Alberta STE Report

The Effectiveness and Safety of Preschool Hearing Screening Programs

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THE EFFECTIVENESS AND SAFETY OF PRESCHOOL HEARING SCREENING PROGRAMS

Alberta STE Report: Policy-driven Health Technology Assessment reports that include an analysis of the social and system demographics, technological effectiveness and economic implications of a health technology. The reports are written under contract with the Alberta Health Technologies Decision Process and contextualized for use in Alberta.

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EXECUTIVE SUMMARY

Technology Effects and Effectiveness

Background

This health technology assessment report has been produced in response to a request from Alberta Health and Wellness (AHW) as part of the Alberta Health Technologies Decision Process (AHTDP) to perform an evaluation of the scientific evidence on the safety, performance, and effectiveness of universal and targeted preschool hearing screening (PHS) to inform the Infant and Preschool Screening Framework being developed by the Community and Public Health (CPH) Division of AHW.

Objectives

The report has two objectives.

1. To examine the safety, performance, and effectiveness of **universal preschool hearing screening** under the following two scenarios:
   (a) UNHS (universal newborn hearing screening) is already in place.
   (b) UNHS is not in place.
2. To examine the safety, performance, and effectiveness of **targeted preschool hearing screening** under the following two scenarios:
   (a) UNHS is already in place.
   (b) UNHS is not in place.

Results

A structured search of electronic bibliographic databases from 2002 to 2012 and a Google search identified two rapid reviews, two systematic reviews, and three primary studies addressing various aspects of preschool hearing screening programs. One systematic review examined the effectiveness of screening children in the general population to identify those requiring treatment for otitis media with effusion (OME) within the first 4 years of life. Two prospective cohort studies assessed the contribution of targeted screening to the identification of hearing loss (HL) in preschool-aged children in traditionally medically underserved regions of the United States that had universal newborn hearing screening (UNHS) programs. All studies examined screening within the context of UNHS. No studies were identified that assessed universal or targeted PHS in the absence of a UNHS program.

Performance of universal hearing screening programs was assessed in a national survey in the United Kingdom, conducted as part of a National Health Service health technology assessment (HTA).[1] Universal screening was assessed also in one retrospective cohort study,[1] while targeted screening was assessed in two prospective cohort studies.[2,3] Overall, referral rates were high, ranging from about 8% or more of those screened, and the yield (those with confirmed hearing loss) represented less than 1% of the population screened. This evidence limits the conclusions that can be drawn about the contribution of universal or targeted PHS to the identification of hearing loss, because of uncertainty regarding the prevalence of hearing loss in populations with UNHS programs and because of differences among screening settings, ages of the study populations, screening tests and protocols.

One retrospective study[1] and one systematic review[4] containing three randomized controlled trials (RCTs) provided evidence on the potential effectiveness of PHS on language and developmental outcomes. None of the study results indicated a statistically significant or clinical meaningful difference between those who were screened and those who were not. No studies reported outcomes concerning the safety of either universal or targeted screening.

Conclusions

No benefit is demonstrated for universal preschool hearing screening within the context of a UNHS program. Potential benefit may exist for targeted screening of at-risk youth; however, the realization of these benefits may be hampered by significant challenges in ensuring timely referral, diagnosis, and treatment. No identified studies evaluated the performance and effectiveness of PHS programs in the absence of UNHS.

Economics Analysis

Objective and Method

The objective was to compare the cost-effectiveness of various strategies used in preschool hearing screening, through a review of the published economic literature.

Results

Potentially cost-effective strategies included universal school entry screening with pure-tone sweep tests (SES-PTS), composite universal school entry screening, and high-accuracy targeted school entry screening; all were associated with additional costs and improved outcomes compared to no screening. Of these, high-accuracy targeted school entry screening may be the most cost effective, but its applicability is dependent upon whether the additional effectiveness is worth the additional costs. Generalizability of the results is limited.

Conclusion

Limited published economic evidence was available regarding the cost effectiveness of hearing screening in preschool–aged children.

ABBREVIATIONS

AAA	American Academy of Audiology
AHTDP	Alberta Health Technologies Decision Process
BCEHP	British Columbia Early Hearing Program
dB	decibels
DPOAE	distortion product otoacoustic emissions
HTA	health technology assessment
HTSPU	Health Technologies and Services Policy Unit
Hz	hertz
NHS	National Health Service
OAE	otoacoustic emissions
OIHP	Ontario Infant Hearing Program
PHS	preschool hearing screening
QALY	quality-adjusted life year
SES	school entry screening
SES-C	composite SES
SES-PQ	SES using parental questionnaire only
SES-PTS	SES using pure-tone sweep audiometry only
SES-SW	SES using spoken word tests only
SES-T	SES using tympanometry only
TEOAE	transient evoked otoacoustic emissions
UNHS	universal newborn hearing screening

GLOSSARY

The glossary terms listed below were obtained and adapted from:
www.phsa.ca/AgenciesAndServices/Services/BCEarlyHearing/ForFamilies/Glossary.htm

Audiologist: A health professional who identifies people who have hearing problems, and works with these people to help to improve their communication. This includes diagnosing hearing loss and fitting hearing aids. Audiologists in Canada have a minimum of a master's degree and are certified by the Canadian Association of Speech–Language Pathologists and Audiologists.

Audiometer: A type of electronic equipment used to test hearing.

Bilateral hearing loss: Hearing loss in both ears.

Conductive hearing loss: A type of hearing loss characterized by problems with the outer or middle ear. An example of an outer ear problem is atresia, where there is no opening to the ear canal. Middle ear problems can be the result of fluid in the middle ear, or there can be something

wrong with the three tiny bones in the middle ear. Conductive hearing loss may be temporary when it is a problem that can be medically treated, such as fluid in the middle ear.

Congenital hearing loss: A hearing loss present at birth, associated with the birth process, or which develops in the first few days of life.

Decibels (dB): Intensity (loudness) of sound is measured in decibels. For instance, 10dB is a very quiet sound and 100dB is a very loud sound.

False positive: A test outcome indicating the presence of a disease or condition when, in fact, that disease or condition is not present.

Impedance/immittance testing: A hearing test during which a small probe is placed in the ear to determine whether a problem exists in the middle ear.

Inner ear: The part of the ear that contains the cochlea and the auditory nerve, as well as the balance organ.

Lost to follow up: This refers to when an individual is not seen for follow-up procedures once having been identified with, or at risk of, hearing impairment. It can be due to such factors as low parental compliance, movement of the individual to another province, lack of services available, no tracking systems in place, and so on.

Middle ear: The middle section of the ear that contains three tiny bones, through which sound is conducted from the eardrum to the inner ear.

Mild hearing loss: Occurs when a person is unable to detect sounds until the sounds are in the loudness range of 26dB to 40dB.

Moderate hearing loss: Occurs when a person is unable to detect sounds until the sounds are in the loudness range of 41dB to 55dB.

Moderately severe hearing loss: Occurs when a person is unable to detect sounds until the sounds are in the loudness range of 56dB to 70dB.

Otoacoustic emissions (OAEs): Low level sound emitted by the cochlea, evoked by an auditory stimulus or echo; related to the functioning of normal outer hair cells of the cochlea.

Otoacoustic emissions (OAEs) test: A test in which a sensitive microphone is placed in the ear while the audiologist presents several soft clicks or tones. If the inner ear (cochlea) is normal, it sends back sounds (called otoacoustic emissions) that are picked up by the microphone. When these responses are present it usually means a person's hearing is normal. If these responses are absent, it may indicate hearing loss. Responses may also be absent due to such things as wax in the ear canal or the presence of fluid in the middle ear.

Outer ear: The visible part of the ear, as well as the ear canal that channels sound from outside through to the eardrum.

Profound hearing loss: Occurs when a person is unable to detect sounds until the sounds are at a loudness level of 90dB or higher.

Screening: The application of rapid and simple tests to a large population consisting of individuals who are undiagnosed and typically asymptomatic, in order to identify those who require additional diagnostic procedures; screening typically results in either a 'pass' or a 'refer' outcome.

Sensitivity: The ability of a test to detect the disorder it was designed to detect; expressed as the percentage of positive screen results in those having the condition.

Sensorineural hearing loss: Hearing loss that results from a problem in the cochlea (inner ear).

Severe hearing loss: Occurs when a person is unable to detect sounds until the sounds are in the loudness range of 71dB to 90dB.

Specificity: The ability of a test to differentiate a normal condition from the disorder the test was designed to detect; expressed as the percentage of negative screen results in patients not having the disorder.

Tympanogram: A graph or chart that records the results of tympanometry testing.

Tympanometry testing: A hearing test during which a small probe is placed in the ear while the movement of the eardrum is measured to determine whether a problem exists in the middle ear.

Unilateral hearing loss: Hearing loss in one ear only.

Universal: Available and applicable to all, without discrimination.

Visual reinforced audiometry (VRA): A hearing test typically used for infants over 6 months of age, and toddlers up to about 2 or 3 years of age. VRA involves teaching a child to turn toward sounds, using toys that light up as the reward. Sounds are presented through headphones and/or speakers, with the goal of identifying the softest sound to which the child will respond, for different kinds of sounds.

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SECTION ONE: EFFECTIVENESS AND SAFETY OF PRESCHOOL HEARING SCREENING

Ken Bond, MA; Dagmara Chojecki, MLIS

INTRODUCTION

This health technology assessment report has been produced in response to a request from Alberta Health and Wellness (AHW) as part of the Alberta Health Technologies Decision Process (AHTDP) to perform an evaluation of the scientific evidence on the safety, performance, and effectiveness of universal and targeted preschool hearing screening (PHS). The Community and Public Health (CPH) Division of AHW has been developing the Infant and Preschool Screening Framework, of which hearing, vision, and developmental screening are possible components. A phased approach is being proposed, in which universal newborn hearing screening (UNHS) may be the first service implemented. CPH has asked the AHW Health Technologies and Services Policy Unit (HTSPU) to aid in the development of this framework by providing an evidence review of preschool hearing screening, both where UNHS is already in place and where it is absent.

OBJECTIVE AND SCOPE

This report is a structured review of the published research literature concerning the performance and effectiveness of hearing screening programs for preschool-aged children. **Screen performance** refers to the number of children who actually receive the screen (also called 'uptake') and to the number of cases identified (also called 'yield'). **Screen effectiveness** refers to the effect of screening on language skills, health-related quality of life, communications skills, social interaction, and educational performance.

The specific aim of this review was to answer the following questions, which were developed a priori:

1. What is the safety, performance, and effectiveness of **universal preschool hearing screening** under the following two scenarios:
    a) UNHS is already in place
    b) UNHS is not in place
2. What is the safety, performance, and effectiveness of targeted preschool hearing screening under the following two scenarios:
    a) UNHS is already in place
    b) UNHS is not in place

BACKGROUND

Hearing screening is a pass–refer procedure for identifying individuals who require further audiologic evaluation or other assessments. Universal screening for congenital hearing loss (HL) in newborns and infants 3 months of age and younger, and treatment of identified hearing loss by age 6 months, are now cornerstones of optimal neonatal care in Canada and the United States.[5–9] Lack of identification and lack of appropriate management of hearing loss in children can have a negative

impact on individual children's educational, cognitive, and social development; this has broad economic effects. To avoid these negative impacts, the goal of early detection of newborn hearing loss is to maximize perception of speech and the resulting attainment of linguistic based skills.[8] The identification of new or emerging hearing loss in one or both ears, followed by appropriate referral for diagnosis and treatment, are first steps toward minimizing these effects.[8] Despite the importance of UNHS programs in identifying congenital hearing losses, these programs will not identify infants with progressive, late-onset, and acquired hearing loss. Data from the Centers for Disease Control and Prevention shows that among the 2% of infants referred for follow-up after UNHS, fewer than 40% received a diagnosis. Though parental suspicion of hearing loss based on a child's inattention or erratic response can be a useful predictor,[10] relying on such concern can delay identification and appropriate treatment. Hence, in addition to using UNHS, it may be appropriate to employ hearing screening of some kind to identify those preschool children most likely to have peripheral hearing impairment that may interfere with communication, development, health, or future academic performance.[9,11]

Condition

Hearing is measured in terms of the lowest level in decibels (dB) at which a tone can be heard 50% of the time compared with average normal hearing in young adults (that is, 0 dB). Hearing loss is the reduced ability to detect and hear sounds; it is defined as unilateral or bilateral sensorineural and/or conductive hearing loss greater than 20 dB HL in the frequency region from 1000 through 4000 Hz.

- Mild hearing loss: People who cannot, with their better ear, detect sounds until they are in the loudness range between 20 and 40 dB, have some difficulty keeping up with conversations, especially in noisy surroundings. Children with this mild loss may have problems hearing in the classroom, learning, acquiring literacy skills, and playing with peers; they may need a hearing aid.
- Moderate to moderately severe hearing loss: Those who cannot, with their better ear, detect sounds until they are in the loudness range between 40 and 70 dB, have difficulty keeping up with conversations when not using a hearing aid.
- Severe to profound hearing loss: Those who cannot, with their better ear, detect sounds until they are in the loudness range between 70 and 95 dB, benefit from powerful hearing aids and possibly cochlear implants; they may rely on lip reading, even when using hearing aids, and some may use sign language.[10]

Although hearing loss can be a progressive congenital impairment that manifests in later childhood, it can also be acquired. Otitis media (OM), the inflammation of the middle ear associated with a buildup of fluid, is the most frequently diagnosed disease in young infants and children. Fluctuating conductive hearing loss nearly always occurs with all types of OM and it is the most common cause of hearing loss in young children. This type of hearing loss is generally temporary; however, when OM occurs repeatedly, damage can occur to the eardrum or even to the hearing nerve, and may cause permanent, sensorineural hearing loss.

Given the diverse nature of conditions that can lead to hearing loss, within the context of an integrated UNHS program, preschool hearing screening targets four independent groups of children.

1. Children who have no impairment at birth but who acquire impairment later in life as a result of some traumatic event such as infection (for example, bacterial meningitis), head

injury, ototoxic therapy (medications taken for a condition, such as infection or cancer, that result in damage to the inner ear, thus impairing hearing or balance), or chemotherapy.

2. Children who have a hearing impairment at birth, but of insufficient severity to be detected by newborn screening. As the child grows, the initially mild condition becomes progressively worse (progressive impairment).
3. Children who develop genuine, late-onset impairment with no obvious causative factor.
4. Children who might have been identified at birth had they undergone newborn hearing screening, but who, for some reason (for example, because of early discharge or parental refusal), did not undergo such screening.

Prevalence

Because of the implementation of UNHS, and because of regional variation in later screening for hearing loss, it is difficult to establish the prevalence and incidence of late-onset and acquired hearing loss. The proportion of children who, at age 5, have hearing impairments that actually are congenital is not well understood, and reported ranges vary.[11] Some researchers[11] have suggested that 5% to 10% of newborns manifest one of the risk indicators for progressive or late onset hearing loss. The category of children who have temporary hearing loss is composed primarily of those having persistent middle ear effusion and sensorineural deficits. The prevalence of temporary problems is approximately 15%, while for persistent problems it is closer to 3%.[11] In the United States, the Early Childhood Hearing Outreach has used otoacoustic emissions (OAE) to help identify hearing loss in children (aged 4 to 5 years) in Head Start programs. Data from these programs indicates that approximately two out of 1000 children are identified with hearing loss and an additional 18 out of 1000 are identified with transient conductive hearing loss.[12]

Screening Tests

A variety of objective tools have been developed for screening tests, and the choice of tool to use in screening depends on available resources and on the child's age and degree of cooperation. However, the following tests are generally used for screening preschool children:[10]

- pure-tone audiometry (conventional audiometry, play audiometry, and visual reinforcement audiometry)
- tympanometry
- otoacoustic emission (transient-evoked or distortion-product)

Pure-tone audiometry (PTA): PTA is the reference standard for the screening and evaluation of hearing loss in infants older than 6 months of age and includes various behavioural tests that rely on operant conditioning.[13] PTA measures auditory thresholds in response to speech and frequency-specific stimuli presented through earphones. Each ear is usually tested at frequencies of 500, 1000, 2000, and 4000 Hz.[10] Depending on the age of the child, PTA can be modified to suit the developmental level of the child by using conventional pure-tone or play audiometry (visual reinforced audiometry is also used to assess hearing, but is not readily applied in screening programs).[10] The appropriateness of any of the PTA tests is dependent on the attention span, understanding, and cooperation of the child. For children aged 4 years and older, conventional screening audiometry can be used. In conventional PTA, the child is asked to raise his/her hand or press a button when a sound is heard. The test should be performed in a quiet environment using earphones. Children between 2 and 4 years of age can be screened using play audiometry, in which the child is conditioned to respond to an auditory stimulus through a play activity such as dropping a

block in a box or putting a peg in a board—any task that is easy and fun for the child to do and also easy for the examiner to judge.[14] The skill and experience required for accurate and consistent play audiometry is considered substantial.[11]

Tympanometry: Tympanometry measures relative changes in tympanic membrane movement as air pressure is varied in the external auditory canal. It is used to assess middle ear status; it is not a test of hearing.[10] Tympanograms (the results of tympanometry) are classified depending on the curve shape relative to 0 as the pressure is changed.[10] A flat tympanogram indicates a high probability that middle ear effusion or a perforated tympanic membrane are the cause of hearing loss; a high-peaked tympanogram indicates a low likelihood that effusion is the cause, and a peak shifted toward negative pressure indicates a low probability of middle ear effusion and associated hearing loss. It can be challenging to conduct successful pure-tone screening with young children or those having special needs; thus, alternatives to pure-tone screening are required for these populations.[10]

Evoked Otoacoustic Emissions (OAE): OAEs are acoustic signals generated from within the cochlea that travel in a reverse direction through the middle ear space and tympanic membrane, out to the ear canal. These signals are generated in response to auditory stimuli (either clicks or tone bursts). The signals may be detected with a very sensitive microphone/probe system. The test allows for individual ear assessment, can be performed quickly on a child of any age, and does not depend on whether the child is asleep or awake. OAE can be used to screen for middle ear abnormalities and cochlear causes of hearing loss. An automated OAE screener provides a pass–fail report.[10] The OAE test does not further quantify hearing loss or hearing threshold level, nor does it distinguish conductive from sensorineural heaing impairment.

OAE assessments have two main variants: transient-evoked (TEOAE) and distortion-product (DPOAE). TEOAE presence implies integrity of sound transmission through the outer and middle ear structures and functional integrity of the outer hair cells, which are the primary sensory transducers with the organ of Corti in the cochlea.[11] DPOAEs are an alternative form of cochlear emission, also having their origin in the outer hair cells of the cochlea. In contrast to TEOAE assessments, DPOAE assessments use two simultaneous, sustained pure tones, typically in the 50 to 70 dB instensity range with a frequency ratio of 1.22. The frequency-specific nature of the DPOAE may provide more precise information than that provided by the TEOAE, but poor recording conditions may affect the accuracy of measurement. The latest devices incorporate both measurement of OAE and otoacoustic immittance capabilities.[11] This allows simultaneous detection of any hearing impairment of at least 30 dB and limited differential diagnosis of the type of impairment (conductive or sensorineural).

Diagnostic audiology is used to confirm or deny the suspected existence of hearing loss based on a hearing screen. If a loss is present, one of the aims of diagnostic testing is to determine the general degree of hearing loss as well as the degree of impairment as a function of frequency or pitch.[14] Another aim of testing is to determine the type of loss or the location of the problem within the auditory system. Both kinds of information are necessary in order to develop appropriate management strategies.[14] Trained audiologists conduct diagnostic testing for children who do not pass hearing screening. Audiologists may repeat, in a sound booth, the test used in screening, and may also use a variety of other tests. A complete examination usually takes 45 minutes to one hour, depending on the age of the child.[15]

Guidelines and Recommendations

Alberta College of Speech-Language Pathologists and Audiologists (ACSLPA) Hearing Screening Guidelines (2008):[5] ACSLPA guidelines provide direction on the hearing screening procedure in general and do not provide specific recommendations for preschool infant screening. The guidelines indicate that any child, preschool (age 3) and older, may be screened, and recommend that any person who is difficult to test should be referred for complete audiological assessment. The guideline provides specific direction on taking a case history, conducting a visual inspection, using pure-tone screening (including pass/fail criteria), and checking and maintaining equipment. At the screening level, a child's failure to respond to two or three tones, presented at any frequency in either ear, constitutes a screening failure. Any child who fails the screening should be referred to an audiologist at a health centre close to the family.

Ontario Infant Hearing Program (OIHP) Audiological Assessment Protocol (2008):[7] The OIHP protocol provides detailed guidelines on assessment goals, overall screening procedures, data collection and documentation, personnel, equipment, and screening environment for the detection of hearing impairment in children younger than age 6, but provides no information on screening programs specifically for preschool-aged children. All assessments funded by the OIHP must be conducted by audiologists registered with the College of Audiologists and Speech-Language Pathologists of Ontario who are authorized by the OIHP as having received training in the assessment protocol.

British Columbia Early Hearing Program (BCEHP) Diagnostic Audiology Protocol (2008):[6] The BCEHP protocol builds on the work of the OIHP and provides detailed guidelines on assessment goals, overall screening procedures, data collection and documentation, personnel, equipment, and screening environment for the detection of hearing impairment in children younger than age 6, but provides no information on screening programs specifically for preschool-aged children. The BCEHP was developed and is delivered by healthcare professionals in BC's six health authorities. Assessment funding for the program is administered locally by 12 regional BCEHP coordinating agencies, according to locally negotiated contracts within provincial funding guidelines. All assessments funded by the BCEHP must be conducted by a BCEHP-registered audiologist who has received training in the assessment protocol.

American Academy of Audiology (AAA):[8] The AAA's minimum practice guidelines for mass hearing screening in school settings recommends screening preschool and kindergarten children to help maximize the identification of new and emerging hearing loss. AAA guidelines provide detailed recommendations on appropriate ages for screening, the type of screening test to be used, the screening environment, personnel and equipment, and quality assurance measures for hearing screening programs. Pure-tone screening should be used to screen populations aged 3 and older. Both TEOAEs and DPOAEs should be used in those populations in which pure-tone audiometry cannot be used, but these tests cannot replace the preferred battery of pure-tone screening and tympanometry. Tympanometry should be used in conjunction with pure-tone screening for the preschool population and should be used as a second-stage screen following failure of pure-tone or OAE screening to help differentiate children with active middle ear effusion and hearing loss from those with possible sensorineural hearing loss.[8]

Because preschool children are at higher risk for hearing screening failure secondary to middle ear effusion, the AAA recommends that school districts include tympanometry at least for children in toddler, preschool, kindergarten, and grade 1. Following failure of PTA and tympanometry screening, the rescreening period will be, at minimum, eight weeks after the initial screening and no

later than 10 weeks afterward. The AAA also recommends against using acoustic reflex or acoustic reflectometry screening in mass preschool hearing screening programs. All screening should take place in an acoustically appropriate environment (as measured by a sound meter or calibrated to a normal-hearing adult). Screening programs using OAE technology must involve an experienced audiologist. Tympanometry equipment should be calibrated daily. To maximize the number of newly identified or emerging hearing losses identified, a lack of response in either ear at any frequency should constitute a 'fail.' A child who receives a 'fail' should be rescreened immediately, preferably by a different tester and with a different audiometer. With respect to program management, the AAA states that responsibilities for a hearing screening program must target accountability, risk management, and program evaluation. Program management responsibilities include implementing a protocol that ensures patient confidentiality, parental notification/permission, appropriate referral, and counseling. Risk factor management includes potential for infection, invalid screening results based on calibration errors or malfunctioning equipment, and an annual review for quality assurance. With respect to evaluation, mechanisms must be in place to allow evaluators to accurately quantify the pass and refer rates, estimate the false-positive and false-negative rates, and assure the effectiveness of follow-up protocols for patients who need testing or are referred from screening. The AAA guideline recommends what types of information ought to be collected to adequately evaluate a program's effectiveness.

American Speech-Language Hearing Association (ASHA) Recommendations:[9] The ASHA guidelines provide separate consideration and recommendations for children aged 7 months to 2 years, 2 to 5 years, and 6 years and older. These guidelines state that any child who has not been screened by 6 months of age should undergo hearing screening; however, they make no other statement about the use of universal preschool hearing screening. Children referred from screening should have their hearing status confirmed within one month and no later than three months from the time of the initial screening. For children functioning at 7 months to 3 years developmental age, visual reinforcement audiometry (VRA) and conditioned play audiometry (CPA) are considered the two most appropriate tools. OAEs could be used for children in this age category who are unable to participate in behavioural procedures. ASHA has separate guidelines for preschool children (aged 3 to 5 years) because it is believed that the testing procedure used for this age (CPA) requires more training, instruction, and caution on the part of the examiner than do traditional screening procedures used with older children. Because many of the hearing impairments in preschool children are associated with middle ear disease, ASHA recommends that children in this age group also be screened for outer and middle ear disorders, with the aim of identifying those children at risk of developing hearing impairment or a medical condition that warrants attention. The guidelines recommend that a child be referred for audiological assessment if the child does not respond at least two times out of three, at the criterion decibel level at any frequency in either ear, or if the child cannot be conditioned to the task. For children aged 7 months to 2 years, the guidelines recommend that personnel conducting screening be limited to ASHA-certified, state-licensed audiologists; for children aged 3 to 5 years, screening personnel should be limited to ASHA-certified and state licensed audiologists or speech-language pathologists, and support personnel under the supervision of a certified audiologists. ASHA emphasizes that, because preschool populations are generally not available in large, organized groups that lend themselves to universal screening, an interdisciplinary, collaborative effort is particularly important for this age group. Furthermore, physicians and other professionals who specialize in child development should be included in the planning and implementation of the hearing screening programs to maximize the likelihood of prompt care of children referred from screening.

For all age groups, the hearing screening program should include an educational component designed to provide parents with information, in lay language, on the process of hearing screening, the likelihood of their child having a hearing impairment, and follow-up procedures.

Canadian Association of Speech Language Pathologists and Audiologists (CASLPA): No relevant practice guidelines were identified from the Canadian Association of Speech Language Pathologists and Audiologists (CASLPA).

Project context

In March 2012, Charis Management Consulting Inc. completed a comprehensive current state assessment[16] that detailed the funding and human resources, physical infrastructure, data management and information sharing systems, and quality management processes and protocols for infant and preschool screening in Alberta. The following information summarizes the information described in the Charis assessment.

Currently, only the North and South zones maintain UNHS programs. Calgary and Edmonton maintain targeted screening programs for infants identified as being at at high risk. The UNHS programs currently capture 85% of all newborns residing in the catchment areas of the respective zones. With respect to PSH screening, the Charis assessment found that the only coherent and standardized PHS services that exist in Alberta are organized at the health-zone level or lower. The only universal hearing screening programs for preschool/kindergarten children in the province are based out of the smaller population centres of Medicine Hat and Grande Prairie (Table T.1). In the spring of 2012, AHS was expected to roll out a universal kindergarten hearing screening program in the greater Grande Prairie and Peace River region to screen all children aged 5 and 6, in as many as 60 schools.[17] The aim of the program is to identify children having any possible hearing loss that may affect their development and overall general health.

Selective hearing screening programs are more common (in place in all zones), but do not provide consistent screening services to the same target populations. Selective screening programs are offered by different agencies (for example, public health, speech–language pathology, Head Start) and program areas in different parts of the province.

Table T.1: Universal preschool hearing screening resources and tests

Location	Resources	Tools	Assessment and diagnosis	Coverage of target population (%)
Grande Prairie	Audiologist and technician	Euroscan and otoacoustic emission	In zone	70
Medicine Hat	Speech language pathologist and audiology/speech assistant	Audiometer	In zone	Not available

Provision of Audiology Services in Alberta

The level of screening, the processes and tools used, and the staff doing the screening all vary from program to program. The two preschool hearing screening services (Medicine Hat and Grande Prairie) are run by audiology services within Allied Health and follow children whose parents have given written consent for an in-school screen. PHS is usually conducted in shared space located in schools or in clinic space in public health or audiology offices. Most programs use audiometers or

otoacoustic emissions; however, tests such as Eroscans, tympanometers, and otoscopes are used as well.

The level of detail and type of information collected by preschool hearing screening programs varies across AHS zones. Preschool hearing screening data is collected and managed in a variety of ways across the province. Program data and staff time are recorded in Meditech by two zones (North and Southeast), but no data is being rolled up into provincial data systems or attached to electronic medical records. Information is shared within the scope of the *Health Information Act* and AHS privacy policies, but no formal mechanisms are in place for sharing information outside of AHS. The requirement of parental consent for the sharing of information poses a particular challenge for school-based preschool hearing screening programs, as consent forms must be sent out in advance and typically have a relatively low return rate.

Professional oversight and quality assurance and clerical support vary among programs. The current-state report found limited information on processes and protocols to ensure quality, accuracy, and interpretation of preschool hearing screening results. For those zones that were able to provide information, audiology staff usually provided 'on-the-job' training as well as the oversight and supervision for screening.

Current Resource Allocations

Staffing resources allocated to PHS vary across the province. In most circumstances, no dedicated personnel are hired for preschool hearing; instead screening is integrated into professionals' and paraprofessionals' roles in overall assessment and diagnosis. As a result, it is difficult to estimate the number of staff currently involved in screening activities across the province. Estimates from targeted PHS programs suggest that the time spent administering an audiometer screen averages around 15 minutes per child, or greater if concerns arise. Physical space in which to conduct PHS is usually shared space located in schools or in clinic space in public health or audiology offices.

Funding in Alberta

None of the PHS programs has dedicated PHS funding; any funding resources allocated to these activities are integrated within a broader (audiology or other) program area.

Key Challenges and System Supports

Based on interviews with key stakeholders across the province, the Charis assessment identified that the key challenges in establishing PHS programs in Alberta include:

- under-resourcing for screening as well as follow-up diagnostic assessment and intervention
- lack of a standardized data management and reporting system
- finding appropriate environments in which to screen
- ensuring parental consent forms are returned so that staff are authorized to screen as many children as possible
- sharing of screening outcomes data

In addition, the Charis report identified that the components of a quality PHS program include:

- a coordinated and integrated system, with a well-defined and supported organizational structure

- a provincial data management system, with standardized screening and diagnostic protocols and outcome measures

Methods

A detailed description of the review methodology, including the search strategy, is provided in Appendix A. Briefly, the methodology was as follows.

Literature search

Electronic searches of the peer-reviewed scientific literature published from 2002 to April 2012 were conducted in the following databases: MEDLINE (including in-process), EMBASE, and CINAHL. In addition, reference lists of reviews and retrieved articles were searched for relevant studies. A Google search was also conducted for relevant information. Searches were limited to English language articles.

Literature selection

One reviewer (KB) screened titles and abstracts, retrieved relevant articles, and determined eligibility of key studies according to the inclusion criteria below (see Table T.2). Studies were excluded if they did not meet all inclusion criteria.

Table T.2: Study selection criteria

Publication	Complete report of study published in English
Study design*	Health technology assessments, systematic reviews, meta-analyses, randomized or non-randomized studies, or cross-sectional screening performance and effectiveness studies
Population	Preschool-aged children (≤6 years)†
Condition	Hearing loss (congenital, permanent progressive, delayed-onset, or acquired)
Screen	Universal or targeted hearing screening program (no restriction on technologies used)‡
Reference standard	Audiologic assessment
Comparator	No preschool hearing screening program
Outcome	Any measures related to safety, screening accuracy, therapeutic efficacy, or functional outcomes (e.g., improved speech and language ability, improved educational outcomes, reduced need for remediation)

*Due to time constraints, HTAs and systematic reviews were sought first. When no relevant research syntheses were identified, the primary literature was examined.
†Based on the age for compulsory schooling in Alberta as defined by the *Alberta School Act*
‡Conducted in a country with a developed market economy as defined by the United Nations (that is, Australia, Canada, Germany, Italy, Japan, New Zealand, United Kingdom, and United States)

Assessment of Methodological Quality

Due to time constraints, quality assessment was not conducted.

Evidence Summary

Based on the included studies, a tabular and narrative summary of the study characteristics and results was developed. In addition, references are provided for included and excluded studies (see Appendix C).

RESULTS

Summary of Evidence

Quantity of research available

The search of electronic databases and the Google search identified two rapid reviews, two systematic reviews, and three primary studies addressing various aspects of the research questions. With respect to universal screening programs, two CADTH rapid response reports examined, respectively, evidence on the appropriate age at which to conduct universal hearing screening following a UNHS program[18] and evidence on the comparative diagnostic accuracy of government versus private hearing screening programs.[19] One NHS HTA[1,20] assessed whether a school-entry screening program in the United Kingdom made a useful contribution to the identification of childhood hearing loss in light of recent UNHS implementation. One prospective cohort study[21] assessed screening and follow-up outcomes of the Long Island Hearing Screening Program (a universal preschool hearing screening program) over a 10-year period. One systematic review[4,22,23]addressed the question of whether long-term outcomes differed between those children in the general population who were screened for OME and those who were not screened for OME within the first four years of life. In terms of targeted screening, two prospective cohort studies[2,3]assessed the contribution to the identification of hearing loss in preschool-aged children in traditionally medically underserved areas of the United States that had UNHS programs.

No studies were identified that assessed universal or targeted preschool hearing screening in the absence of a UNHS program.

Summary of study characteristics

Characteristics of the included studies are summarized in Table T.3.

Universal Hearing Screening for Hearing Loss (HL)

CADTH 2011[18] conducted a limited search of electronic bibliographic databases and of the Internet (English-language reports published between 2004 and 2009) to identify any scientific evidence on the comparative diagnostic accuracy of government-funded versus private hearing screening programs. No literature addressing the topic was identified.

CADTH 2009[19] conducted a limited search of electronic bibliographic databases and of the Internet (English-language reports published between 2006 and 2011) to identify any HTAs, systematic reviews, or evidence-based guidelines that examined the appropriate age at which to conduct universal hearing screening following UNHS. The search identified one HTA on school-entry hearing screening.[1] As that was the only HTA addressing this topic and it was identified via this review as well, the characteristics and results of the HTA are summarized below.

Bamford et al.[1,20] conducted an HTA for the NHS HTA program to better understand the practice and contribution of universal school entry screening for detecting children with hearing loss in light of the implementation of a UNHS program.

The HTA consists of three sections:

- a national questionnaire survey of current school-entry hearing screening practice in the United Kingdom
- a systematic review of the literature on the accuracy and effectiveness of hearing screening tests
- a cost-effectiveness analysis of school-entry hearing screening

The systematic review sought evidence on the accuracy of school-based hearing screening (children aged 4 to 6, assessed in school settings), the yield and uptake of school-based hearing screening, the effectiveness of school-based screening on language, education, and social outcomes, and any adverse events of school-based hearing screening. The review authors conducted a comprehensive search of electronic bibliographic databases and trial registries from inception to May 2005. Studies with no clear comparator were excluded and no restrictions were placed on the tests used. No meta-analysis was conducted because of the small number of studies on screen performance and effectiveness.

The review authors identified three previous systematic reviews and 25 primary studies that addressed the review questions. Of the three previous systematic reviews, two examined the accuracy of tests on children whose age (up to age 12) went well beyond the scope of this review and, therefore, are not described here. The remaining review examined the relative effectiveness of selective versus routine school-entry medical examination (vision assessment, hearing test, and general medical examination by a doctor). The review included 16 primary studies but the summary data did not distinguish between vision, hearing, growth, or other physical problems, so it was not possible to draw any conclusions about the effectiveness of school-entry medical examination for the identification of hearing loss.

In addition to the reviews, the authors identified 25 primary studies; however, only four of the included studies included exclusively children between 4 and 6 years of age, and four studies did not describe the age of the children but described the study population as 'kindergarten' or 'preschool.' The ages of the children in the remaining studies varied from as young as 2.5 to as old as 14.

A range of different test comparisons was found, with some studies comparing individual tests (for example, tympanometry versus PTA), and others comparing combinations of tests or different protocols for the same test. The majority of studies used PTA as the reference test. When reported, screening was carried out in a variety of settings, including within the school or primary care/community facility, and under tightly controlled conditions. In the majority of studies a qualified professional, such as a school (or public health) nurse or an audiologist, conducted the screen. Studies failed to identify the conditions (for example, hearing loss, conductive impairment) being sought, or the severity of hearing loss identified. Lastly, although the review authors summarized the information on test accuracy and screen performance, they noted that the studies providing these data were experimental test accuracy assessments rather than 'real world' community based screening evaluations (that is, screening programs), and that none of the studies investigated reported their true case yields. For these reasons, test accuracy and screen performance data from the review was not considered relevant to the current assessment of PHS programs.

With respect to screen effectiveness, the review authors identified one study examining the effectiveness of preschool hearing screening. This retrospective cohort study compared two groups of 730 children from different geographical areas in Ontario. One group received verbal audiometric hearing screening by a public health nurse before school entry while the other group did not.

Hearing loss was assessed by a PTA in both groups at 6 to 12 months after screening. The study found no statistically significant difference in the prevalence of hearing impairment in the two groups after 6 to 12 months. However, the review authors note several potential sources of bias in the design and conduct of the study that may explain the observed results, namely, selection bias, lack of blinding to screening status, non-compliance with treatment, a wrong timescale, and so on.

No studies were identified that assessed the long-term impact of preschool hearing screening on educational, language, and social outcomes. No studies reported any adverse effects of screening.

In addition to conducting the systematic review, HTA researchers conducted a postal questionnaire survey to determine whether the school-entry screening program continued to make a useful contribution toward identification of hearing impairment in light of UNHS implementation. Questionnaires were sent to 229 screening service leads; 195 (85%) responded. All respondents reported screening was done on school premises and took place in a quiet classroom all or most of the time. Most services used pure-tone sweep audiometry as the first test and 71.7% (n = 124) implemented a two-test screen before referring a child to diagnostic services. The time between the two tests ranged from a few hours to more than 12 weeks and the criteria for retesting and referral varied greatly. In most cases, school health nurses or school health nurse assistants conducted the screening. Coverage of the population was considered high, with 75% of services achieving greater than 90% coverage. The median referral rate was 7.9% (range: 1.91% to 23.4%). No data were available on false negative results (that is, children who were not referred but were subsequently identified as having hearing impairment). The median yield from 18 sites reporting the number of cases of hearing impairment was 0.12% (range: 0.05% to 0.59%) for sensorineural impairment and 0.09% (range: 0.07 to 0.44) for permanent conductive hearing impairment. The median positive predictive value from the number of children referred was, respectively, 1.71% (range: 0.62% to 12.16%) for sensorineural impairment and 3.42% (range: 1.24% to 17.56%) for permanent conductive impairment.

The authors noted that the UNHS program would not have captured many of the children screened at school entry, because newborn screening was only fully implemented in 2006, or even later, in some parts of the United Kingdom. Hence, the authors concluded that no evidence was available to support a decision to either continue or discontinue the school-entry hearing screen. It was emphasized that a proper evaluation of the school-entry hearing screening program requires: national guidance and a single national protocol for the implementation and conduct of school-entry hearing screening; a coordinated system of data collection; and the development of an evidence base showing the comparative effectiveness of alternative approaches for the identification of permanent hearing impairment in this age group.

Serpanos & Jarmel[21] conducted a 10-year retrospective assessment of quantitative follow-up data obtained from the Long Island Hearing Screening Program (LIHSP), a universal preschool hearing screening program for children between 3 and 5 years of age. Hearing screening procedures were based on a modified version of American Speech-Language-Hearing Association (ASHA) guidelines. Screening was conducted in private, nonprofit or public preschools, day care centres, or Head Start programs, by graduate students in audiology or speech-language pathology, under the supervision of ASHA-certified audiologists. Each child underwent a pure-tone screen followed by a tympanometry screen. Play audiometry was used when a child could not be reliably screened using conventional pure-tone audiometry (that is, hand raising). When one or more frequencies were not perceived at a loudness level of 20 dB in either ear, the child was retested in the same test session by a different examiner. A referral for hearing evaluation was made when a pure tone was not perceived at at a loudness level of 20 dB at any one frequency in either ear, or when a child could not be tested

reliably following the screening retest. The audiology supervisor performed otoscopy for children with known tympanostomy tubes, reduced peak static acoustic admittance, or when testing could not be conducted due to lack of a seal. A child was referred for medical evaluation when there was visual observation of ear drainage. No children who passed the screen were evaluated using diagnostic audiological assessment.

A total of 34,979 children were screened between 1995 and 2005; 82% (n = 28, 642) of the children passed both the pure-tone and tympanometry screen. Demographic data was not reported for the overall screened population. The overall referral rate was 18% (n = 6337): 2% (n = 663) did not pass the pure-tone screen, 3% (n = 1185) could not be tested, 6% (n = 2006) could not pass the tympanometry, and 7% (n = 2483) did not pass either the pure-tone or tympanometry screens. Follow-up data was available for 21% (n = 1316) of those who completed the referral. The age distribution of the follow-up group was: age 3, 19.4%; age 4, 21.7%; age 5, 9.8%; age unspecified, 49.1%. The follow-up group consisted of 49.0% males and 46.1% females, gender was unspecified for 4.9%. Unilateral or bilateral hearing loss, ranging in degree from slight to profound (loudness level of >90 dB), was diagnosed in 18% (n = 239) of children (156 conductive, 15 sensorineural, 5 mixed, and 63 unspecified). Of the 682 children receiving medical assessment, 480 (70.4%) presented with outer or middle ear disorders. Otitis media (with or without effusion) accounted for 25% (n = 335) of otologic disorders identified in the follow-up group.

Universal Hearing Screening for Otitis Media with Effusion (OME)

Simpson et al.[4,22,23] conducted a systematic review of RCTs to address the question of whether long-term outcomes differed between children in the general population who were screened for OME and those who were not screened for OME within the first four years of life. The review was originally published in 2003, and was updated in 2006 and again in 2009. The review also examined the effect on outcomes of treatment of children identified with OME through screening (early detection and treatment). The authors conducted a comprehensive search of electronic bibliographic databases from inception to 2009. They also used reference tracking and a Google search to identify unpublished reports.

No randomized trials were identified that compared outcomes for children who were screened with those who were not.

The authors identified three trials (published in seven reports) that reported the effect of screening and treatment on children identified with clinically significant OME in the first four years of life; 668 participants were involved. The studies had two phases.

1) Children were screened for OME.
2) Those found to have clinically significant OME were invited to participate in a randomized trial of treatment.

All three trials included children during their first 4 years of life and one trial included subjects who were followed up when they were between 9 and 11 years of age. The studies were conducted in the Netherlands and the United States. The children in the studies from the Netherlands were either invited to take part in a screening program or were drawn from a population that was already part of a screening program. The studies in the United States were screened for the purpose of describing the natural history of OME and for identifying children suitable for inclusion in a trial of early versus later OME treatment. The main outcome in all three trials was language development, with secondary outcomes of effusion resolution and improved hearing. Because of important differences

in study settings, inclusion criteria, outcome measures, follow-up, and results, no meta-analysis was conducted.

One study screened children at 24 months of age, using tympanometry. The children were screened in their own homes every 3 months for nine consecutive sessions. If the tympanogram was positive for OME, otoscopy was done to exclude causes other than OME and then OME was confirmed with a second tympanogram performed by an ENT surgeon. Children with a diagnosis of OME were randomized to receive either ventilation tubes or no treatment, and were followed up for 6 months. Language was tested using a standard Reynell test (a test that measures language skills in young children, through assessment of verbal comprehension and expressive language). Of 1050 children who completed the last round of screening, 288 met the criteria for treatment. Of these 288 children, 84 (8% of those screened) received a positive diagnosis and were eligible for the trial; 52 children participated in the trial. The mean age of the children was 39.5 months for those in the treatment group and 39.2 months for the non-treatment group. When children in the treatment phase were analyzed as a single group, bilateral OME lasting 3 to 6 months caused significant impairment of expressive language skills, but its effect on verbal comprehension was not significant. However, there was no significant difference in language development (expression and comprehension) between those children who received treatment after a positive screen for OME and those who did not receive treatment after the same screen result.

The second study was embedded within a cohort study of 30,099 children. Children were invited for screening at 9 months of age, using the Ewing test (a behavioural response hearing test for babies and young infants that uses familiar, complex sounds rather than pure tones). Children who failed a hearing test were recalled 1 month later. Those who failed three successive tests were referred to an ENT clinic for diagnostic testing. Children with bilateral OME confirmed (via tympanometry and otoscopy) were randomized to receive either ventilation tubes or watchful waiting. Children in the watchful waiting group who needed treatment received management other than ventilation tubes, (for example, antibiotics). Children were followed for 12 months and tympanometry and otoscopy were performed every 3 months. The mean age was 19.5 months for the treatment group and 19.4 months for the watchful waiting group. Hearing loss and expressive language and comprehension were assessed every six months. Language was assessed using the Reynell test, the Schlichting test, and the Lexi test. Of the 30,099 children screened, 1081 (3.6%) failed three successive tests and were referred for diagnostic assessment. Of those referred, 386 (1.3% of those screened) were identified (at 4 to 6 months of age) with persistent OME; 158 were randomized and assessed at 12 months. In follow-up at 3, 6, 9, and 12 months of age, 15%, 29%, 27%, and 27%, respectively, of children in the ventilation group were diagnosed with bilateral OME. In the watchful waiting group, these percentages were 77%, 66%, 57%, and 52%. The differences between the two groups in language comprehension was statistically non-significant ($p = 0.18$). The difference between the two groups in expressive language was also statistically non-significant ($p = 0.17$). The groups also did not differ significantly in terms of their performance on the Lexi test (words the children speak spontaneously; $p = 0.32$). The effect of screening and treatment with ventilation tubes on group average hearing levels was evident at 6-month follow-up, but had all but disappeared by 1-year follow-up.

The third study invited 6350 children for screening to describe the natural history of OME. Healthy infants aged 2 to 61 days were evaluated for OME, monthly for 3 years, using tympanometry and otoscopy. Children were eligible to be enrolled the trial if, beginning at the age of 2 months and within the first 3 years of life, they had middle ear effusions that appeared substantial and that persisted, despite treatment with antimicrobial drugs, for 90 days (bilateral effusion) or 135 days (unilateral effusion). Children were assigned to early treatment (ventilation tubes as soon as possible)

or late treatment (the same procedure 6 months later, if unilateral effusion persisted). The mean age of children at randomization was 15 months (median 14 months). Developmental testing of the children in the trial was undertaken using a variety of tests (for example, picture vocabulary test, mean length of utterances, Child Behaviour Checklist, Woodcock Reading Mastery Test, Wechsler Intelligence Scale for Children), after the child's third, fourth, and sixth birthdays, and again between their ninth and 12th birthdays. Of 6350 children screened, 588 (9.3%) were eligible for the trial; 429 were randomized. During the first 12 months, the percentage of children in the late treatment group who'd had effusion for more than 50% of the time was approximately three times that of the percentage of children the early treatment group. During the first 24 months, the percentage in the late treatment group was approximately twice the percentage of those in the early treatment group. At age 4, no significant differences were evident between children in the early and late treatment groups on parent-rated measures of parent–child stress and children's behaviour. At age 6, no significant differences were evident between early and late treatment groups except for a moderately higher score among children in the late-treatment group on the Nonword Repetition Task ($p = 0.05$). At ages 9 and 11, no significant differences were evident between the two groups on any measures. Based on the review data, the authors calculated that, in order to identify one child with OME considered eligible for treatment, the number of children needing to be screened was from between four and 78.

Overall, no evidence was found of a clinically important benefit in terms of language development and behaviour from screening a general population of children in the first four years of life for OME and treating those identified with OME. Nevertheless, Simpson et al. note that many parents describe definite and sometimes dramatic improvements in their children after OME treatment. The authors conclude that it is possible that the threshold for treatment was too low in the studies included in this review, and that beneficial effects might have been demonstrated if eligibility for treatment had been limited to children with more severe OME. However, such children may be symptomatic and more easily identified through routine care, rather than by general population screening.

Targeted Hearing Screening for Hearing Loss

Allen et al.[2]conducted a prospective 4-year assessment of pass and refer rates in a hearing screening program for 3- and 4-year-old children attending seven Head Start centres in rural eastern North Carolina, an area that is traditionally medically underserved. Head Start is a comprehensive, early childhood development program primarily serving at-risk preschool-aged children and their families (with the families having an earned income at or below the United States federal poverty level, which, for a family of four in 2011, was considered to be USD $22,350).[24] The hearing screening was part of a multidisciplinary health screening program (speech–language, vision, dental, fine and gross motor skills, blood pressure, and so on) that aimed to screen all children in the program regardless of whether or not they were receiving medical management. Hearing screening procedures were based on American Speech-Language-Hearing Association (ASHA) guidelines, and screening was conducted in quiet rooms at each of the Head Start centres. Children were screened using conditioned play audiometry, tympanometry, and otoscopy. A child was considered to have passed the audiometry screen if two of three reliable responses were obtained at a loudness level of 20dB for all three frequencies in both ears. Children who did not pass the audiological screen were rescreened within two to four weeks and, if the child did not pass the audiological screen, the child was referred for a diagnostic audiological assessment. For middle and outer ear assessments (otoscopy and tympanometry), a pass indicated a negative result for ear drainage, a previously undetected structural deficit, or ear canal abnormalities. Children who did not pass the initial

assessment received a medical referral. Thus, a 'pass' in screening meant that a child had passed all three screening components (pure-tone audiometry, otoscopy, and tympanometry). None of the children who passed the screen were evaluated using diagnostic audiological assessment.

Approximately 65% (n = 1462) of the total student body was tested (with no overlap or retesting of the same individuals across years). Eighty percent of the children screened were 4 years old with similar numbers of males and females. More than 75% of the children came from families receiving Medicaid or families that had no health insurance at all. Of the 1170 children who passed the audiologic screen and the 25 who received diagnostic audiology, six (0.5%) were confirmed with hearing loss (four conductive and two sensorineural). Of the 15 children seen for medical examination, 11 presented with abnormal findings (otitis media or impacted cerumen). Overall, follow-up assessment compliance was poor, with roughly only 25 (9%) of 272 children receiving the referred diagnostic audiologic assessment. In addition, the hearing status of approximately 18% (n = 263) of those children initially screened was never determined. The authors highlighted the low initial pass rate (53.8%) of audiological screening, noting that this finding was consistent across the four-year study period. It was unclear whether transient middle ear effusion or an inappropriate screening protocol resulted in a high initial refer rate. The authors concluded that the goal of efficiently identifying children who had a medical condition or hearing loss was not met due to a high over-referral rate and a low identification of hearing impairment.

Eiserman et al.[3] conducted a 36-month prospective assessment of hearing loss in infants and toddlers (≤3 years) attending 65 Early, Migrant, and American Indian Head Start centres in Kansas, Oregon, Utah, and Washington. The hearing screening was part of a mandatory annual health screening program and was conducted by lay screeners who had attended a 6-hour training session. Screening took place in a range of natural environments, including classroom play settings and homes. Hearing screening was conducted in accordance with standardized procedures (not further described). Children were screened first using visual inspection and, if they passed the visual screen, could undergo up to three OAE screenings over a 2- to 4-week period. OAE was sensitive to hearing loss to 25 dB. Children who received a 'refer' rating from the OAE unit were referred for evaluation by a health care provider and pediatric audiologist or other hearing specialist. In addition, screeners could provide a direct referral for any child who received a 'refer' on either the first or second OAE screen or who could not receive an accurate screen result. None of the children who passed the screen were evaluated using diagnostic audiological assessment.

In total, 4519 children were screened. The mean age of the children screened was 22 months (13 SD; range 0 to 48), with 52% (n = 2347) of those screened being male. Of the 4519 children screened, eight (0.2%) did not pass the initial visual screen. Of the 4511 who were screened using OAE, 24% (n = 1099) did not pass the initial screen. Of the 4519 children screened, 257 (6%) were referred for audiological follow-up; 159 (61.8%) of those referred completed diagnostic assessment. Of the 4519 children screened, seven (0.2%) were identified with permanent hearing loss (four sensorineural and three conductive), 83 (1.8%) with otitis media, and 15 (0.3%) with excessive ear wax or congestion. Of the seven children identified with permanent hearing loss, four had passed newborn screening, two had not been screened at birth, and one had not received follow-up services after referring from newborn screening. The diagnostic status of 38% (n = 98) of those referred was unknown because they exited the Head Start program before completion of diagnostic assessment. Based on the within-study prevalence, the author estimated that an additional four cases of permanent hearing loss may have been identified in this group. Using the study data, the authors calculated the positive predictive value of the screen to be 67.3%.

Table T.3: Summary of study characteristics

First author, publication year Country Study design	Screening guideline used Study period	Population characteristics	Screen Diagnostic test setting Personnel	Results
Universal Screening for Hearing Loss				
CADTH, 2011[18] Canada Rapid review	NA	No studies identified	NA	NA
CADTH, 2009[19] Canada Rapid review	NA	One HTA (Bamford et al., 2007[1]) identified	NA	NA
Bamford et al., 2007[1,20] UK HTA	ND	Three systematic reviews, 25 primary studies, published between 1970–2005 National survey of 195 school-entry screening services	*Systematic review* Various screening tests PTA (in most cases) Various settings including schools and community facilities Qualified health professionals such as physicians and audiologists *National survey* PTA (in most cases) Diagnostic test ND Quiet classroom Personnel not described	One retrospective cohort reported no significant difference in prevalence of hearing impairment, 12 mo. after screening, between those screened and those not screened Survey results Median referral rate 7.9% (range: 1.91–23.4) Median yield *Sensorineural HI:* 0.12% (range: 0.05–0.59) *Permanent conductive HI:* 0.09% (range: 0.07–0.44)
Serpanos et al., 2007[21] USA Retrospective cohort	Modified ASHA 1995–2004	No. screened: 34,979 Followed up: 1316 Age: 3 yrs –19.4% 4 yrs – 21.7% 5 yrs – 9.8% age unspecified – 49.1% Male: 49.0%, Female: 46.1% Unspecified: 4.9%	PTA, tympanometry Quiet rooms on site (day care, Head Start centre, etc.) Graduate students in audiology/speech language pathology and audiology; supervised by ASHA-certified audiologist	Referral rates *Overall:* 6, 337 (18%) *Follow-up:* 1,316/6,337 (21%) Yield *Hearing loss:* 239/34,979 (0.7%) *Medical disorder:* 682/34,979 (1.9%)

Universal Screening for OME				
Simpson et al., 2009[4,22,23] UK Systematic review	Guidelines ND Study periods ND	Three RCTs: 1. No. screened: 1050 No. followed up: 288 Age: 24 mo. 2. No. screened: 30,099 No. followed up: 386 Age: 9 mo. 3. No. screened: 6350 No. followed up: 588 Age: 2–61 days	1. Tympanometry Confirmation by ENT surgeon In home Personnel ND 2. Ewing test ENT clinic diagnostic testing Setting ND Personnel ND 3. Tympanometry and otoscopy Diagnostic test ND Setting ND Personnel ND	1. No statistically significant difference in language development at 6 mo. 2. No statistically significant difference in language development at 12 mo. 3. No statistically significant difference across a range of developmental outcomes, including language, reading, behaviour, and intelligence.
Targeted Screening				
Allen et al., 2004[2] USA Prospective cohort	ASHA 1998–2002	Children attending Head Start programs, coming from families with incomes below the poverty line No. screened: 1462 Age: 3 yrs – 20.0% 4 yrs – 80.0% Male: 52.1%	CPA, tympanometry, otoscopy Audiologic assessment Quiet rooms in Head Start centres ASHA-certified auidologists/graduate students in speech language pathology and audiology	Referral rates *Initial:* 675/1462 (46.2%) *At re-screen:* 227/550 (41.3%) Yield *Hearing loss:* 6/1195 (0.5%) *Hearing status undetermined:* 267/1195 (22.3%)
Eiserman et al., 2008[3] USA Prospective cohort	Study-specific protocol 36-month period	Children attending Head Start programs, coming from families with incomes below the poverty line No. screened: 4519 Age: Male:	OAE Audiologic assessment Classroom play areas and homes Lay screeners who had completed a 6-hr training session	Referral rates *Initial OAE:* 809/4511 (18%) *Second screen:* 295/1055 (28.0%) *Third screen:* 135/359 (38%) Total: 257/4511 (5.7%; 84 direct referrals from OAE screen 1 and 2) Yield *Hearing loss:* 7/4,519 (0.2%) PPV: 67.3%

ASHA = American Speech-Language-Hearing Association; CPA = conditioned play audiometry; HI = hearing impairment; NA = not applicable; ND = not described; OAE = otoacoustic emissions; OME = otitis media with effusion; PPV = positive predictive value; PTA = pure-tone audiometry; RCT = randomized controlled trial

DISCUSSION

Summary

This structured review of the research literature on PHS programs identified two rapid literature reviews, one HTA, and one retrospective cohort study that examined the performance and effectiveness of universal preschool screening for hearing loss. One systematic review examined the effectiveness of screening children in the general population to identify those requiring treatment for OME within the first 4 years of life. Two prospective cohort studies[2,3] assessed the contribution of targeted screening to the identification of hearing loss in preschool-aged children in Head Start programs in areas of the United States. All studies examined screening within the context of UNHS; no studies were identified that assessed universal or targeted preschool hearing screening in the absence of a UNHS program.

Performance of universal screening programs was assessed in a national survey in the United Kingdom conducted as part of an NHS HTA,[1] and in one retrospective cohort study,[1] while targeted screening was assessed in two prospective cohort studies.[2,3] Overall, referral rates were high, about 8% or more of those screened, and the yield (those with confirmed hearing loss) represented less than 1% of the population screened. The uncertainty regarding the prevalence of hearing loss in populations with UNHS programs and the differences among screening settings, ages of the study populations, screening tests, and protocols limits the conclusions that can be drawn from this evidence regarding the contribution of universal or targeted preschool hearing screening for identifying hearing loss.

One retrospective study[1] and one systematic review[4] containing three RCTs provided evidence on the potential effectiveness of preschool hearing screening on language and developmental outcomes. None of the studies indicated a statistically significant or clinical meaningful difference between those who were screened and those who were not. No studies reported outcomes concerning the safety of either universal or targeted screening.

Of note, only one study[21] examined the recommended screening approach for universal PHS. This 10-year study used a retrospective design and followed children aged 3 to 5 years; hence, little evidence is available regarding the potential uptake and yield of preschool screening programs for children younger than age 3. In addition, the study did not assess outcomes of effectiveness, such as improved educational outcomes. For the data that was available, the retrospective design and high loss-to-follow-up suggest a need for caution in drawing any uptake or yield conclusions from the study results.

Based on a similar lack of evidence, Feightner[25] concluded that the evidence spoke against including routine hearing assessment in the preschool-age periodic health examination. He argued that, while hearing screening carries little or no risk to the individual child, its benefit has not been demonstrated and screening detracts resources from other health maintenance maneuvers. Similarly, the lack of benefit in screening for OME found by Simpson et al.[4,22,23] reflects, in part, the uncertainty regarding the need to treat OME. A systematic review and meta-analysis[26] that examined the relationship between OME or OME-associated hearing loss in early childhood to children's later speech and language development found no to very small negative associations between OME and later speech and language development. The authors concluded that the study results suggest that just ignoring OME and associated hearing loss for a young child is a reasonable approach. Nevertheless, the authors note that in any particular case, the relative risk of not screening hearing

and missing a moderate degree of hearing loss caused by OME must be weighed against the advantages of giving the child the optimal language and learning environment. Similarly, a New Zealand HTA on screening programs for OME and conductive hearing loss in preschool and new-entrant children[27] concluded that, despite an association between OME and conductive hearing loss, few persistent cases exist; therefore, it was not possible to draw conclusions about the effectiveness of screening programs.

The referral rates found here are similar to those reported by other researchers. For example, referral rates from initial OAE screens for children aged 0 to 3 years have been reported to be as high as 25% to 30%, with re-screening referral rates as high as 30% to 40% (approximately 8% of those initially screened).[12] Abnormal hearing test results require clinically appropriate referral for appropriate diagnosis, counseling, and treatment, including otolaryngology, audiology, and speech-langauge pathology.[10] Such high referral rates have important implications for the resources required to ensure timely diagnosis and, if necessary, intervention. While a well-designed follow-up tracking procedure that ensures compliance to the screening recommendations is as important as the screening itself, the results of the cohort studies described in this review indicate that this remains one of the major challenges to audiology screening. This echoes the findings of the Charis current state assessment,[16]which indicated that AHS staff saw the need for a coordinated and integrated system with well-defined and supported organizational structure and a provincial data management system with standardized screening and diagnostic protocols and outcome measures. In addition, current guidelines emphasize the necessity of a coordinated approach to ensure appropriate and timely referral.

Information gaps

To establish the comparative effectiveness of PHS, one needs to know, or to be able to estimate, the number of cases that remain to be identified by the PHS after the identification of these cases by the UNHS. The uncertainty around this baseline is perhaps the greatest limitation of the research literature in this area. In addition, an effective screening protocol must balance the risk of false positive results and the potential over-referring of children for assessment against the need for timely referrals. However, because negative screens are not generally followed, no information is available about false negative rates in screening programs. Thus, neither the sensitivity nor the negative predictive value of screening can be calculated from study data.

Although many of the screening tests that may be used in preschool screening programs have been examined in experimental accuracy studies, not all (for example, the whisper test[28]) have been formally evaluated in program settings, raising questions about the feasibility of using some of these tests. For example, though PTA is often considered the standard, its appropriate application requires conditions and equipment that might limit its use as a screening method.[25] Additionally, as pointed out above, even with technologies such as OAE that are 'automatic,' it may be a challenge to ensure the appropriate expertise, as recommended in audiology guidelines, is available.

Limitations

This structured review has several limitations. First, the search covered a period of only 10 years. Given the use of UNHS in developed countries since the 1990, studies published before this time may exist that examined PHS in the absence of UNHS programs. Nevertheless, an examination of systematic reviews that examined this literature base[1,27,29] suggests that additional research relevant to this topic is unlikely to exist.

Conclusions

Within the context of a UNHS program, universal preschool hearing screening has not been demonstrated to be effective. Potential benefit may exist for targeted screening of at-risk youth; however, the realization of these benefits is likely hampered by significant challenges in ensuring timely referral, diagnosis, and treatment. Currently, no evidence is available with which to assess the safety of PHS programs. No identified studies have evaluated the performance and effectiveness of PHS programs in the absence of UNHS.

APPENDICES

Appendix T.A: Methodology

A systematic review of the primary and secondary scientific literature will be conducted to identify any HTAs, systematic reviews, or screening accuracy studies that examine preschool hearing programs.

Literature Search

Electronic searches of the peer-reviewed scientific literature published from 2002 onward will be conducted in the following databases: MEDLINE (including in-process), EMBASE, and CINAHL. In addition, reference lists of reviews and retrieved articles will be searched for relevant studies. A Google search will also be conducted for relevant information. Searches will be limited to English language articles.

Publication period: 2002 to 2012

Sources:

- **Electronic databases:** Medline (including in-process), EMBASE, CINAHL, and the Cochrane Library
- **Grey literature** search for HTAs or evidence based reports, clinical trial registries, clinical practice guidelines, position statements, and regulatory and coverage status
- **Reference lists** of the retrieved articles
- **World Wide Web:** A Google search will be conducted and the first 100 results will be assessed for relevance

Search terms: Terms for hearing loss and for hearing loss, screening, and diagnosis and their related terms, as well as terms for specific screens, will be used in the searches (see Appendix A).

Search limitation: limited to human studies; limited to studies published in English

Literature Search Summary: Final Search—T Section

General Information

The IHE research librarian conducted the literature search. The search was limited to articles published between 2002 and 2012. The search was developed and carried out prior to the study selection process. In addition to the strategy outlined below, reference lists of retrieved articles were reviewed for potential studies.

Table T.A.1: Search strategy

Database	Edition or date searched	Search Terms[††]
MEDLINE (includes in-process and other non-indexed citations) OVID Licensed Resource	2002 to April 4, 2012	1 hearing loss/ or deafness/ or hearing loss, bilateral/ or hearing loss, conductive/ or hearing loss, functional/ or hearing loss, high-frequency/ or hearing loss, mixed conductive-sensorineural/or hearing loss, sensorineural/ or hearing loss, central/or hearing loss, noise-induced/ or hearing loss, sudden/ or hearing loss, unilateral/ 2 (PCHI or deaf* or auditory neuropathy).tw. 3 (hearing adj2 (loss or impairment)).tw. 4 1 or 2 or 3 5 mass screening/ 6 (screen* or diagnos* or test or tests or testing).ti. 7 5 or 6 8 4 and 7 9 diagnostic techniques, otological/ or hearing tests/ or acoustic impedance tests/ or audiometry/or audiometry, evoked response/ or audiometry, pure-tone/ or exp audiometry, speech/ or psychoacoustics/or dichotic listening tests/ or recruitment detection, audiologic/ or otoscopy/ or vestibular function tests/or caloric tests/ or electronystagmography/ 10 ((hearing or audiological or auditory) adj1 (assessment* or screening or evaluation*)).tw. 11 Evoked Potentials, Auditory, Brain Stem/ or Otoacoustic Emissions, Spontaneous/ 12 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE).tw. 13 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*).tw. 14 9 or 10 or 11 or 12 or 13 15 14 and 4 16 8 or 15 17 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage).tw. 18 16 and 17 19 limit 16 to "preschool child (2 to 5 years)" 20 18 or 19 21 limit 20 to yr="2002 - 2012" 22 21 not cochlear implant*.ti. **1885 results**
Embase	2002 to April 4, 2012	1 hearing impairment/ or conduction deafness/ or congenital deafness/ or deaf-blindness/ or hearing loss/ or hypoacusis/ or mixed hearing loss/ or monaural hearing/ or perception deafness/ or sudden deafness/ or unilateral hearing loss/ 2 noise injury/ 3 vestibulocochlear nerve disease/ 4 (PCHI or deaf* or auditory neuropathy).tw. 5 (hearing adj2 (loss or impairment)).tw. 6 1 or 2 or 3 or 4 or 5 7 screening/ or mass screening/ or screening test/ 8 anonymous testing/ or auditory screening/ or developmental screening/

		9 (screen* or diagnos* or test or tests or testing).ti. 10 7 or 8 or 9 11 6 and 10 12 auditory system examination/ or hearing test/ or otoscopy/ or tonotopy/ or tympanometry/ 13 audiometry/ or audiography/ or evoked response audiometry/ or impedance audiometry/ or pure tone audiometry/ or speech audiometry/ 14 dichotic listening/ 15 exp vestibular test/ 16 electronystagmography/ 17 acoustic impedance/ 18 psychoacoustics.tw. 19 ((hearing or audiological or auditory) adj1 (assessment* or screening or evaluation*)).tw. 20 evoked brain stem auditory response/ 21 spontaneous otoacoustic emission/ 22 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE).tw. 23 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*).tw. 24 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 25 6 and 24 26 11 or 25 27 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage).tw. 28 26 and 27 29 limit 26 to preschool child <1 to 6 years> 30 28 or 29 31 limit 30 to yr="2002 -Current" 32 31 not cochlear implant*.ti. **2621 results**
Cochrane Library	April 5, 2012	#1 MeSH descriptor Hearing Loss explode all trees #2 (PCHI or deaf* or auditory neuropathy):ti,ab,kw #3 (hearing NEAR/2 (loss or impairment)):ti,ab,kw #4 (#1 OR #2 OR #3) #5 MeSH descriptor Mass Screening, this term only #6 (screen* or diagnos* or test or tests or testing):ti #7 (#5 OR #6) #8 (#4 AND #7) #9 MeSH descriptor Diagnostic Techniques, Otological explode all trees #10 ((hearing or audiological or auditory) NEAR/1 (assessment* or screening or evaluation*)):ti,ab,kw #11 MeSH descriptor Evoked Potentials, Auditory, Brain Stem explode all trees #12 MeSH descriptor Otoacoustic Emissions, Spontaneous explode all trees #13 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE):ti,ab,kw

		#14 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*):ti,ab,kw #15 (#9 OR #10 OR #11 OR #12 OR #13) #16 (#15 AND #4) #17 (#16 OR #8) #18 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage):ti,ab,kw #19 (#18 AND #17) #20 (#19), from 2002 to 2012 **60 results**
CINAHL	April 10, 2012	S26 S23 or S24 Limiters - Published Date from: 20020101-20121231 S25 S23 or S24 Search modes S24 S21 Limiters - Age Groups: Child, Preschool: 2-5 years S23 S21 and S22 S22 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage) S21 S11 or S20 S20 S5 and S19 S19 S12 or S13 or S14 or S15 or S16 or S17 or S18 S18 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*) S17 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE) S16 (MH "Otoacoustic Emissions, Spontaneous") S15 (MH "Evoked Potentials, Auditory, Brainstem") S14 ((hearing or audiological or auditory) N1 (assessment* or screening or evaluation*)) S13 (MH "Psychoacoustics") S12 (MH "Diagnosis, Ear") OR (MH "Hearing Tests") OR (MH "Acoustic Impedance Tests") OR (MH "Audiometry") OR (MH "Audiometry, Evoked Response") OR (MH "Audiometry, Pure-Tone") OR (MH "Audiometry, Speech") OR (MH "Dichotic Listening Tests") OR (MH "Otoacoustic Emissions, Evoked") OR (MH "Otoscopy") OR (MH "Vestibular Function Tests") OR (MH "Electronystagmography") S11 S9 or S10 S10 (MH "Hearing Screening") S9 S5 and S8 S8 S6 or S7 S7 TI (screen* or diagnos* or test or tests or testing) S6 (MH "Health Screening") S5 S1 or S2 or S3 or S4 S4 (hearing N2 (loss or impairment)) S3 (PCHI or deaf* or auditory neuropathy) S2 (MH "Auditory Neuropathy") S1 (MH "Hearing Disorders") OR (MH "Deafness") OR (MH "Deaf-Blind Disorders") OR (MH "Hearing Loss, Partial") OR (MH "Hearing Loss, Conductive") OR (MH "Hearing Loss, Functional") OR (MH "Hearing Loss, High-Frequency") OR (MH "Hearing Loss, Sensorineural") OR (MH "Hearing Loss, Central") OR (MH "Hearing Loss, Noise-Induced") **1663 results**

Guidelines		
AMA Clinical Practice Guidelines www.topalbertadoctors.org/cpgs.php?sid=1	April 16, 2012	Browsed list of topics **0 results**
NICE Guidance www.nice.org.uk/	April 16, 2012	("hearing screening" OR "hearing tests") **0 results**
CALSPA www.caslpa.ca	April 16, 2012	Browsed list **2 results**
ACSLPA www.acslpa.ab.ca/	April 17, 2012	Browsed list **1 result**
CMA Infobase http://mdm.ca/cpgsnew/cpgs/index.asp	April 16, 2012	Browsed list of publications Hearing **1 result**
National Guidelines Clearinghouse www.ngc.gov	April 16, 2012	Hearing **2 results**
Coverage/Regulatory/Licensing Agencies		
Aetna Clinical Policy Bulletins www.aetna.com/about/cov_det_policies.html	April 16, 2012	"Hearing screening" OR "hearing tests" **0 results**
HTA resources		
INESS www.inesss.qc.ca/	April 16, 2012	Hearing **0 results**
CADTH www.cadth.ca/index.php/en	April 16, 2012	Hearing **5 results**
Institue for Clinical and Evaluative Sciences (ICES), Ontario www.ices.on.ca/	April 16, 2012	Browsed list **0 results**
Health Technology Assessment Unit at McGill www.mcgill.ca/tau	April 16, 2012	Browsed list **0 results**
Medical Advisory Secretariat www.health.gov.on.ca/english/providers/program/mas/mas_mn.html	April 16, 2012	Browsed list **0 results**
Other Grey Literature Sources		
Proquest Dissertations and Theses	April 17, 2012	(Hearing screening OR hearing test*) AND (preschool OR child* OR school-age) **1 result**
Provincial Health Websites British Columbia www.gov.bc.ca/health/ Alberta www.health.alberta.ca/ Saskatchewan www.health.gov.sk.ca/ Manitoba www.gov.mb.ca/health/ Ontario www.health.gov.on.ca/en/	April 18, 2012 and April 25, 2012	("hearing screening" OR "hearing tests") British Columbia – **3 results** Alberta – **0 results** Saskatchewan – **1 result** Manitoba – **1 result** Ontario – **0 results**

Quebec www.msss.gouv.qc.ca/ New Brunswick www.gnb.ca/0051/index-e.asp Nova Scotia www.gov.ns.ca/DHW/ PEI www.gov.pe.ca/health/ Newfoundland/Labrador www.health.gov.nl.ca/health Yukon www.hss.gov.yk.ca NWT www.hlthss.gov.nt.ca/ Nunavut www.hss.gov.nu.ca/en/Home.aspx		Quebec – **0 results** New Brunswick – **0 results** Nova Scotia – **0 results** Prince Edward Island – **0 results** Newfoundland/Labrador – **0 results** Yukon – **0 results** Northwest Territories – **2 results** Nunavut – **0 results**
Search Engines		
Google	April 17, 2012	screening OR tests OR audiology preschool OR children OR school-age "hearing" –pubmed **11 results**

Literature Selection

One reviewer (KB) screened titles and abstracts, retrieved relevant articles, and determined eligibility of key studies, according to the inclusion and exclusion criteria below.

Inclusion Criteria

Publication status: complete report of study published in English

Study design: health technology assessments, systematic reviews, meta-analyses, randomized or non-randomized studies or cross-sectional screening efficacy studies. Due to time constraints, HTAs and systematic reviews[1] will be sought first. If no relevant research syntheses are found, the primary literature will be examined.

Population: preschool-aged children (≤6 years)

Intervention: universal or targeted hearing screening program (no restriction on technologies used) conducted in a country with a developed market economy as defined by the United Nations (that is, Australia, Canada, Germany, Italy, Japan, New Zealand, United Kingdom, United States)

Comparator: no preschool hearing screening program

Target condition: hearing loss (congenital, permanent progressive, delayed-onset, and acquired)

Setting: audiology clinic, school, pediatric or primary care physician clinic, other

Outcome of interest: at least one of the following:

- screening accuracy—that is, sensitivity and specificity
- therapuetic efficacy—for example, change-in-management

[1] A review is considered to be *systematic* if it meets the following criteria: (1) a focused clinical question, (2) explicit search strategy, (3) use of explicit, reproducible and uniformly applied criteria for article selection, (4) formal critical appraisal of the included studies, (5) narrative summary or quantitative data synthesis (that is, meta-analysis)

- patient-important functional outcomes—for example, improved speech and language ability, improved educational outcomes, reduced need for remediation, outcome of false-negative results

Language: limited to English

Publication period: January 2002 to February 2012

Exclusion Criteria

Publication status: conference abstracts, letters, news, editorial comments; not English-language publication

Study design: reports of single cases

Population: newborns and children aged >6 years

Condition: studies examining only children with conditions already associated with hearing difficulty, for example, Turner's syndrome, rubella, otitis media

Intervention: screening programs other than those designed specifically for hearing (for example, combined screening programs), or programs that take place in the absence of universal newborn hearing screening, or programs taking place in countries that do not have a developed economy (including countries having transitional economies)

Outcomes: studies that do not report quantitative data on any of the outcomes listed in the inclusion criteria

Quality assessment

Due to time constraints, no quality assessment of the collected reports was performed. Details of the population and screen were described and potential applicability to the Alberta context was also indicated.

Data extraction

Data was extracted by one reviewer (KB) according to a predetermined data extraction form.

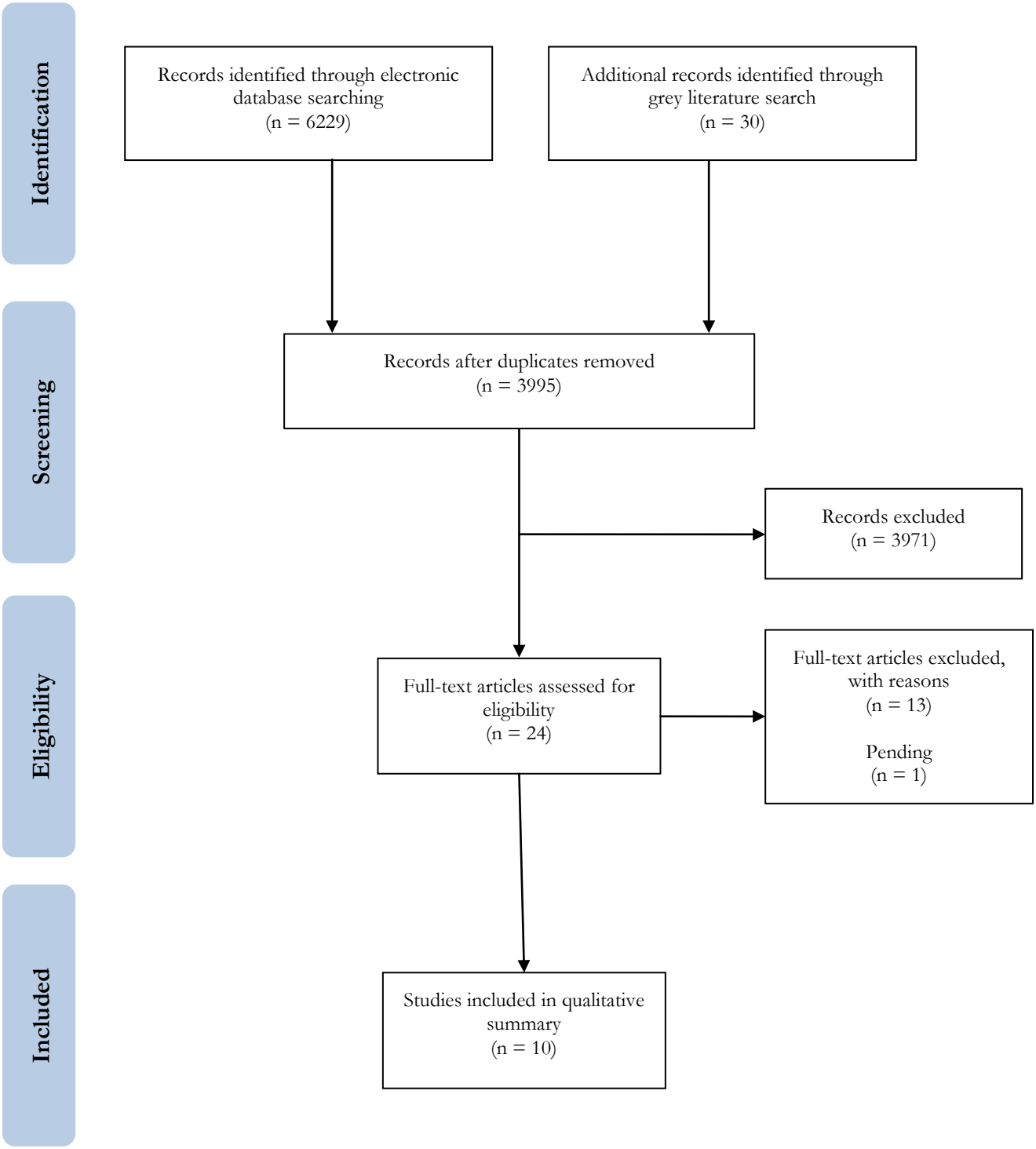
- **Study information:** first author, year of publication, country, number of sites
- **Study characteristics:** study design, timing of study, method of patient selection
- **Program characteristics:**
    - target population
    - eligibility criteria (age, developmental milestones, and so on)
    - patient recruitment (consecutive, random sampling, or other)
    - description of target conditions
    - prevalence of target condition (hearing loss overall and sensorineural vs conductive)
- **Screening test characteristics:**
    - technology used
    - device(s) used; (manufacturer)
    - timing of measurement

  - test positivity threshold
  - setting in which the test was performed
  - personnel responsible for performing the test
  - quality control procedure
- **Reference standard**
  - complete audiological examination
  - definition provided by study author
- **Outcomes**
  - screen accuracy:
    - sensitivity (for different cut-off) and 95% CI
    - specificity (for different cut-off) and 95% CI
  - diagnostic utility—for example, change in diagnosis
  - therapuetic efficacy—for example, change in treatment, reduced need for further testing
  - patient-important—for example, reduced need for remediation or more successful remediation, outcome of false-negative results

Data analysis and synthesis

A narrative approach was used to summarize the findings for accuracy (sensitivity and specificity), safety, therapeutic utility, patient-important outcomes. Due to time limitations, no quantitative analysis of the data was conducted.

Appendix T.B: Literature Search and Selection



Appendix C: Excluded Studies

Thirteen studies were excluded based on the literature selection criteria. One study was pending retrieval at cut-off time for study assessment and data extraction.

Not report of screening program (n = 4)

The following reports were excluded because they were not reports of primary research.

1. Beyond infant screening: what comes next? *Hearing Journal* 2002;55(11):13-59.
2. Biddle AM. School health services: education, screening and testing. *DNA Reporter* 2005;30(4):8.
3. Eiserman WD, Shisler L, Foust T. Hearing screening in early childcare settings. *ASHA Leader* 2008;13(15):34-7.
4. Kubba H. Whispered voice test for screening hearing impairment in adults and children: systematic review. *Journal of Pediatrics* 2004;144(5):684.

Not primary screening study (n = 3)

The following reports were excluded because they were not screening effectiveness studies.

1. Ayukawa H, Lejeune P, Proulx JF. Hearing screening outcomes in Inuit children in Nunavik, Quebec, Canada. *International Journal of Circumpolar Health* 2004;63 Suppl 2:309-11.
2. Johnson KC, Winter ME. Assessment of hearing in infants and toddlers. *Volta Review* 2003;103(4):219-409.
3. Taylor A, Bell A. Decreasing the Hearing Screen Referral Rate, One Unit's Experience. *Journal of Obstetric, Gynecologic & Neonatal Nursing* 2011;40:S25-S26.

Not population of interest (n = 1)

The following study was excluded because it did not include the population of interest.

1. Bigbee JL. Prevalence of failed hearing screening in ESL school-age children... 35th Annual Communicating Nursing Research Conference/16th Annual WIN Assembly, "Health Disparities: Meeting the Challenge," held April 18 to 20, 2002, Palm Springs, California. *Communicating Nursing Research* 2002;35:321.

Not condition of interest (n = 3)

The following studies were excluded because they did not examine the condition of interest.

1. Ho V, Daly KA, Hunter LL, Davey C. Otoacoustic emissions and tympanometry screening among 0-5 year olds. *Laryngoscope* 2002;112(3):513-9.
2. Pirozzo S, Papinczak T, Glasziou P. Whispered voice test for screening for hearing impairment in adults and children: systematic review. *BMJ* 2003;327(7421):967.
3. Sideris I, Glattke TJ. A comparison of two methods of hearing screening in the preschool population. *Journal of Community Disorders* 2006;39(6):391-401.

No quantitative outcomes of screening effectiveness or safety (n = 1)

The following study was excluded because it did not report quantitative outcomes of screening effectiveness or safety.

1. Driscoll C, Kei J, McPherson B. Hearing screening for children in community settings using transient evoked otoacoustic emissions. *Asia Pacific Journal of Speech, Language and Hearing* 2003;8(3):179-84.

Not English language (n = 1)

The following report was excluded because the full-text report was not published in English.

1. Brunner M, Pfeiffer B, Heinrich C, Proschel U. Development and testing of the Heidelberg preschool screening for auditory perception and speech processing (HVS). *Folia Phoniatrica et Logopedica* 2005;57(1):48-58.

Pending retrieval (n = 1)

The following report was not retrieved by the cut-off time for full-text assessment.

1. Eubanks CG. A model program for periodic childhood hearing screening in the medical home. *Perspectives on Administration & Supervision* 2007;17(3):20-2.

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2. Allen RL, Stuart A, Everett D, Elangovan S. Preschool hearing screening: pass/refer rates for children enrolled in a head start program in eastern North Carolina. *American Journal of Audiology* 2004;13(1):29-38.
3. Eiserman WD, Hartel DM, Shisler L, Buhrmann J, White KR, Foust T. Using otoacoustic emissions to screen for hearing loss in early childhood care settings. *International Journal of Pediatric Otorhinolaryngology* 2008;72(4):475-82.
4. Simpson SA, Thomas CL, van der Linden MK, Macmillan H, van der Wouden JC, Butler C. Identification of children in the first four years of life for early treatment for otitis media with effusion. *Cochrane Database of Systematic Reviews* 2007;(1):CD004163.
5. Alberta College of Speech-Language Pathologists and Audiologists. *Hearing screening guidelines*, 2008 (accessed 2012 May 28).
6. BC Early Hearing Program Diagnostic Audiology Advisory Group. *BC early hearing program diagnostic audiology protocol,* 2008.
7. Otologic Function Unit MSH. *Ontario infant hearing program audiologic assessment protocol (ver. 3.1).* Toronto, ON: 2008.
8. AAA Subcommittee on Childhood Hearing Screening. *American Academy of Audiology Childhood Hearing Screening Guidelines,* 2011.
9. ASHA panel on audiologic assessment. *Guidelines for audiologic screening,* 1997 (accessed 2012 May 27).
10. Harlor AD, Jr., Bower C, Committee on Practice and Ambulatory Medicine, Section on Otolaryngology-Head and Neck Surgery. Hearing assessment in infants and children: recommendations beyond neonatal screening. *Pediatrics* 2009;124(4):1252-63.
11. Sokol J, Hyde M. Hearing screening. *Pediatric Review* 2002;23(5):155-62.
12. Eiserman WD, Shisler L, Foust T. Hearing screening in early childcare settings. *ASHA Leader* 2008;13(15):34-7.
13. Sheykholeslami K, Kaga K. The otolithic organ as a receptor of vestibular hearing revealed by vestibular-evoked myogenic potentials in patients with inner ear anomalies. *Hearing Research* 165(1-2):62-7.
14. Widen JE, Keener SK. Diagnostic testing for hearing loss in infants and young children. *Mental Retardation & Developmental Disabilities Research Reviews* 9(4):220-4.
15. Psarommatis I, Valsamakis T, Raptaki M, Kontrogiani A, Douniadakis D. Audiologic evaluation of infants and preschoolers: a practical approach. *American Journal of Otolaryngology* 2007;28(6):392-6.

16. Damberger L, Flynn S. *Alberta Health and Wellness infant and preschool screening: current state assessment*. Edmonton, AB: Charis Management Consulting, Inc.; 2012.

17. Alberta Health Services. Kindergarten hearing screening program expands in northern Alberta. *Alberta Health Services webpage*; 2012 Jan 23 (accessed 2012 May 27).

18. Canadian Agency for Drugs and Technologies in Health. *Auditory screening and hearing loss prevention: a review of the clinical evidence and guidelines*. Ottawa, ON: Canadian Agency for Drugs and Technologies in Health; 2011 (accessed 2012 May 23).

19. Canadian Agency for Drugs and Technologies in Health. *Screening and detection of hearing loss: diagnostic accuracy of government versus private programs*. Ottawa, ON: Canadian Agency for Drugs and Technologies in Health; 2009.

20. Bristow K, Fortnum H, Fonseca S, Bamford J. United Kingdom school-entry hearing screening: current practice. *Archives of Disease in Childhood* 2007;93(3):232-5.

21. Serpanos YC, Jarmel F. Quantitative and qualitative follow-up outcomes from a preschool audiologic screening program: perspectives over a decade. *American Journal of Audiology* 2007;16(1):4-12.

22. Butler CC, van der Linden MK, Macmillan H, van der Wouden JC. Screening children in the first four years of life to undergo early treatment for otitis media with effusion. *Cochrane Database of Systematic Reviews* 2003;(2):CD004163.

23. Butler CC, van der Linden MK, MacMillan HL, van der Wouden JC. Should children be screened to undergo early treatment for otitis media with effusion? A systematic review of randomized trials. *Child: Care, Health and Development* 2003;29(6):425-32.

24. National Head Start Association. *Basic Head Start facts*; 2012. Available: www.nhsa.org (accessed 2012 May 24).

25. Feightner JW. Routine preschool screening for visual and hearing problems. In: *Canadian Task Force on the Periodic Health Examination Canadian Guide to Clinical Preventive Health Care*. Ottawa, ON: Health Canada; 1994:298-304.

26. Roberts JE, Rosenfeld RM, Zeisel SA. Otitis media and speech and language: a meta-analysis of prospective studies. *Pediatrics* 2004;113:238-48.

27. *Screening programmes for the detection of otitis media with effusion and conductive hearing loss in pre-school and new entrant school children*. Christchurch, NZ: 1998. NZHTA Report 3 (accessed 2012 May 29).

28. Pirozzo S, Papinczak T, Glasziou P. Whispered voice test for screening for hearing impairment in adults and children: systematic review. *BMJ* 2003;327(7421):967.

29. Rance G, Barker E, Mok M, Dowell R, Rincon A, Garratt R. Speech perception in noise for children with auditory neuropathy/dys-synchrony type hearing loss. *Ear and Hearing* 2007;28(3):351-60.

SECTION TWO: ECONOMICS ANALYSIS

Charles Yan, PhD; Anderson Chuck, PhD

OBJECTIVE AND SCOPE

The objective was to compare the cost-effectiveness of various strategies used in preschool hearing screening (PHS).

METHODS

A review was conducted of the published economic literature on the cost effectiveness of alternative strategies for PHS.

Search strategy

Selected databases were searched for economic evaluation studies of PHS. Databases searched included: Medline; EMBASE; CINAHL; Cochrane Database of Systematic Reviews; and grey literature. To supplement the electronic searches, reference lists of retrieved articles were also reviewed to find further studies. The literature search summary is presented in Appendix E.1.

Selection criteria

The search was limited to human and English language publications. Eligible studies were those that met the following predefined inclusion/exclusion criteria:

Inclusion Criteria

- **Study design**: Health technology assessment reports, systematic reviews and economic evaluation studies including studies of cost effectiveness, cost-utility, or cost-benefit
- **Population:** preschool-aged children (≤6 years)
- **Interventions and comparators:** various hearing screening strategies. Note that studies had to report the specific hearing tests used.
- **Language:** English
- **Search period:** from 2002 onward

Exclusion Criteria

- Abstracts, case studies, narrative reviews, letters and editorials
- Studies that reported the cost and outcomes of only one PHS strategy (without a comparator)
- Newborns and children aged >6 years

Outcomes of interest

- Quality-adjusted life years (QALYs)
- Number of correctly detected cases referred for follow-up/confirmatory testing
- Number of correctly identified non-cases not referred for follow-up/confirmatory testing

- Proportion of children whose hearing disorders were diagnosed within a follow-up period
- Cost per screen
- Cost per case detected
- Additional cost per health outcome gained

Quality assessment

A formal quality assessment of full economic studies was conducted with the Quality of Health Economic Studies (QHES) instrument.[1] The QHES instrument is designed to evaluate the quality of health economics, including cost-minimization, cost-effectiveness, and cost-utility analyses. It includes a scoring system to weight scores across 16 criteria. Scores are aggregated to provide a summative quality index. The quality index ranges from 0 to 100, with a score of 75 or greater indicating acceptable quality.

Data extraction

Data extracted from studies includes study objective, PHS strategies under investigation, cost components, health outcome measures, results, and conclusions.

RESULTS

Search results

In the literature search, 226 references were identified. After reviewing the titles and abstracts/summaries, 29 were retrieved for further review. Of these 29 studies, one HTA report (2007)[2] met the final inclusion/exclusion criteria. See Appendix E.2 for data extraction and Appendix E.3 for its quality assessment scores.

Evidence from economic literature

Bamford et al.[2] conducted a systematic review of economic studies evaluating the cost effectiveness of hearing screening in children. They identified no economic evaluation studies in the literature. However the authors conducted their own primary economic evaluation to assess the cost-effectiveness of school entry hearing screening for children in the United Kingdom aged 4 to 6 years, using a decision analytic model. The analysis compared seven screening options, including:

- no hearing screening
- universal school entry screening (SES) using pure-tone sweep audiometry only (SES-PTS)
- universal SES using parental questionnaire only (SES-PQ)
- universal SES using tympanometry only (SES-T)
- universal SES using spoken word tests only (SES-SW)
- targeted SES (low-accuracy and high-accuracy targeted SES)
- composite SES (SES-C)

The authors reported adopting a societal perspective by adopting the perspectives of NHS, education services, patients, and families. However, we could only detect resources associated with costs directly related to the screening service itself, such as labour and equipment costs associated with screening, diagnosis, treatment, and non-surgical hearing aids following a diagnosis of hearing

impairment. Hence, we could not verify that a societal perspective was applied correctly. Health outcomes were primarily measured using quality-adjusted life years (QALYs). The time horizon for the main analysis was 1 year.

Results showed that the expected QALYs were:

- 0.979 for no SES
- 0.983 for SES-C
- 0.983 for SES-PTS
- 0.975 for SES-T
- 0.977 for SES-PQ
- 0.964 for SES-SW
- 0.980 for low-accuracy targeted screening
- 0.988 for high-accuracy targeted screening

The expected costs per child were:

- £0.22 for no SES
- £10 for SES-C
- £9.9 for SES-PTS
- £10 for SES-T
- £23 for SES-PQ
- £30 for SES-SW
- £13 for low-accuracy targeted screening
- £11 for high-accuracy targeted screening

The marginal analysis indicated that the costs and outcomes of SES-PTS were similar to those of SES-C. Compared to no screening, SES-PTS/SES-C was more costly and more effective with a cost per additional QALY gained of approximately £2,500. Compared to low-accuracy screening (that is, SES-T, SES-PQ and SES-SW) and low-accuracy targeted screening, SES-PTS/SES-C was less costly and more effective. Compared to SES-PTS/SES-C, high-accuracy targeted screening was more costly and more effective, with an associated cost per QALY gained of £200. Compared to no screening, high-accuracy targeted screening was associated with a cost per QALY gained of £1198. Note that a difference in QALYs of 0.03 is considered clinically important. The study was assessed with a quality score of 80, indicating that it was of acceptable quality.

DISCUSSION

This review found one study[2] evaluating the cost-effectiveness of preschool screening for hearing impairment that was deemed to be of good quality, based on our quality assessment. Not surprisingly, compared to no screening, screening strategies were associated with additional costs and improved health outcomes. However, comparisons between screening strategies show that SES-PTS and SES-C were almost identical in costs and outcomes, and dominated the low-accuracy screening strategies of SES-T, SES-PQ, and SES-SW by being both cheaper and more effective.

Thus SES-T, SES-PQ, and SES-SW can be ruled out as cost-effective alternatives. Compared to SES-PTS/SES-C, high-accuracy targeted screening was associated with additional costs and improved health outcomes, although the improvement in health outcomes was not shown to be clinically important.

When comparing SES-PTS/SES-C with no screening, the cost per additional QALY gained was approximately £2,500. While £2,500 would be considered good value for money by most cost-effectiveness thresholds, because adopting SES-PTS/SES-C will both add costs while improving health outcomes compared to no screening, its cost-effectiveness is dependent on the opportunity cost of its adoption. That is, does investing the resources in the next best alternative use provide even greater health outcomes for the resources invested (that is, < £2,500 per QALY gained)? If not, then SES-PTS/SES-C is cost effective.

Additional effectiveness can be achieved in addition to SES-PTS/SES-C with high-accuracy targeted screening at a cost per QALY gained of approximately £200, but whether this is cost effective is, as described above, dependent on the opportunity cost of its adoption. However, it is important to note that high-accuracy targeted screening may indirectly dominate SES-PTS/SES-C. Compared to no screening, the incremental cost per additional QALY gained was £2,500 for SES-PTS/PTS-C and £1198 for high-accuracy targeted screening. Hence, compared to SES-PTS/SES-C, the same unit of effectiveness can be purchased at a lower cost with high-accuracy targeted screening. If the policy question is what screening strategy should be adopted, then high-accuracy targeted screening could provide the best value for money. Nonetheless, we must emphasize that, due to lack of primary data and the wide variation of estimates used in the analysis, these results are, as the authors point out, exploratory only.

Even if there were a high degree of confidence in the study results, there remains the question of whether the results are generalizable to the Alberta setting. An assessment of generalizability requires a comparison of the hearing screening services described in the study with the hearing screening services provided to preschool-aged children across Alberta. A universal hearing screening program is available for children entering kindergarten in Medicine Hat, and one is available on a pilot basis in Grande Prairie; all other zones within Alberta provide targeted hearing screening for preschoolers identified as being at risk.[3] However, the specific screening services provided in both universal and targeted screening contexts differ in terms of the screening tools, the professionals conducting the screen, and the location of the screening. Furthermore, the services are not provided in a systematic fashion. The wide variation in providers, tools, and processes across Alberta, combined with potential differences in the cost of resources between Alberta and the United Kingdom, make generalizability unlikely.

In conclusion—based on a single study—high-accuracy targeted screening may be the most cost effective among the screening alternatives assessed. However, the validity of the results and their applicability to the Alberta setting is unclear.

APPENDICES

Appendix E.1: Literature Search Summary: Preschool Hearing Screening Search—Economics

The IHE research librarian conducted this literature search. The search was limited to English-language publications and was developed and carried out prior to the study selection process. In addition to the strategy outlined below, reference lists of retrieved articles were reviewed for potential studies.

Database	Edition or date searched	Search Terms ††
MEDLINE (includes in-process and other non-indexed citations) OVID Licensed Resource	April 13, 2012	1 hearing loss/ or deafness/ or hearing loss, bilateral/ or hearing loss, conductive/ or hearing loss, functional/ or hearing loss, high-frequency/ or hearing loss, mixed conductive-sensorineural/ or hearing loss, sensorineural/ or hearing loss, central/ or hearing loss, noise-induced/ or hearing loss, sudden/ or hearing loss, unilateral/ 2 (PCHI or deaf* or auditory neuropathy).tw. 3 (hearing adj2 (loss or impairment)).tw. 4 1 or 2 or 3 5 mass screening/ 6 (screen* or diagnos* or test or tests or testing).ti. 7 5 or 6 8 4 and 7 9 diagnostic techniques, otological/ or hearing tests/ or acoustic impedance tests/ or audiometry/ or audiometry, evoked response/ or audiometry, pure-tone/ or exp audiometry, speech/ or psychoacoustics/ or dichotic listening tests/ or recruitment detection, audiologic/ or otoscopy/ or vestibular function tests/ or caloric tests/ or electronystagmography/ 10 ((hearing or audiological or auditory) adj1 (assessment* or screening or evaluation*)).tw. 11 Evoked Potentials, Auditory, Brain Stem/ or Otoacoustic Emissions, Spontaneous/ 12 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE).tw. 13 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*).tw. 14 9 or 10 or 11 or 12 or 13 15 14 and 4 16 8 or 15 17 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage).tw. 18 16 and 17 19 limit 16 to "preschool child (2 to 5 years)" 20 18 or 19 21 limit 20 to yr="2002 - 2012" 22 21 not cochlear implant*.ti. 23 exp "Costs and Cost Analysis"/ 24 (cost* or economic* or expensive*).tw.

		25 (expenditures or price or fiscal or financial or burden or efficiency or pay or valuation or spending or resource*).ti. 26 23 or 24 or 25 27 22 and 26 **103 results**
Embase	April 13, 2012	1 hearing impairment/ or conduction deafness/ or congenital deafness/ or deafblindness/ or hearing loss/ or hypoacusis/ or mixed hearing loss/ or monaural hearing/ or perception deafness/ or sudden deafness/ or unilateral hearing loss/ 2 noise injury/ 3 vestibulocochlear nerve disease/ 4 (PCHI or deaf* or auditory neuropathy).tw. 5 (hearing adj2 (loss or impairment)).tw. 6 1 or 2 or 3 or 4 or 5 7 screening/ or mass screening/ or screening test/ 8 anonymous testing/ or auditory screening/ or developmental screening/ 9 (screen* or diagnos* or test or tests or testing).ti. 10 7 or 8 or 9 11 6 and 10 12 auditory system examination/ or hearing test/ or otoscopy/ or tonotopy/ or tympanometry/ 13 audiometry/ or audiography/ or evoked response audiometry/ or impedance audiometry/ or pure tone audiometry/ or speech audiometry/ 14 dichotic listening/ 15 exp vestibular test/ 16 electronystagmography/ 17 acoustic impedance/ 18 psychoacoustics.tw. 19 ((hearing or audiological or auditory) adj1 (assessment* or screening or evaluation*)).tw. 20 evoked brain stem auditory response/ 21 spontaneous otoacoustic emission/ 22 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE).tw. 23 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*).tw. 24 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 25 6 and 24 26 11 or 25 27 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage).tw. 28 26 and 27 29 limit 26 to preschool child <1 to 6 years> 30 28 or 29 31 limit 30 to yr="2002 -Current" 32 31 not cochlear implant*.ti. 33 Health economics/ or exp economic evaluation/ or exp health care cost/ 34 exp "cost"/ 35 (cost* or economic* or expensive*).tw. 36 (expenditures or price or fiscal or financial or burden or efficiency or pay or

		valuation or spending or resource*).ti. 37 33 or 34 or 35 or 36 38 32 and 37 **143 results**
Cochrane	April 16, 2012	#1 MeSH descriptor **Hearing Loss** explode all trees #2 (PCHI or deaf* or auditory neuropathy):ti,ab,kw #3 (hearing adj2 (loss or impairment)):ti,ab,kw #4 (#1 OR #2 OR #3) #5 MeSH descriptor **Mass Screening**, this term only #6 (screen* or diagnos* or test or tests or testing):ti #7 (#5 OR #6) #8 (#4 AND #7) #9 MeSH descriptor **Diagnostic Techniques, Otological** explode all trees #10 ((hearing or audiological or auditory) NEAR/1 (assessment* or screening or evaluation*)):ti,ab,kw #11 MeSH descriptor **Evoked Potentials, Auditory, Brain Stem**, this term only #12 MeSH descriptor **Otoacoustic Emissions, Spontaneous** explode all trees #13 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE):ti,ab,kw #14 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*):ti,ab,kw #15 (#9 OR #10 OR #11 OR #12 OR #13 OR #14) #16 (#15 AND 4) #17 (#8 OR #16) #18 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage):ti,ab,kw #19 (#17 AND #18) #20 MeSH descriptor **Costs and Cost Analysis** explode all trees #21 (cost* or economic* or expensive*):ti,ab,kw #22 (expenditures or price or fiscal or financial or burden or efficiency or pay or valuation or spending or resource*):ti #23 (#20 OR #21 OR #22) #24 (#23 AND #19) **23 results**
CINAHL	April 16, 2012	S29 S27 and S28 S28 economic* or cost* S27 S23 or S25 S26 S23 or S25 S25 S21 S24 S21 S23 S21 and S22 S22 (child* or pediatric* or paediatric* or preschool or pre-school or school-age or schoolage)

		S21 S11 or S20 S20 S5 and S19 S19 S12 or S13 or S14 or S15 or S16 or S17 or S18 S18 (conditioned oriented response* or tympanometry or otoscopy or COR or VRA or audiometry or behavi?ral audiogram*) S17 (auditory brainstem response or ABR or AABR or otoacoustic emission* or OAE or AOAE or TEOAE or DPOAE) S16 (MH "Otoacoustic Emissions, Spontaneous") S15 (MH "Evoked Potentials, Auditory, Brainstem") S14 ((hearing or audiological or auditory) N1 (assessment* or screening or evaluation*)) S13 (MH "Psychoacoustics") S12 (MH "Diagnosis, Ear") OR (MH "Hearing Tests") OR (MH "Acoustic Impedance Tests") OR (MH "Audiometry") OR (MH "Audiometry, Evoked Response") OR (MH "Audiometry, Pure-Tone") OR (MH "Audiometry, Speech") OR (MH "Dichotic Listening Tests") OR (MH "Otoacoustic Emissions, Evoked") OR (MH "Otoscopy") OR (MH "Vestibular Function Tests") OR (MH "Electronystagmo-graphy") S11 S9 or S10 S10 (MH "Hearing Screening") S9 S5 and S8 S8 S6 or S7 S7 TI (screen* or diagnos* or test or tests or testing) S6 (MH "Health Screening") S5 S1 or S2 or S3 or S4 S4 (hearing N2 (loss or impairment)) S3 (PCHI or deaf* or auditory neuropathy) S2 (MH "Auditory Neuropathy") S1 (MH "Hearing Disorders") OR (MH "Deafness") OR (MH "Deaf-Blind Disorders") OR (MH "Hearing Loss, Partial") OR (MH "Hearing Loss, Conductive") OR (MH "Hearing Loss, Functional") OR (MH "Hearing Loss, High-Frequency") OR (MH "Hearing Loss, Sensorineural") OR (MH "Hearing Loss, Central") OR (MH "Hearing Loss, Noise-Induced") **85 results**
Guidelines		
AMA Clinical Practice Guidelines www.topalbertadoctors.org/cpgs.php?sid=1 /	April 16, 2012	Browsed list of topics **0 results**
NICE Guidance www.nice.org.uk/	April 16, 2012	(“hearing screening” OR “hearing tests”) **0 results**
CALSPA www.caslpa.ca	April 16, 2012	Browsed list **2 results**
ACSLPA www.acslpa.ab.ca/	April 17, 2012	Browsed list **1 result**
CMA Infobase http://mdm.ca/cpgsnew/cpgs/index.asp	April 16, 2012	Browsed list of publications Hearing **1 result**

National Guidelines Clearinghouse www.ngc.gov	April 16, 2012	Hearing **2 results**
Coverage/Regulatory/Licensing Agencies		
Aetna Clinical Policy Bulletins www.aetna.com/about/cov_det_policies.html	April 16, 2012	("hearing screening" OR "hearing tests") **0 results**
HTA resources		
INESS www.inesss.qc.ca/	April 16, 2012	Hearing **0 results**
CADTH www.cadth.ca/index.php/en/	April 16, 2012	Hearing **5 results**
Institue for Clinical and Evaluative Sciences (ICES), Ontario www.ices.on.ca/	April 16, 2012	Browsed list **0 results**
Health Technology Assessment Unit At McGill www.mcgill.ca/tau/	April 16, 2012	Browsed list **0 results**
Medical Advisory Secretariat www.health.gov.on.ca/english/providers/program/mas/mas_mn.html	April 16, 2012	Browsed list **0 results**
Other Grey Literature Sources		
Proquest Dissertations and Theses	April 17, 2012	(hearing screening OR hearing test*) AND (preschool OR child* OR school-age) **1 result**
Provincial Health Websites British Columbia www.gov.bc.ca/ health/ Alberta www.health.alberta. ca/ Saskatchewan www.health.gov.sk. ca/ Manitoba www.gov.mb.ca/ health/ Ontario www.health.gov.on. ca/en/ Quebec www.msss.gouv.qc. ca/ New Brunswick www.gnb.ca/0051/ index-e.asp Nova Scotia www.gov.ns.ca/ DHW/ PEI www.gov.pe.ca/health/ Newfoundland/Labrador www.health.gov.nl. ca/health/ Yukon www.hss.gov.yk.ca/ NWT	April 18, 2012 and April 25, 2012	("hearing screening" OR "hearing tests") British Columbia – **3 results** Alberta – **0 results** Saskatchewan – **1 result** Manitoba – **1 result** Ontario – **0 results** Quebec – **0 results** New Brunswick – **0 results** Nova Scotia – **0 results** Prince Edward Island – **0 results** Newfoundland and Labrador – **0 results** Yukon – **0 results** Northwest Territories – **2 results**

www.hlthss.gov.nt.ca Nunavut www.hss.gov.nu.ca/en/Home.aspx		Nunavut – **0 results**
Search Engines		
Google	April 17, 2012	screening OR tests OR audiology preschool OR children OR school-age "hearing" –pubmed **11 results**

Note:

††, *, #, and ? are truncation characters that retrieve all possible suffix variations of the root word, for example, surg* retrieves surgery, surgical, surgeon, and so on.

Search Strategy: # Searches Results

Appendix E.2: Summarized Evidence from Selected Studies

#	Item	Description
1	Study	Authors/publish year: Bamford et al./2007; country: the UK; study type: CEA, CUA; Setting: school/community; study perspective: society
	Objective	The objective was to assess the cost-effectiveness of various screening options for amblyopia and strabismus.
	Population	Children aged 4 to 6 years
	Intervention	The compared strategies were: • no school-entry hearing screening (SES) • universal SES, using pure-tone sweep audiometry only (SES-PTS) • universal SES, using parental questionnaire only (SES-PQ • universal SES, using tympanometry only (SES-T • universal SES, using spoken word tests only (SES-SW) • targeted SE • composite SEC (SES-C) The targeted SES screens children identified as being at risk of hearing impairment. The current practice in the UK (i.e., SES-C) was the combination of pure-tone sweep audiometry (99%) and tympanometry (1%).
	Time horizon/ discount rate	1, 6, and 11 years /3.5%
	Currency/price year	£/2004
	Outcomes measure	QALY and years with no or mild disability due to hearing impairment
	Cost components	Cost categories considered in the analysis were screening, diagnosis, surgical treatment, and non-surgical hearing aids, following a diagnosis of hearing impairment. The costs of screening and diagnosis were for equipment, maintenance, supplies and consumables, and staff time. The cost of non-surgical hearing aids were for staff, follow-up monitoring, and hearing aid replacement.
	Results	
	Outcomes	Over 1 year, the expected QALYs were 0.979 for no SES, 0.983 for SES-C and SES-PTS, 0.975 for SES-T, 0.977 for SES-PQ, 0.964 for SES-SW, 0.980 for low-accuracy targeted screening, and 0.988 for high-accuracy targeted screening. In the population with a low prevalence of unidentified permanent hearing impairment, the expected QALYs were 0.985 for SES-C and 0.983 for no SES; and in the population with a high prevalence, the expected QALYs were 0.986 for SES-C and 0.985 for no SES. The expected years with no to mild hearing impairment (YNHIs) was 0.999 for SES-C and 1 for no SES. YNHIs for other strategies were not reported. Over 6 (and 11) years, the expected QALYs were 5.37 (and 9.07) for SES-C and 5.27 (and 8.91) for no SES. QALYs for other strategies were not reported.

Costs	Over 1 year, the expected costs per child were £0.22 for no SES, £10 for SES-C, £9.9 for SES-PTS, £10 for SES-T, £23 for SES-PQ, £30 for SES-SW, £13 for low-accuracy targeted screening, and £11 for high-accuracy targeted screening. In the population with a low prevalence of unidentified permanent hearing impairment, the expected costs per child were £9 for SES-C and £0.11 for no SES; and in the population with a high prevalence, the expected QALYs were £9 for SES-C and £0.08 for no SES. Over 6 (and 11) years, the expected costs per child were £25 (and £30) for SES-C and £2 (and £3) for no SES. Costs for other strategies were not reported.
Marginal Analysis	Over 1 year, the incremental cost-effectiveness ratio (ICER) of SES-C over no screening was £2445 per QALY gained. SES-C generated the same QALYs at a similar cost to SES-PTS. Sensitivity analysis indicated that: • compared with no screening, SES-C is more than 50% cost effective over 1 year and more than 99% cost-effective over 6 and 11 years if the WTP threshold value is more than £2,000 • compared with SES-PTS, SES-C is 60% cost effective • compared with no screening, SES-PQ and SES-SW were less cost effective and SES-T was more cost effective • compared with SES-C, SES-T, SES-PQ, and SES-SW were less cost effective • SES-C was more cost effective than targeted SES When considering YNHIs as the benefit measure, no screening is likely to be cost-effective over 1 year.
Conclusion	The screening based on pure-tone sweep audiometry is more cost effective than no screening and other less accurate alternatives. Targeted screening using high accuracy approach could be more cost effective than universal SES.

Appendix E.3: QHES Instrument

#	Questions	QHES Scores
		Banford, 2007
1	Was the study objective presented in a clear, specific, and measurable manner?	7
2	Were the perspective of the analysis (societal, third-party payer, etc.) and reasons for its selection stated?	1
3	Were variable estimates used in the analysis from the best available source (i.e., randomized control trial—best, expert opinion—worst)?	6
4	If estimates came from a subgroup analysis, were the groups pre-specified at the beginning of the study?	1
5	Was uncertainty handled by (1) statistical analysis to address random events, (2) sensitivity analysis to cover a range of assumptions?	7
6	Was incremental analysis performed between alternatives for resources and costs?	5
7	Was the methodology for data abstraction (including the value of health states and other benefits) stated?	5
8	Did the analytic horizon allow time for all relevant and important outcomes? Were benefits and costs that went beyond one year discounted (3% to 5%) and justification given for the discount rate?	7
9	Was the measurement of costs appropriate and the methodology for the estimation of quantities and unit costs clearly described?	8
10	Were the primary outcome measure(s) for the economic evaluation clearly stated and did they include the major short-term, long-term, and negative outcomes?	5
11	Were the health outcomes measures/scales valid and reliable? If previously tested valid and reliable measures were not available, was justification given for the measures/scales used?	4
12	Were the economic model (including structure), study methods and analysis, and the components of the numerator and denominator displayed in a clear, transparent manner?	8
13	Were the choice of economic model, main assumptions, and limitations of the study stated and justified?	5
14	Did the author(s) explicitly discuss direction and magnitude of potential biases?	0
15	Were the conclusions/recommendations of the study justified and based on the study results?	8
16	Was there a statement disclosing the source of funding for the study?	3
	TOTAL POINTS	**80**

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Author Contribution Statements

Ken Bond contributed to study conception and design, data analysis and interpretation, and approved the final version for publication.

Christa Harstall contributed to study conception and design, revision of manuscript for critical content, and approved the final version for publication.

Charles Yan contributed to study conception and design, statistical analysis, economic expert review of the literature, revision of manuscript for critical content, and approved the final version for publication.

Anderson (Andy) Chuck contributed to study conception and design, statistical analysis, economic expert review of the literature, manuscript preparation, and approved the final version for publication.

Dagmara Chojecki developed and executed the literature search.

This report performs an evaluation of the scientific evidence on the safety, performance, and effectiveness of universal and targeted preschool hearing screening (PHS) to inform the Infant and Preschool Screening Framework being developed by the Community and Public Health (CPH) Division of AHW, as well as comparing the cost-effectiveness of various strategies used in preschool hearing screening, through a review of the published economic literature.

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