of case-finding balanced with expenditure on medical care as a whole** • yes

Evaluation of the Evidence: Questionnaire Surveillance

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
Yoong (2008)	**Origin of study** • UK **Study setting** • Data Audit	**A comparative study without concurrent controls** • Historical control study [Info] *review of screening program*	**Level of evidence** • level III-3 **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes [Info] *infant* • no [Info] *school-entry* **Acceptable to public** • yes [Info] *infant & school-entry* **Repeatable** • unknown [Info] *infant & school-entry* **Sensitive** • no [Info] *infant & school-entry* **Specific** • yes [Info] *infant & school-*	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • no **Latent or early symptomatic stage** • yes **Suitable test or examination** • no **Test acceptable to the population** • yes **Natural history adequately understood** • yes **Agreed policy on whom to treat** • no **The cost of case-finding balanced with expenditure on medical care as a whole** • no

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				entry	

Evaluation of the Evidence: Primary Care Setting

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
Bhatia (2013)	**Origin of study** • USA **Study setting** • Federally qualified health centers	**Description of study type** • Other (write in) [Info] *prospective chart review*	**Level of evidence** • level III-3 **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	NA/Not Required
Ciccia (2011)	**Origin of study** • USA **Study setting** • health clinic	**Description of study type** • screening intervention	**Level of evidence** • level IV **Paper: Overall Grading** • Fair • Poor	**Simple, quick & easy to interpret** • unknown **Acceptable to public** • yes **Repeatable** • yes **Sensitive** • unknown **Specific** • unknown	NA/Not Required

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
Foust (2013)	**Origin of study** • USA **Study setting** • health clinic [Info] *primary care setting* • school-based clinic [Info] *primary care setting*	**Description of study type** • screening intervention	**Level of evidence** • level III-3 **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • yes **Sensitive** • unknown **Specific** • unknown	NA/Not Required
Halloran (2005)	**Origin of study** • USA **Study setting** • health clinic	**Description of study type** • screening intervention	**Level of evidence** • level III-3 **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • no **Repeatable** • unknown [Info] *not reported* **Sensitive** • unknown [Info] *not reported* **Specific** • unknown [Info] *not reported*	NA/Not Required
Halloran (2009)	**Origin of study** • USA **Study setting** • health clinic	**comparative study with concurrent controls** • Cohort study [Info] *prospective cohort study*	**Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • no **Acceptable to public** • no **Repeatable** • unknown	NA/Not Required

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Sensitive • no **Specific** • no	
Lim (2013)	**Origin of study** • USA **Study setting** • hospital [Info] *Pediatrix - provides the staff, administrative support and management necessary for high-quality newborn hearing screen services to hospital partners*	**Description of study type** • screening intervention [Info] *median birth weight 3,255 g (2,440-3,880) median gestational age 39 weeks (36-40)*	**Level of evidence** • level III-3 **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	NA/Not Required

Evaluation of the Evidence: Preschool Setting

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
Chen (2013)	**Origin of study** • China **Study setting** • preschool/day-care centre	**Description of study type** • Other (write in) [Info] *cross-sectional, representative cluster sample survey*	**Level of evidence** • NHMRC - cannot be classified **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • unknown [Info] *not reported*	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes **Latent or early symptomatic stage** • unknown **Suitable test or examination** • yes

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Specific • unknown [Info] *not reported*	**Test acceptable to the population** • yes **Natural history adequately understood** • unknown **Agreed policy on whom to treat** • yes **The cost of case-finding balanced with expenditure on medical care as a whole** • unknown
Dille (2007)	**Origin of study** • USA **Study setting** • preschool/day-care centre	**A comparative study without concurrent controls** • Two or more single arm study	**Level of evidence** • level III-3 **Paper: Overall Grading** • Poor	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	NA/Not Required
Eiserman (2008)	**Origin of study** • USA **Study setting** • preschool/day-care centre	NA/Not Required	**Level of evidence** • level IV **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • yes **Sensitive** • unknown	NA/Not Required

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Specific • unknown	
Lü (2014)	**Origin of study** • China **Study setting** • preschool/day-care centre	NA/Not Required	**Level of evidence** • level III-3 **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • unknown **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes **Latent or early symptomatic stage** • unknown **Suitable test or examination** • yes **Test acceptable to the population** • yes **Natural history adequately understood** • yes **Agreed policy on whom to treat** • yes **The cost of case-finding balanced with expenditure on medical care as a whole** • unknown
Samelli (2012)	**Origin of study** • Brazil **Study setting** • preschool/day-care centre	**A comparative study without concurrent controls** • Two or more single arm study	**Level of evidence** • level III-3 **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • yes **Acceptable to public** • no **Repeatable** • no	NA/Not Required

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Sensitive • unknown **Specific** • unknown	
Serpanos (2007)	**Origin of study** • USA **Study setting** • preschool/day-care centre	**Description of study type** • screening intervention	**Level of evidence** • level IV **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • no **Acceptable to public** • no **Repeatable** • unknown **Sensitive** • unknown **Specific** • unknown	NA/Not Required
Sideris (2006)	**Origin of study** • USA **Study setting** • preschool/day-care centre	**A comparative study without concurrent controls** • Two or more single arm study	**Level of evidence** • level III-3 **Paper: Overall Grading** • Fair	**Simple, quick & easy to interpret** • yes [Info] *TEOAE* • no [Info] *pure tone* **Acceptable to public** • yes [Info] *TEOAE* • no [Info] *Pure tone* **Repeatable** • unknown [Info] *TEOAE & pure tone* **Sensitive** • unknown [Info] *TEOAE & pure tone*	NA/Not Required

Study	Setting	Study Type	Level of Evidence	Screening Test	Screening Program
				Specific • unknown [Info] *TEOAE & pure tone*	
Yin (2009)	**Origin of study** • USA **Study setting** • preschool/day-care centre	**A comparative study without concurrent controls** • Historical control study	**Level of evidence** • level III-3 **Paper: Overall Grading** • Good	**Simple, quick & easy to interpret** • yes **Acceptable to public** • yes **Repeatable** • unknown **Sensitive** • no **Specific** • yes	**Important health problem** • yes **Accepted treatment** • yes **Facilities for diagnosis & treatment** • yes **Latent or early symptomatic stage** • yes **Suitable test or examination** • yes **Test acceptable to the population** • yes **Natural history adequately understood** • yes **Agreed policy on whom to treat** • no **The cost of case-finding balanced with expenditure on medical care as a whole** • yes

Appendix 10: Citation List by Ranking

Type of Program/Service Model	Studies
Supported	
	None
Promising	
• Targeted Surveillance	• Beswick, Driscoll, Kei, & Glennon (2012) • Beswick, Driscoll, Kei, Khan, & Glennon (2013) • Wood, Davis, & Sutton (2013)
• Molecular Testing	• Lim, Clark, Kelleher, Lin & Spitzer (2013)
Unknown	
• Preschool/Day-care Setting	• Serpanos & Jarmel (2007) • Chen, Fu, Luo, Zhang, & Yang (2013)
Not Supported	
• Questionnaire Surveillance	• Yoong & Spencer (2008)
• Primary Care Setting	• Bhatia, Mintz, Hecht, Deavenport, & Kuo (2013) • Ciccia, Whitford, Krumm, & McNeal (2011) • Foust, Eiserman, Shisler, & Geroso (2013) • Halloran, Wall, Evans, Hardin, & Woolley (2005) • Halloran, Hardin, & Wall (2009) • Lim et al., (2013)
• Preschool/Day-care Setting	• Dille, Glattke, & Earl (2007) • Eiserman, Hartel, Shisler, Buhrmann, White, & Foust (2008) • Lü, Huang, Ma, Li, Mei, Yao, et al. (2014) • Samelli, Rabelo, Pereira, Portela, Sanches, & Neves-Lobo (2012) • Sideris & Glattke (2006) • Yin, Bottrell, Clarke, Shacks, & Poulsen (2009)