Oto-acoustic emission audiometry

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MSAC application 1002

Final assessment report

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The Secretary
Medicare Services Advisory Committee
Commonwealth Department of Health and Aged Care
Mail Drop 107
GPO Box 9848
Canberra ACT 2601

Enquiries about the content of the report should be directed to the above address.

The Medicare Services Advisory Committee is an independent committee which has been established to provide advice to the Commonwealth Minister for Health and Aged Care on the strength of evidence available on new medical technologies and procedures in terms of their safety, effectiveness and cost-effectiveness. This advice will help to inform Government decisions about which new medical services should attract funding under Medicare.

This report was prepared by the Medicare Services Advisory Committee (MSAC). The report was endorsed by the Commonwealth Minister for Health and Aged Care on 9 August 1999.

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MSAC recommendations do not necessarily reflect the views of all individuals who participated in the MSAC evaluation.

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Executive summary

The procedure

Oto-acoustic emission audiometry (OAEA) is a diagnostic test for hearing impairment.

Oto-acoustic emissions (OAE) are narrowband acoustic signals generated by the inner ear of normal individuals, either in the absence of acoustic stimulation (spontaneous emissions) or in response to acoustic stimulation (evoked emissions). These emissions can be detected by analysing the signals obtained by placing a tiny microphone at the entrance to the ear canal, a simple and non-invasive procedure.

Medicare Services Advisory Committee – role and approach

The Medicare Services Advisory Committee (MSAC) is a key element of a measure taken by the Commonwealth Government to strengthen the role of evidence in health financing decisions in Australia. MSAC advises the Commonwealth Minister for Health and Aged Care on the evidence relating to the safety, effectiveness and cost-effectiveness of new medical technologies and procedures, and under what circumstances public funding should be supported.

A rigorous assessment of the available evidence is thus the basis of decision making when funding is sought under Medicare. A team from the Australasian Cochrane Centre was engaged to conduct a systematic review of literature on OAEA. A supporting committee with expertise in this area then evaluated the evidence and provided advice to MSAC.

Assessment of Oto-acoustic Emission Audiometry

The clinical studies undertaken to date on the sensitivity and specificity of OAEA all have methodological limitations. A common flaw identified in many of the studies is the failure to evaluate OAEA independently of the reference test (blind), leaving open the possibility of bias. While there is strong theoretical evidence that earlier intervention in hearing impairment may result in better outcomes for infants with permanent hearing impairment, there are no randomised controlled trials available to support this and it is considered not feasible to conduct such studies. However, the cohort studies which have been conducted in this area indicate that intervention before the age of three to six months results in improved communication skills.

Clinical need

Estimates of the prevalence of hearing impairment in the population vary depending on the definition used. The Working Party on 'Early Identification of Hearing Impairment in Children in NSW' used the criterion of permanent hearing loss of 40dB or worse in the better ear.[1]

In Australia, there were 253,673 live births in 1997.[2] Given the reported prevalence of hearing impairment in children of 0.1 to 0.2 per cent, the expected number of children affected in Australia is 254 to 507 per year.

Safety

OAEA is a non-invasive test. No adverse outcomes were reported in any of the trials included in the review. Risks associated with the test are the consequences of false positive or false negative test results.

Effectiveness

Review of the literature has shown OAEA is reasonably sensitive and specific when compared with other forms of audiology, although there was significant variation between the results of the studies included in the review. It is particularly useful for the pre-lingual child where testing requiring behavioural responses may be unreliable. It has a particularly high negative predictive value, that is, where the test indicates that hearing impairment is not present, it is highly likely that the child's hearing is normal. On the other hand the positive predictive value of the test is lower. In the included studies this varies between 4 per cent and 73 per cent. This is partly because it will detect large numbers of children with otitis media with effusion, in whom the benefits of detection with OAEA are not proven. In addition, the relative rarity of permanent hearing impairment means that even a test with high sensitivity and specificity will detect large numbers of patients who on further testing do not have the condition.

Cost-effectiveness

The main focus of this report has been a systematic review of the effectiveness of OAEA as a diagnostic test. It was not possible within the scope of this review to do a full economic evaluation of the technology.

Recommendations

It is recommended that on the strength of evidence pertaining to OAEA, public funding should be supported for this procedure for the detection of permanent congenital hearing impairment in groups of children at high risk due to the following factors:

- admission to a neonatal intensive care unit
- family history of hearing impairment
- perinatal infection (either suspected or confirmed)
- birthweight <1.5kg
- craniofacial deformity
- birth asphyxia
- chromosomal abnormality, including Down syndrome
- exchange transfusion

In addition, it is recommended that OAEA be preceded by a specialist assessment to exclude middle ear pathology.

Introduction

The Medicare Services Advisory Committee (MSAC) has reviewed the use of oto-acoustic emission audiometry (OAEA), which is a diagnostic hearing test for infants and children at risk of, or actively suspected of, impaired hearing. MSAC evaluates new health technologies and procedures for which funding is sought under the Medicare Benefits Scheme in terms of their safety, effectiveness and cost-effectiveness, while taking into account other issues such as access and equity. MSAC adopts an evidence-based approach to its assessments, based on reviews of the scientific literature and other information sources, including clinical expertise.

MSAC's terms of reference and membership are at Appendix A. MSAC is a multidisciplinary expert body, comprising members drawn from such disciplines as diagnostic imaging, pathology, surgery, internal medicine and general practice, clinical epidemiology, health economics and health administration.

This report summarises the current evidence of the effectiveness of OAEA as a diagnostic hearing test for infants and children at risk of, or actively suspected of, impaired hearing. Whilst much of the literature has assessed the use of OAEA as a screening tool in infants, this report does not seek to address the effectiveness of OAEA in screening for disease.

Background

Oto-acoustic Emission Audiometry

The procedure

OAEA is a diagnostic test for hearing impairment.

In 1978, Kemp made the first recordings of sounds produced by the cochlea – generally known as 'oto-acoustic emissions' (OAE). OAEs are narrowband signals generated by the cochlea in non-hearing impaired individuals and occur either spontaneously (spontaneous oto-acoustic emissions) or in response to a sound stimulus (evoked oto-acoustic emissions).[3] It is thought that these signals are generated by the same biofeedback mechanisms that are involved in the ability of the normal ear to detect and analyse low level sounds. The weakening or loss of this process is one of the earliest pathological changes in hearing impairment.

It is possible to record and analyse OAEs with the use of a tiny probe, which contains a microphone and earphone, placed inside the entrance of the ear canal. Because the response generated by the cochlea is small, it is necessary to present a number of stimuli and to average the response. Generally, OAEs will be present when hearing is better than 20 to 30 decibel measure of hearing level (dB HL). The better a person's hearing, the smaller the decibel hearing level, with up to 15dB HL generally regarded as the level of hearing considered optimal for 'normal' speech development in children.[4] The OAEs are generated by sound stimuli of differing magnitudes and are recorded and graphed using computerised frequency analysis. The presence of OAEs can therefore determine if a person has a functioning cochlea since a pass or fail results. However, the method is unable to detect the threshold level of hearing. Therefore, it is unable to differentiate between moderate, severe and profound levels of hearing loss. Once the possibility of hearing impairment has been detected by OAEA, a full auditory assessment is necessary to confirm the diagnosis.

OAEs are evoked either by a 'click' sound stimulus, referred to as transient evoked oto-acoustic emission audiometry (TOAEA), or by a two-tone sound stimulus, referred to as distortion product oto-acoustic emission audiometry (DPOAE). There are also newer versions of OAEA which are quicker and are likely to be more useful in noisy situations. All of the studies included in this review used OAEA, using the commercially available IL088 device.

The test takes 10 to 30 minutes to perform which includes preparation of the patient, explanation and recording of results. A nurse or technician can perform the test with a sample of results reviewed by a supervising audiologist. Initial training to use the equipment takes four hours.[5] A proportion of children will be unable to be tested because it is not possible to settle the child and a proportion of responses will be borderline.

OAEA can also be used for the diagnosis of hearing loss in adults. Because it measures the function of the cochlea hair cells, it is highly sensitive and can detect the loss of the function of these cells prior to any detectable loss in hearing. This has potential value in monitoring hearing loss due to occupational or environmental exposure. DPOAE is more suited to the testing of adult ears because it achieves technically better results above 4kHz (the high frequency loss which is more common in adults) and is able to extract responses to more intense sustained stimulation.[6]

Intended purpose

The MSAC application proposed that OAEA is indicated for individuals in whom hearing loss is suspected and/or in whom risk factors for hearing impairment have been identified, where conventional hearing tests are unlikely to be adequate or appropriate. For the purposes of this MSAC application, OAEA is not under consideration as a method of universal neonatal hearing screening.

Conventional audiology, such as visual reinforcement audiometry (VRA), commonly requires a behavioural response to a sound stimulus. Such methods are difficult to use reliably in a pre-lingual child. OAEA offers a diagnostic method that requires no active cooperation from the individual being assessed and can even be performed whilst the patient is asleep

In the current review, OAEA is proposed for use in high risk infants. The application specifies no restrictions on clinical setting. However, it is recommended that the procedure only be undertaken under specialist supervision. OAEA is proposed as a second line diagnostic test where other methods are considered to be inappropriate and is not under consideration as a method of universal neonatal hearing screening.

Clinical need/burden of disease

Estimates of the prevalence of hearing impairment in the population vary depending on the definition used. Diagnostic criteria range from greater than 40dB HL to greater than 90dB HL and also depend on whether bilateral or unilateral impairment is considered. The Working Party on 'Early Identification of Hearing Impairment in Children in NSW' used the criterion of permanent hearing loss of 40dB or worse in the better ear[1]. They chose this level because it is a level of hearing impairment which:

- may lead to significant educational and psychosocial delay;
- can practically be detected in young children; and
- in the absence of an internationally agreed standard, is commonly used in research.

Study of the literature has confirmed the last point, in that the majority of studies included in this report used a criterion of permanent loss of 40dB or worse in the better ear as the definition of hearing impairment. Most studies have averaged the threshold of hearing loss over the frequencies 0.5, 1, 2 and 4 kHz.

A summary of estimates of the prevalence of hearing impairment in children is shown in Table 1.

Table 1 Studies of prevalence of permanent hearing impairment in children

Population	Country	Prevalence of hearing impairment	Criteria used	Studies
Total newborn population	UK	0.2%	>50dB HL in better ear in infants up to 3 months	Watkin, 1996[7]
	UK	0.12%	>50dB PHL at 5 years	Davis, 1993[8]
	UK	0.1%	>50dB in children up to 3 years	Davis and Wood, 1992[9]
	UK	0.13%	>40dB hearing loss in the better ear in children 21 months to 4.5 years	Fortnum and Davis, 199710
	UK	0.02%	>100dB hearing loss in better ear in same study	Fortnum and Davis, 199710
	USA	0.6%	>60dB hearing loss (sensorineural) in better ear in infants up to 6 months	White et al, 199311
	Australia	0.08%	>60dB hearing impairment per live births in period 1979 to 1988	AHS Annual Statistics, 1996 reported in Birtles et al, 19981
	Europe	0.1%	>50dB hearing loss in better ear, ascertained by 1977 for children born during 1969	Martin et al, 198112
Neonatal intensive care	UK	1.4%	>30dB hearing loss in better ear for babies in one hospital's NICU during 1986	Rowe, 199113

dB HL= decibel measure of hearing level; PHL = permanent hearing loss; NICU = neonatal intensive care unit

In Australia, there were 253,673 live births in 1997.[2] Assuming that approximately 0.1 to 0.2 per cent of children will be affected with a permanent congenital hearing impairment (PCHI) of 40dB or greater, the expected number of children affected in Australia is 254 to 507 per year.

Approximately 50 per cent of infants with PCHI have an identifiable risk factor. Risk factors for permanent hearing loss include:

- admission to a neonatal intensive care unit (NICU)
- family history of hearing impairment
- perinatal infection (either suspected or confirmed)
- birthweight <1.5kg
- craniofacial deformity
- birth asphyxia
- chromosomal abnormality, including Down syndrome
- exchange transfusion[14]

Testing of neonates who have been admitted to special care and intensive care units accounts for approximately 40 per cent of infants identified with PCHI.[14,15] It is possible that an improvement in the mortality rate for low birth weight infants and infants admitted to NICUs has resulted in an increase in the prevalence of permanent hearing impairment in children. There is some anecdotal evidence of an increase in sensorineural hearing loss seen in paediatric clinics, which is speculated to be a result of this.[16] More ready identification of children with mild or moderate hearing impairment may also influence reported rates.

About 10 to 20 per cent of permanent childhood hearing impairment will be acquired after birth.[10] The majority of acquired hearing impairment is secondary to bacterial meningitis. Permanent sensorineural hearing loss occurs in 3 to 10 per cent of children who have had bacterial meningitis. This may be bilateral or unilateral.[17] In the majority of cases this is due to damage to cochlea cells, but meningitis may also affect acoustic nerve and brainstem function, which will not be detected by OAEA.

A small number of children with a permanent hearing impairment loss will have a retrocochlea lesion. This will not be detected by testing with OAEA, but the majority of these children will show other signs of neurological impairment. Prevalence data were not available, but expert opinion is that this is less than 1 per cent of all PCHI.[5]

A common reason for audiological testing is suspected hearing loss secondary to middle ear infection. The prevalence of this form of conductive hearing loss is high and will be detected by OAEA: 'Otitis media with effusion, though extremely common, only rarely causes problems severe and prolonged enough to warrant special educational assistance. It will, however, be a common reason for children failing primary screening'.[1] In addition, the benefits of early intervention in such cases of mild or temporary hearing impairment are uncertain.[18,19,20]

The impact of permanent hearing impairment in children

It has long been appreciated that there are critical periods in infant development for speech and language development. If appropriate auditory stimuli are not provided at these times, the result is lifelong linguistic and communication deficit.[21] Greater severity of hearing loss is associated with worse outcomes in terms of communication abilities.

A number of studies have documented the effect of moderate or greater hearing impairment on language and communication skills (for example, Bench and Bamford,[22] Levitt et al,[23] and Gregory and Mogford[24]). Hearing impairment also results in reading disability, with more than half of profoundly deaf children unable to read.[25] This results in decreased educational, employment and life opportunities.[26,27] Deaf children also frequently suffer from social isolation.[5,28] One estimate of the lifetime economic cost of congenital deafness was in excess of US$1 million.[4]

The key interventions for moderate to profound permanent childhood hearing impairment are:

- family support, advice and information
- provision of hearing aids or cochlea implants as required

- provision of communication support (spoken and/or signed)
- provision of pre-school educational support
- provision of other devices, eg radio aids[29]

Patients with a hearing loss >95dB are considered profoundly deaf. Such children may be considered for a cochlea implant.

The effect of mild or unilateral PCHI is uncertain. Mostly this is due to the lack of rigorous data on the epidemiology of these conditions.[5] In the past, unilateral hearing loss was thought to have little developmental impact. However, some studies suggest that any degree of unilateral loss may be associated with behavioural problems and learning difficulties. Oyler et al[30] found that 23.7 per cent of children with unilateral loss had repeated at least one grade, approximately 10 times the proportion for unimpaired children in the same school district. They conclude that unilateral loss may greatly increase the risk of academic failure. Brookhouser et al[31] found 59 per cent of children with unilateral hearing loss had a history of behavioural problems at school, of which 17 per cent had speech or language delay. The authors suggest that a relatively late average age of diagnosis and subsequent inadequate intervention may influence the impact of the hearing loss. In this sample even profound hearing loss was not identified until six years or older. As discussed by the authors, behavioural problems and learning difficulties may stem from an inability to localise and extract individual auditory signals, such as a teacher's voice, from normal background classroom noise.

Does early identification reduce the impact of permanent hearing impairment?

A review on this question was undertaken by Davis et al,[5] as part of the United Kingdom's Health Technology Assessment report on the universal screening of neonates for hearing impairment.

There are no randomised controlled trials on the benefits of early intervention in children with a permanent hearing impairment. A randomised trial of early intervention versus no intervention would not be feasible because of strong parental and physician preference for intervention. To do a trial of screened versus non-screened children would require large numbers of infants to be assessed to find the 0.1 to 0.2 per cent of children affected and in whom the outcomes of interest could be measured.

The review by Davis et al[5] identified 18 studies on the impact of early intervention in children with PCHI. Thirteen of these had strong methodological problems such as a lack of a reliable control group or a high degree of confounding. Four studies provide some weak evidence that earlier identification is associated with better outcomes, but all four of the studies provide only Level III evidence.[32]

An observational study of children identified by a neonatal screening program in Colorado provides some evidence of the effect of early intervention.[33] This study showed that children fitted with a hearing aid before the age of three months (n=69) scored 87 per cent of what was regarded as normal expressive language for the age of the child at the time of assessment, compared with only 66 per cent for children fitted with a hearing aid between the ages of 3 and 12 months. Yoshinago-Itano et al[34] examined the receptive and expressive language ability of 72 hearing impaired children identified before the age

of six months with 78 children identified after this age. The authors found that earlier-identified children with normal cognitive abilities had significantly higher language scores across all test ages, communication modes – spoken or sign language, degrees of hearing loss, socioeconomic strata and a variety of other variables generally associated with language abilities. In her longitudinal study of communicative and linguistic development, Robinshaw[35] found that five hearing impaired children fitted with hearing aids by six months acquired vocal communicative and linguistic skills at comparable ages and in a similar pattern to their normal hearing matched partners. While later-identified children from an earlier study also followed a similar developmental pattern, they did so at a far less 'typical' age than the early-identified group.

Markides[36] studied 153 children who were fitted with a hearing aid before six months and assessed by teachers for speech intelligibility. Children identified and fitted with a hearing aid earlier achieved higher scores than children fitted with hearing aids after this age. The study participants were matched for age, sex, age of onset of hearing loss, degree of deafness and schooling.

Ramkalawan and Davis[37] examined spoken language skills in 16 children at 27 to 79 months. They found that, after controlling for age, earlier intervention was associated with better linguistic skills.

The problem with all of these studies is that they are based on observational data. Such studies can result in biased measures of effectiveness. Other confounding factors may be associated with earlier intervention. For example, parental involvement and intervention may be associated with earlier use of a hearing aid. The outcomes measured are also surrogate outcomes. The outcomes which are of primary interest have not been evaluated, that is, the long-term effects of earlier intervention on social, educational and employment opportunities. However, expert opinion in this area supports early intervention.

One study, however, found that the initial benefits of early intervention on spoken language did not persist in 118 children assessed between three and nine years old.[38] In this study, however, 'early intervention' was defined as before 36 months.

There has been some concern in the past that testing and early identification of infants may interfere with the bonding process between parents and their children. There has never been any clinical evidence of this and studies of families where PCHI has been identified show that the families preferred early confirmation.[14,39,40]

There is little evidence regarding the relative effectiveness of various habilitation programs used. Because of strong physician and parental preferences, it is unlikely that it would be possible to conduct randomised controlled trials in this area.

In summary, there are theoretical reasons for believing that earlier intervention will result in reduced disability due to hearing impairment. The evidence from clinical research, however, is relatively weak.

Existing procedures

There is considerable variation in the existing procedures for diagnosis of congenital and acquired hearing impairment in Australia and it is difficult to establish current

procedures. OAEA is available in many public hospitals and several of the NICUs are testing hearing in infants prior to discharge.[1,16] In 1992, the Centre for Community Child Health and Ambulatory Paediatrics (Royal Children's Hospital, Victoria) implemented a two-stage screening protocol in neonates and infants considered at risk of hearing impairment: the Victorian Infant Hearing Screening Program. The protocol involves auditory brainstem response (ABR) testing of at-risk neonates and distraction test screening of infants considered not at-risk at seven to nine months. Infants are identified as having risk factors for hearing impairment prior to discharge from hospital at birth and are referred to the program. After discharge, health professionals at maternal and child health centres also identify and refer at-risk children. According to figures provided by the screening program, 319 children were diagnosed with sensorineural hearing loss and fitted with hearing aids in the first three years of the program, compared to 329 children in the three years to 1992. Median age at diagnosis, however, fell from 27.1 months to 24.5 months in the same period. Of the children diagnosed with PCHI, 12.4 per cent of diagnoses were made before six months, compared to 2.8 per cent in the three years immediately prior to the commencement of the program. In the first three years of the program 55 per cent of infants had been diagnosed by the age of 12 months, compared to 32 per cent in 1989–91. Amongst children thought to be at high risk of PCHI, 22.5 per cent of infants diagnosed with PCHI were diagnosed before six months, compared to 4 per cent prior to 1992. Since the inception of the program, the proportion of children in Victoria fitted with a hearing aid before the age of 11 to 23 months has been higher in Victoria than in other States (see Table 2).

Table 2 Observed versus expected fitting of hearing aids in Australia

State	Live births (1994)[1]	Expected number of PHL[a]	Hearing aids fitted in birth cohort by 11 months[2]	Hearing aids fitted in birth cohort by 23 months[2]
NSW	87,916	88 – 176	24	62
Vic	64,119	64 – 128	24	56
Qld	47,037	47 – 94	12	22
WA	24,929	25 – 50	6	15
SA	19,425	20 – 40	2	9
Other	14,957	15 – 30	4	12
Total	258,426	259 – 518	72	176

PHL = permanent hearing loss
Source: Australian Bureau of Statistics[2]
Source: Australian Hearing Services data, reported in Birtles et al[1]
Permanent hearing loss is defined as >40dB HL in the better ear. The expected number is based on a prevalence estimate of 1 to 2 per 1,000 live births (see Table 1).

In an examination of the diagnostic and management patterns of children with sensorineural hearing loss entering early intervention programs within Victoria during a 12-month period in the early 1990s, Rickards et al[41] found that the mean age of initial referral to an audiologist was 15.6 months among the 49 children studied. Confirmation of diagnosis occurred at a mean age of 18.4 months, and the fitting of hearing aids occurred at a mean age of 20.2 months. Referrals to audiologists and subsequent diagnoses were found to occur significantly earlier in children referred due to known risk factors than children referred from maternal and child health centre screening. Referral and diagnosis occurred even later in children referred due to parental concern. The authors also found that diagnostic delay decreased with increasing severity of hearing loss.

Presently, only 11 to 14 per cent of children with congenital hearing impairment are detected by the age of 12 months in New South Wales.[1] The other States show similar deficits between the expected number of children with hearing impairment and the number actually detected and fitted with hearing aids. The difference between the number of children expected to have a PCHI and the number detected is illustrated in Table 2.

It appears from these data that many children in Australia with a permanent hearing impairment are presently not being detected until later in childhood.

Comparators

When assessing a diagnostic test, the important issue is to assess the ability of the test to differentiate between those who have the disease in question and those who do not. This needs to be done against a comparison test. This is preferably a 'gold standard' test, which is able to discriminate accurately in 100 per cent of patients between those who have the disease and those who do not. Unfortunately, there is no gold standard for audiological testing in young infants. There are, however, a number of audiological tests with which OAEA can be compared. The following is a very brief description of each of these tests.

Distraction testing

The child's attention is obtained by one tester with a visual stimulus. Another tester then presents an auditory stimulus to one side of the child, and the child indicates that it has heard the stimulus by a behavioural response such as a head turn. It is typically used in infants aged 6 to 12 months, but obviously cannot be used in younger infants and has a high degree of observer variability.[42] Despite these drawbacks, it has been used as a screening test for infants aged seven to eight months in the United Kingdom.

Auditory brainstem response (ABR)

This test is based on the brainstem response to auditory stimuli as visualised by a three-lead electroencephalographic (EEG) recording of the child's brain activity. The auditory stimuli are multiple brief duration clicks. The response is a small electrical potential embedded in the ongoing EEG which can be detected by signal averaging techniques. The test requires the child to be resting quietly and is unaffected by sleep or sedation. In an active child, the EEG activity drowns out the auditory response. The test can be used to test each ear separately and to test the threshold of hearing.

ABR requires highly trained staff and is a more sophisticated technology. Testing of a sleeping newborn takes around 20 to 30 minutes, but testing for threshold levels can take longer.[1]

A further development of ABR is automated auditory brainstem response. This uses improved microprocessor control systems to increase the speed of testing and evaluation. The test appears to detect fewer false positives as a result of middle ear effusion. Most of the studies used in this evaluation have used the Algo-1 automated screener. A newer

version, Algo-2e, is not yet available in Australia and has not yet been extensively assessed.

Visual reinforcement audiometry (VRA) is a test based on rewarding a child's response to an auditory stimulus. The reward may be anything that will holds the child's interest, such as flashing lights, an illuminated toy, or even interesting pictures. The sound stimulus may be delivered by a speaker for sound field or by headphones and may consist of a pure tone or speech stimulus. The examiner watches the child very carefully for responses which may include anything from blinking to head localisation to the sound source.[43]

VRA can occasionally be used in infants as young as five months, but in some children, such as those with Down syndrome, it may not be possible to reliably test the child until after they are 10 months old. While the use of sound field may be more successful in children unable to tolerate headphones, VRA may only be testing the better ear, especially in older children who have more developed sound localisation skills.[4]

Play audiometry

In this test the examination takes the form of a game, where the child is trained to perform an activity in response to a sound stimulus. This may be anything, such as dropping a bead in a bucket or placing a quoit on a peg. The sound stimulus usually consists of tone sounds delivered at various frequencies and decibels. The requirement for the active participation of the child limits the applicability of the test to infants older than 12 months, while the examiners' relative ability to maintain the motivation of the child may greatly influence the success of the test as well as the time taken to complete it.[4,43] Play audiometry is considered appropriate for children aged one to five years.

Marketing status of the diagnostic test

The equipment used for OAEA is listed by the Therapeutic Goods Administration (TGA) and currently 69 companies are listed as manufacturing these products. No indication is specified in the TGA listing approval.

Current reimbursement arrangement

OAEA is not currently covered under existing Medicare Benefits Schedule (MBS) arrangements.

Approach to assessment

Review of literature

The methodology used in this review of the evidence of literature on the effectiveness and safety of OAEA has followed the methods outlined in the *Cochrane Collaboration Handbook*[44] and the *Guidelines for meta-analyses evaluating diagnostic tests.*[45]

Literature search

The medical literature was searched to identify relevant studies and reviews. Searches were conducted of the entire electronic databases of Medline, The Cochrane Library, Embase and Healthstar. This search identified a total of 308 articles. In addition, the reference lists of all primary studies and review articles were checked. Information was also sought via the Internet and from international technology assessment agencies.

The following search terms were used:

Primary search 1	*Primary search 2*
Otoacoustic or Oto-acoustic *(and)*	Otoacoustic or Oto-acoustic *(and)*
Diagnosis / diagnostic *(or)*	Neonatal / infant / paediatric / child(ren) *(and)*
Sensitivity and specificity *(or)*	Evaluation / comparison
False negatives / false positives *(or)*	
Prevalence *(or)*	
Predictive value	
Additional searches	
Otoacoustic or Oto-acoustic *(and)*	Otoacoustic or Oto-acoustic *(and)*
Cost / costs and cost analysis *(or)*	Diagnostic accuracy *(or)*
Economics	Diagnosis / hearing loss diagnosis *(and)*
	Newborn / infant / prematurity / childhood disease / adolescent

Inclusion / exclusion criteria

The number of studies reporting primary data on OAEA as a result of this search was 38. Studies were then assessed by two reviewers for inclusion in the review. The inclusion criteria were:

(a) a primary study comparing OAEA with another form of audiology; and

(b) the study reported at least one of the outcomes of sensitivity, specificity, positive predictive values (PPV) or negative predictive values (NPV) or time taken for examination, or the data necessary for calculating these outcomes.

Studies were excluded if at least one of the above outcomes was not reported or if there was incomplete reporting of the data necessary for calculating these outcomes. In addition, it was not possible to obtain data from a number of studies because they were not available in English. Of the reviews, 26 were excluded for the reasons outlined in Table 3.

A list of included and excluded studies is at Appendix B.

Table 3 Reasons for exclusion of studies identified in search

Reason for exclusion	Number of studies
Incomplete reporting of data or outcomes listed above not reported	17
Data included in another study	2
English language version of study not available	7

Extraction of data

Twelve studies were included in the final analysis. Data were extracted independently by two reviewers. Any differences found in the data extracted were discussed or referred to a third reviewer.

Assessment of quality

Each of the studies included in this review was assessed for quality using the following criteria:

- The study examined a consecutive series or a random selection of a consecutive series of patients.
- All participants in the study received both OAEA and a comparator test.
- The results for each test were interpreted without knowledge of the results of the other test.

These criteria are based on recommendations for assessing the scientific validity of estimates of diagnostic accuracy as outlined in the *Guidelines for meta-analyses evaluating diagnostic tests.*[45]

Each study was assessed as clearly adequate, inadequate or unclear for each criterion. No studies in the review were clearly adequate for all criteria.

Expert advice

A supporting committee including members with expertise in relation to hearing impairment was convened to assess the evidence on this procedure. In selecting members for supporting committees, MSAC's practice is to approach appropriate medical colleges, associations or specialist societies for nominees. Membership of the supporting committee is shown at Appendix C.

Results of assessment

Is it safe?

OAEA is a non-invasive test. No adverse outcomes were reported in any of the trials included in the review. Risks associated with the test are the consequences of false positive or false negative test results.

In the one study where it was reported, parental acceptance of the test was high.[16]

Is it effective?

The ideal method for assessing the effectiveness of a diagnostic test would be a randomised controlled trial examining outcomes of importance to patients, such as quality of life, in those who have had the test compared with those who have not had the test. No trial of this sort was available, and as has been noted above, it is not feasible to conduct such studies. Because of this, the appropriate method of assessment is to determine how valid and reliable the test is for differentiating those patients with the disease (in this case permanent hearing impairment of greater than 40dB) from those patients without the disease. This can then be combined with knowledge of how diagnosis impacts on the outcome of the disease.

The accuracy of a diagnostic test is measured primarily by its sensitivity and specificity. Sensitivity is the probability that a patient with a hearing impairment will have a positive test result, which in this context is a 'fail' result. The specificity is the probability of a negative test in those without hearing impairment. The formulae for calculating these characteristics of the test are:

$$\text{Sensitivity} = \frac{\text{true positive results}}{\text{true positive} + \text{false negative results}}$$

$$\text{Specificity} = \frac{\text{true negative results}}{\text{true negative} + \text{false positive results}}$$

Ideally, the sensitivity and specificity would be calculated by comparison with a 'gold standard' test, that is, one with a sensitivity and a specificity as close to 100 per cent as possible. No such gold standard exists, particularly for testing young infants. The studies in this review compared OAEA with either ABR (eight studies) or other forms of audiological testing (five studies). One study used both ABR in the neonatal period and distraction testing at eight months (12 studies in total). The details of the studies included in the review are listed in Table 4.

Another measure of the diagnostic usefulness of a test is its PPV and NPV. The PPV is the proportion of patients who have a 'positive' result, that is, those who failed the OAEA, who turn out to have hearing impairment. The NPV is the proportion of patients who have a 'negative' result, that is, those who pass the OAEA, who are confirmed to not have a hearing impairment. The formulae are:

$$\text{PPV} = \frac{\text{true positive results}}{\text{true positive} + \text{false positive results}}$$

$$\text{NPV} = \frac{\text{true negative results}}{\text{true negative} + \text{false negative results}}$$

Table 4 Studies included in the review

Author(s)	Location	Population	Quality*	Subjects	Reference test
Beppu et al, 1997[46]	Nagoya City, Japan	47 children with suspected hearing deficit as outpatients of a health care centre	a: U b: A c: U	Mean age 3 years 3 months. Males:Females = 32:15	Play audiometry (Peep show test)
Doyle et al, 1997[47]	California, USA	200 neonates as hospital inpatients	a: A b: A c: I	Mean age 24 hours. Males:Females = 101:99	Automated ABR
El-Refaie et al, 1996[48]	Manchester, UK	20 neonates in a NICU	a: U b: A c: A	Mean age 12.2 days.	ABR
François et al, 1997[17]	Paris, France	39 infant inpatients recovering from meningitis	a: A b: A c: U	Mean age 13.8 months. Males:Females = 11:18	VRA
Gill et al, 1998[16]	Newcastle, NSW Australia	144 very low birth weight infants prior to discharge from a NICU	a: A b: A c: U	Mean gestational age 36 weeks, mean postnatal age 6.8 weeks	VRA
Guo and Yao, 1996[49]	Singapore	132 high risk infant hospital inpatients	a: A b: A c: U	Preterm neonates to 7 months	ABR
Jacobson and Jacobson, 1994[50]	Norfolk, USA	119 infant well babies and high risk infants	a: A b: A c: U	Postconception age: 33–41 weeks. Males:Females = 70:49	ABR
Meredith et al, 1994[51]	Cardiff, Wales	516 neonates in a NICU	a: A b: A c: I	Mean age 18.7 days	Distraction
Plinkert et al, 1990[52]	Tübingen, Germany	53 high risk infants (setting not described)	a: U b: A c: U	Age range 11 days to 7 months	ABR
Salamy et al, 1996[53]	San Francisco, California, USA	95 infants in recovery or prior to discharge from a NICU	a: A b: A c: I	Mean gestational age 33.6 weeks, mean postnatal age 4.5 weeks. Males:Females = 53:42	ABR
Stevens et al, 1990[54]	Sheffield, UK	723 infants in a NICU	a: A b: A c: I	Mean gestational age 34 weeks. Followed up 8 months later	**a** ABR and **b** Distraction (at 8-month follow-up)
Stevens et al, 1987[55]	Sheffield, UK	33 well neonates and 112 inpatients of SCBU (complete data for SCBU infants only)	a: A b: A c: U	Mean postconception age for SCBU infants 36.8 weeks. Males:Females = 62:50	ABR

SCBU = special care baby unit; ABR = auditory brainstem response; VRA = visual reinforcement audiometry

* The criteria used to establish quality were:

a. A consecutive series of patients or a random sample of a consecutive series.

b. All the patients in the series had both the reference and the OAEA tests.

c. The results of both tests were interpreted blinded to the results of the other test.

Each test was scored as:

A = adequate; I = inadequate; U = unclear

Results

For studies using ABR as the comparator, OAEA sensitivity ranged from 50 to 100 per cent, and specificity from 52 to 95 per cent. False positive rates were 0 to 50 per cent and false negative rates were 5 to 48 per cent (see Table 5). For studies using other comparators, such as VRA and distraction, OAEA sensitivity ranged from 39 to 94 per cent, and specificity from 68 to 94 per cent. False positive rates were 6 to 45 per cent and false negative rates were 6 to 32 per cent (see Table 6).

Table 5 Sensitivity and specificity of OAEA compared with ABR

Author(s)	Reference test	Sensitivity	Specificity	False positive rate (1 – sensitivity)	False negative rate (1 – specificity)	Prevalence
Doyle et al, 1997[47]	Automated ABR	50%	82%	50%	18%	12%
El-Rafaie et al, 1996[48]	ABR	NR	53%	NR	47%	NR
Guo and Yao, 1996[49]	ABR	90%	95%	10%	5%	8%
Jacobson and Jacobson, 1994[50]	ABR	50%	52%	50%	48%	4%
Plinkert et al, 1990[52]	ABR	90%	91%	10%	9%	22%
Salamy et al, 1996[53]	ABR	100%	67%	0%	33%	5%
a Stevens et al, 1990[54]	ABR	93%	84%	7%	16%	4%

NR = not reported/extractable; ABR = auditory brainstem response

Table 6 Sensitivity and specificity of OAEA compared with other reference tests

Author(s)	Reference test	Sensitivity	Specificity	False positive rate (1 – sensitivity)	False negative rate (1 – specificity)	Prevalence
Beppu et al, 1997[46]	Play audiometry	94%	68%	6%	32%	47%
François et al, 1997[17]	VRA	90%	94%	10%	6%	14%
Gill et al, 1998[16]	VRA	86%	87%	14%	13%	16%
Meredith et al, 1994[51]	Distraction	39%	79%	61%	21%	11%
b Stevens et al, 1990[54]	Distraction (at 8-month follow-up)	55%	82%	45%	18%	6%

NR= not reported/extractable; VRA = visual reinforcement audiometry

PPVs and NPVs depend on the prevalence of the condition being tested for in the population. They are, however, a guide to the usefulness of the test in the clinical setting. The proportion of patients who will require further testing but who do not have a hearing impairment can be estimated as 1–PPV. The proportion of patients who have a negative test result, but who will later be found to have the condition, is estimated from 1–NPV (see Tables 7 and 8).

Table 7 Yield of OAEA compared with ABR

Author(s)	Population	PPV	NPV	Prevalence
Doyle et al, 199747	Neonatal hospital inpatients	27%	93%	12%
El-Rafaie et al, 199648	NICU	NR	NR	NR
Guo and Yao, 199649	High risk neonatal hospital inpatients	59%	99%	8%
Jacobson and Jacobson, 199450	Well babies and high risk infants combined	4%	97%	4%
Plinkert et al, 199052	High risk infants (setting not described)	73%	97%	22%
Salamy et al, 199653	Infants in recovery and NICU	13%	100%	5%
a Stevens et al, 199054	NICU	20%	99%	4%
Stevens et al, 198755	SCBU	63%	95%	16%

NR = not reported/extractable; SCBU = special care baby unit; NICU = neonatal intensive care unit; PPV = positive predictive value; NPV = negative predicitive value

Table 8 Yield of OAEA compared with other reference tests

Author(s)	Reference test	Population	PPV	NPV	Prevalence
Beppu et al, 199746	Play audiometry	Referrals to hospital hearing clinic	72%	93%	47%
François et al, 199717	VRA	Infant meningitis patients	69%	98%	14%
Gill et al, 199816	VRA	NICU	57%	97%	16%
Meredith et al, 199451	Distraction	NICU	19%	92%	11%
b Stevens et al, 199054	Distraction (at 8-month follow-up)	NICU	16%	97%	6%

NR = not reported/extractable; SCBU = special care baby unit; NICU = neonatal intensive care unit; PPV = positive predictive value; VRA = visual reinforcement audiometry; NPV = negative predicitive value

The other variable of interest reported in several of the studies was the time taken to perform the test. In all studies where it was reported, OAEA took less time to perform than ABR. Time estimates for OAEA ranged from 3 to 17.5 minutes and ABR ranged from 12 to 30 minutes (see Table 9). The time reported varied considerably between studies because some measured only the time to perform the test and others reported the total time, including time to settle the child, explain the test and results to parents and record results. Most established programs test between two and four children per hour.[5]

Table 9 Time taken to complete the test

Authors	Reference test	Time taken for reference test	Time taken for OAEA
Doyle et al, 1997[47]	ABR	24 minutes	13 minutes
El-Rafaie et al, 1996[48]	ABR	NR	NR
Guo and Yao, 1996[49]	ABR	30 minutes	3 minutes
Jacobson and Jacobson, 1994[50]	ABR	26.3 minutes	16.6 minutes
Plinkert et al, 1990[52]	ABR	NR	1.59 minutes
Salamy et al, 1996[53]	ABR	12 minutes	11 minutes
a Stevens et al, 1990[54]	ABR	21 minutes	12.1 minutes
Stevens et al, 1987[55]	ABR	25.6 minutes	17.5 minutes
Meredith et al, 1994[51]	Distraction	NR	10 minutes
b Stevens et al, 1990[54]	Distraction (at 8-month follow-up)	NR	12.1 minutes

NR = not reported/extractable; ABR = auditory brainstem response; OAEA = Oto-acoustic emission audiometry

Because of heterogeneity between the studies, it was not possible to combine the results in any form of meta-analysis. One of the few consistent findings across studies, however, was that NPVs were almost exclusively in the upper 90 per cent in both comparator groups.

Discussion

Large variations in results may be partly explained by non-standardised test conditions and differences in the age group studied. There are also many different versions of the pass/refer criteria being used. In addition, while some tests were performed within sound controlled environments, others were done 'cribside' in potentially noisy conditions. Indeed, compensation for environmental noise led to modifications in pass and fail criteria for OAEA in some studies.

While the methodology of OAEA testing was fully described in all the studies reviewed, full descriptions of the reference test were rarely reported, especially for those studies using references other than ABR. As is known, the method of sound delivery in VRA (headphones or sound field) may result in false negatives if only the better ear is tested. Headphones are not commonly used with young infants and hence unilateral loss may be missed.

In one of the larger studies, Stevens et al[54] reported that among 723 NICU infants, OAEA achieved a sensitivity of 93 per cent and a specificity of 84 per cent when compared with ABR. At 8-month follow-up using distraction audiology, sensitivity of the initial OAEA test was reduced to 55 per cent while specificity remained relatively stable at 83 per cent. The authors concluded that OAEA is able to identify most infants who will fail ABR.

The approach taken in the current evaluation was to restrict the review to studies of the highest available quality which were directly applicable to the problem of interest, that is, the use of OAEA in groups at risk of hearing impairment. Unfortunately, the available literature must be regarded as less than rigorous. For example, sample selection is often inadequately described or not described at all. An apparently almost universal failure to evaluate OAEA independently of the reference test (blind), leaving open the possibility of bias, further undermines confidence in the findings. Indeed, in an effort to measure the possible effect of such bias, Doyle et al[47] alternated the order of testing between OAEA and automated ABR and found that pass rates for OAEA were significantly higher in the group tested first with ABR. Ideally, data from individual studies would be pooled in an overall estimate of accuracy, a meta-analysis. However, because of the substantial differences between the available studies, it is impossible to perform such an analysis.

Borderline results/retesting

In a study of the OAEA in infants, approximately 1 per cent could not be settled and 2 per cent had borderline results.[7] The proportion of tests where a recording is not possible increases with the average age of testing. In a study of survivors of bacterial meningitis, aged 6 to 24 months, 5.1 per cent of tests were of inadequate quality.[17]

In the study by Gill[16] on neonates in a NICU, 34 per cent of 144 infants required retesting. This high rate of indeterminate results may be partly due to difficulty in testing infants still on nasal oxygen and also due to transient middle ear effusion in very young neonates.

Factors affecting the effectiveness of the test

The number of false positive test results will increase when testing is carried out in neonates less than 48 hours old because of debris in the neonatal ear and possibly other mechanisms such as oxygenation of the outer hair cells.[56] As has been reported,[47] pass rates for OAEA and, to a lesser extent, ABR may increase as a function of infant age, even within a matter of hours.

There will also be a higher false positive rate if testing is carried out in a population with a high incidence of otitis media. The failure rate in a study of children tested at 12 weeks old was 9.6 per cent, principally because of the presence of otitis media with effusion.[57] The false positive rate in children with middle ear effusion can be reduced by the use of pneumatic otoscopy and impedance audiometry in children more than six months old and multi-frequency tympanometry in children less than six months old.

Where a condition occurs only rarely, such as PCHI, even a test with a high sensitivity and specificity will have a low PPV. This means that a large number of the children who are referred for further testing will be found not to have a hearing impairment.

What are the economic considerations?

The main focus of this report has been a systematic review of the effectiveness of OAEA as a diagnostic test. It was not possible within the scope of this review to do a full economic evaluation of the technology. However, some comments can be made on the costs and consequences of the use of this technology.

Costs

The equipment required for OAEA consists of the probe and a dedicated computer. The cost is between $5,900 and $20,000. Newer versions of the technology are relatively cheaper and more portable. A study of low birth weight neonates at John Hunter Children's Hospital in Newcastle conducted between April 1994 and March 1996 estimated the cost per test in a public hospital NICU to be $5 to $10 per test.[16]

Some of the studies included in this review reported details of the costs involved in testing. Often these were in the form of cost per test performed and cost per case of hearing impairment detected. Quoted costs included the cost of follow-up testing in those children who failed the OAEA, for example, testing by ABR and confirmatory diagnostic assessment. A summary of these estimates is shown in Table 10. Cost appears to be primarily a function of the length of time taken to do the test and the hourly rate of pay of the tester.

The Working Party on 'The Early Identification of Hearing Impairment in Children in NSW'[1] also estimated costs for various combinations of screening and diagnostic testing

(Table 11). As may be expected, the more targeted the testing program, the higher the cost per child tested, because of the need to carry out confirmatory testing in a higher proportion of patients. Conversely, the cost per case of hearing impairment detected and the overall cost of the program will be lower. The more targeted the program, however, the greater the proportion of children with hearing impairment who will remain undetected. The New South Wales report concluded that the most cost-effective form of screening, in terms of cost per case detected, appears to be the combination of OAE as a first line and ABR as a second line screening tool.

To examine the financial impact and consequences of testing, we have developed a model considering alternative diagnostic strategies in a hypothetical cohort of 1,000 children. In each case the use of a screening ABR test is compared with a two-stage use of OAEA followed by a screening ABR in those children found to be positive on OAEA. Children found positive by either strategy would then require full audiological testing. The sensitivity of ABR is assumed to be 94 per cent and specificity 89 per cent.[11] The cost of ABR is assumed to be $145 per test and the cost of OAEA to be $25 per test. In the first model, the costs and consequences of testing in a population with a prevalence of PCHI of 4 per cent using both diagnostic strategies are compared. Model 1A uses low estimates of sensitivity and specificity.[50] Model 1B uses high estimates.[53] The second model assumes a prevalence of PCHI of 16 per cent, again varying the estimated sensitivity and specificity of the OAEA. The prevalence estimates were chosen as the supporting committee assessed that they reflect the prevalence rates for children in a specialist referral clinic.

Consequences of testing

There are a number of possible positive consequences from the use of OAEA:

(a) There is some evidence that the use of OAEA has resulted in lowering the average age of diagnosis of PCHI.[14] Prior to distraction test screening, the average age of diagnosis in the United Kingdom was 18 months. Presently in the United Kingdom, the median age for referral for PCHI is 10.4 months, for confirmation of diagnosis 17.1 months and for fitting a hearing aid 26.3 months.[10] There remains considerable geographic variation. The distribution was also highly skewed to the right, that is, there was a significant proportion of children not detected until quite late in childhood. In the United States, the typical age of diagnosis is 2.5 years. The average age for fitting a hearing aid for children with sensorineural hearing loss in Australia is 2.5 years.[41] As discussed above, there is some evidence that earlier intervention could be beneficial to the overall development of the child, particularly in language and communication skills.

(b) For children diagnosed with a permanent hearing impairment, the majority will be fitted with a hearing aid and referred to a specialist educator. There is uncertainty, however, about the best method of intervention to minimise the short-term and long-term impact of permanent hearing impairment.[5] The effectiveness of intervention in mild cases of hearing impairment has not been evaluated.

(c) In a small minority of patients, usually regarded as having a hearing loss greater than 100dB, the level of hearing impairment is so profound that amplification is not applicable. In these children, a cochlea implant may be considered. Ideally this should be done before the age of two years.[58] In the United Kingdom the cost of

implantation and maintenance over 10 years is estimated to be £50,000. Assuming a prevalence rate of 0.02 to 0.04 per cent, this level of hearing loss occurs in approximately 50 to 100 children per year in Australia. A study of the cochlea implantation program in South Australia reported on 17 children who were provided with cochlea implants in the first two years of the program. Only one of these children was less than two years old.[59] There is some evidence that cochlear implantation leads to a shift in educational placements in favour of mainstreaming with support.[60]

(d) For the parents of children who have been assessed by OAEA and who have a 'pass' result, there is the reassurance that their child's hearing is highly likely to be normal.

(e) For parents with a child with PCHI, earlier diagnosis may allow greater opportunity for genetic counselling.

As with any test, there are possible risks associated with testing:

(a) With inpatients who have been tested and have passed OAEA, but who have an actual hearing impairment (false negatives), the test may falsely reassure parents and professionals about the child's hearing. This would affect approximately 1 to 8 per cent of children tested, that is, 1–NPV.

(b) A proportion of children initially fail the OAEA test but are subsequently found not to have a hearing impairment. These false positive results may cause either temporary or permanent anxiety in the child's parents regarding their child's hearing. The false positive rate is high in the first 48 hours of life, falls, and then rises again with the increasing prevalence of otitis media with effusion.

(c) With use of the test in children to detect those with mild hearing loss, particularly temporary conductive hearing loss due to otitis media with effusion, the effect of such hearing loss is not yet known and the effectiveness of intervention in such cases is not yet proven.[5,18] If identified by this test, such children may be then subject to medical or surgical interventions with the associated risks of adverse effects.

Implications for current resources

The use of OAEA may reduce the demand for some other forms of testing such as distraction testing.

Those children who fail the initial OAEA will require confirmatory audiological testing. In the studies of universal screening, approximately 3 per cent of children required follow-up ABR.[7,14] If all children in Australia were tested in the first 12 months of life (approximately 253,000 children in 1997), this would mean that 7,590 children would require ABR. This could be thought of as a maximum demand for ABR as a result of OAEA. Those children who then fail ABR would need a full diagnostic audiological assessment.

Children diagnosed with permanent hearing impairment need access to habilitation and specialist resources as soon as possible. By allowing earlier identification of hearing impairment, it is likely that the use of OAEA will result in an increased demand for hearing aids and cochlea implants in younger infants. Communication and educational

support is also likely to be required at a younger age. This may be offset by reduced support required by children with hearing impairment after school entry and may also result in less demand for specialised education after school entry.

Other considerations

Access to technology

The application sought that the technology be available only at the request of a specialist paediatrician or oto-rhino-laryngologist. The benefit of this approach is that demand for the technology will be more limited and inappropriate use, such as testing in the presence of middle ear effusion, is likely to be less. The disadvantage of this approach is that access to the technology will also be limited. In the situation where an infant is determined by a primary carer or other practitioner to be at high risk of hearing impairment, the need for a specialist referral may delay the diagnosis. This delay may result in increased parental anxiety and, as has been discussed in this report, there is evidence that earlier diagnosis and intervention may result in improved outcomes. Children in rural and indigenous communities are particularly likely to be disadvantaged by the need for specialist referral before testing.

Further research and development

There needs to be further study on the prevalence and identification of children with permanent hearing impairment in Australia. If OAEA is funded, it would also be recommended that there should be monitoring of its effectiveness, principally the age of diagnosis and the commencement of habilitation. Other areas which require further research are:

- the effectiveness of early intervention;
- the prevalence and consequences of mild or unilateral PCHI; and
- the prevalence and consequences of temporary conductive hearing loss (this last area is presently being investigated by a United Kingdom Medicare Research Council funded randomised controlled trial).

Table 10 Comparative costs of screening programs for universal and specific paediatric populations

Study	Year of trial	Location	Study Population	Prevalence of PCHI	Test used	Cost/child tested (a)	Cost/child detected with PCHI (a)
Maxon, 1995	1993	Rhode Island, USA	Screening of neonatal population (14% from NICU)	1.1%	OAEA ABR follow-up	$41.30	$6,940.44
Friedland et al, 1996	1993	Mt Sinai Hospital, USA	Screening of high risk register (420 of approx 16,000 births)	0.057% (1.27% of those tested)	ABR	$951.18	$67,023.31
Watkin, 1996a	1992-5	Whipps Cross Hospital, UK	Universal screening of neonates	*0.2%	OAEA ABR follow-up	$24.87	$12,436.69
Francois et al, 1997[5]	1989-95	Hopital Robert Debre, France	Survivors of bacterial meningitis	2.6%*	OAEA & VRA ABR follow-up	$128.41	$1,252.39
Davis et al, 1997	1997	Estimate (b)	Universal screening of neonates	0.07-0.14%	OAEA	$35.03	$25,127 – 50,000
Davis et al, 1997	1997	Estimate (b)	Targeted screening of neonates	0.5-0.75%	OAEA	$179.19	$23,858 – 35,787

a) Cost in $A based on exchange rates in March 1999.
b) The estimates of cost in this report were based on an estimate of the cost per test and any follow-up tests combined with estimates of prevalence.

Table 11 Estimated staff and equipment costs for screening program

Factors				**OAE Screen**	**ABR Screen**	Distraction Screen	High Risk Register	**Diagnostic**
Time per child								
Time per child-screener				22*	40	20		
Time of coordinator - mins				5	5	5	5	120
Time of administrator - mins				5	5	5	5	7
Staff	Salary	Plus Oncosts	Hours					
Salary of screener	$30,000	$36,000	1400	$9.43	$17.14	$8.57	0	0
Salary of coordinator	$45,000	$54,000	1400	$3.21	$3.21	$3.21	$3.21	$77.14
Salary of administration staff	$30,000	$36,000	1400	$2.14	$2.14	$2.14	$2.14	$3.00
Equipment		Equipment	Amortisedover 5year					
Equipment costs-OAE		$17,000	$3,400	$1.13^	$0.00			$80
Equipment cost-ABR		$10,000	$2,000		$0.67*			
Equipment cost-Warble		$2,000	$800			$0.13		
Maintenance of equipment		$250	$50	$0.08	$0.08	$0.02		$1
Computers/printer		$2,000	$400	$0.13	$0.13	$0.13	$0.13	$0.13
Stationary, phones, etc				$1.00	$1.00	$1.00	$1.00	$1.00
Total				**$17.14**	**$24.38**	**$15.21**	**$6.49**	**$162.28**

*based on Rhode Island and Whipps Cross programs
^based on 3,000 children per year
Source: Birtles et al, 1998 p 63

Model 1A

Prevalence of hearing impairment 4%

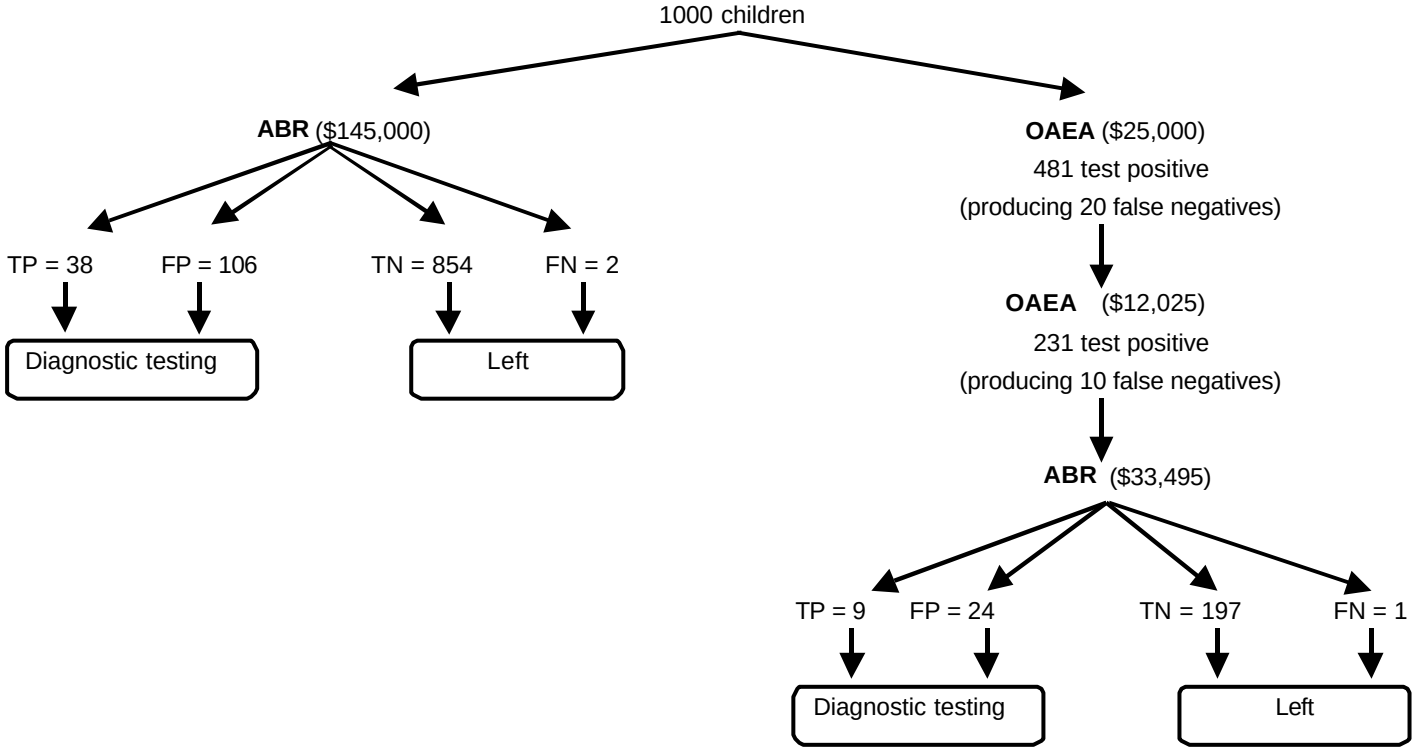
Assumptions:

ABR sensitivity 94%, specificity 89% (White et al 1993)

OAEA sensitivity 50%, specificity 52% (Jacobson and Jacobson 1994)

ABR cost of approximately $145 per test

OAEA cost of approximately $25 per test



Comments:

TP = true positive; FP = false positive; TN = true negative; FN = false negative

Single ABR screening would produce 2 false negatives and 106 false positives

2-stage screening with OAEA followed by ABR would produce 31false negatives and 24 false positives

Cost: (excluding diagnostic testing)

ABR only = $145,000

2-stage screening with OAEA followed by ABR = $70,520

Model 1B

Prevalence of hearing impairment 4%

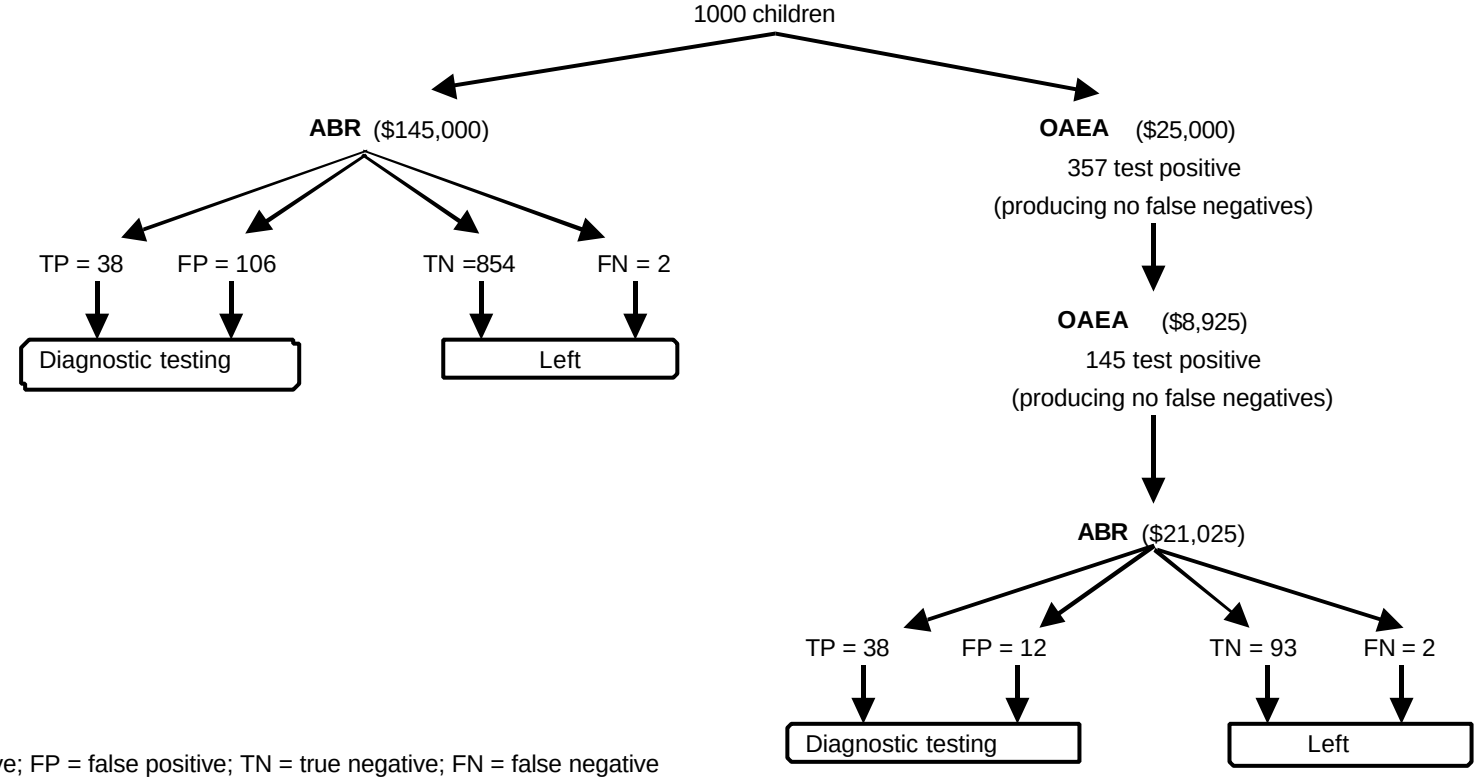
Assumptions:

ABR sensitivity 94%, specificity 89% (White et al 1993)

OAEA sensitivity 100%, specificity 67% (Salamy et al 1996)

ABR cost of approximately $145 per test

OAEA cost of approximately $25 per test



Comments:

TP = true positive; FP = false positive; TN = true negative; FN = false negative

Single ABR screening would produce 2 false negatives and 106 false positives

2-stage screening with OAEA followed by ABR would produce 2 false negatives and 12 false positives

Cost: (excluding diagnostic screening)

ABR only = $145,000

2-stage screening with OAEA followed by ABR = $54,950

Model 2A

Prevalence of hearing impairment 16%

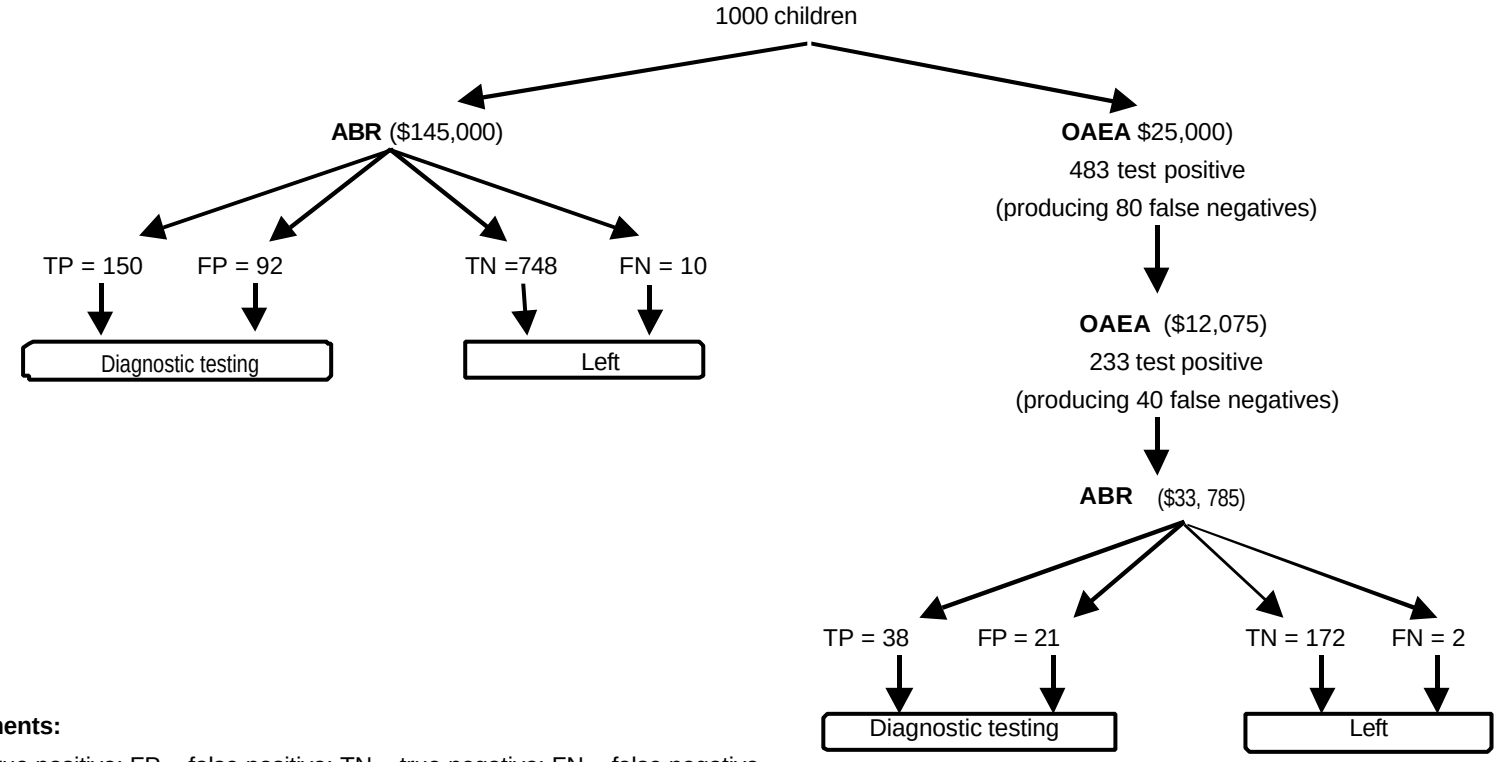
Assumptions:

ABR sensitivity 94%, specificity 89% (White et al 1993)

OAEA sensitivity 50%, specificity 52% (Jacobson and Jacobson 1994)

ABR cost of approximately $145 per test

OAEA cost of approximately $25 per test



Comments:

TP = true positive; FP = false positive; TN = true negative; FN = false negative

Single ABR screening would produce 10 false negatives and 92 false positives

2-stage screening with OAEA followed by ABR would produce122 false negatives and 21 false positives

Cost: (excluding diagnostic screening)

ABR only = $145,000

2-stage screening with OAEA followed by ABR = $70, 860

Model 2B

Prevalence of hearing impairment 16%

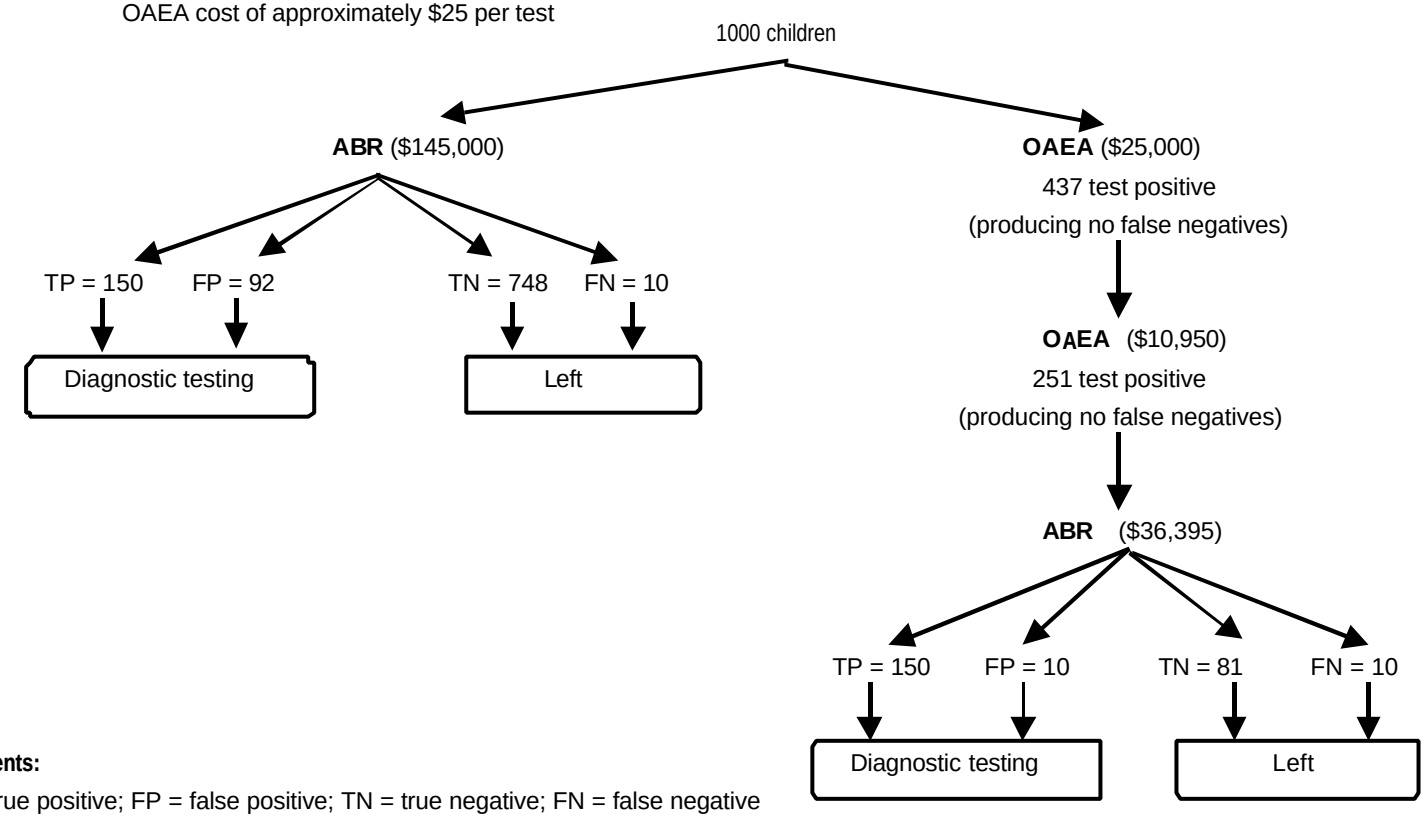
Assumptions :

ABR sensitivity 94%, specificity 89% (White et al 1993)

OAEA sensitivity 100%, specificity 67% (Salamy et al 1996)

ABR cost of approximately $145 per test

OAEA cost of approximately $25 per test



Comments:

TP = true positive; FP = false positive; TN = true negative; FN = false negative

Single ABR screening would produce 10 false negatives and 92 false positives

2-stage screening with OAEA followed by ABR would produce10 false negatives and 10 false positives

Cost: (excluding diagnostic screening)

ABR only = $145,000

2-stage screening with OAEA followed by ABR = $72,245

Conclusions

Safety

OAEA is a non-invasive test which requires only a short exposure and testing time. There are no safety concerns with this test.

Effectiveness

A systematic review of studies on the sensitivity and specificity of OAEA shows that it is reasonably sensitive and specific when compared with other forms of audiology. Because of the high prevalence of otitis media with effusion in the population to be tested, there can be a high false positive rate resulting in a low PPV. That is, a high proportion of children who are tested and 'fail' OAEA will be found on further testing not to have the condition of interest, which is a permanent hearing impairment of greater than 40dB. OAEA will also have a low PPV because of the relative rarity of the condition being tested for. On the other hand, the test has a high NPV, that is, it is able to accurately predict those children who do not have the disease. It is also more convenient and quicker than other forms of audiometry. OAEA is, therefore, useful as a first line test to identify children who may have permanent hearing loss, particularly in pre-lingual children where other methods of testing may not be possible. Children who fail OAEA will require further testing with other diagnostic audiological methods.

While there is strong theoretical evidence that earlier intervention in hearing impairment may result in better outcomes for infants with permanent hearing impairment, there are no randomised controlled trials available to support this and it is considered not feasible to conduct such studies.

Cost-effectiveness

Cost has not been fully evaluated but it appears that OAEA compares favourably with other forms of audiological testing, particularly in infants. The equipment and resources required for testing are relatively low cost. The use of this technology appears to allow earlier identification of hearing impairment at less cost than alternative forms of testing.

Other considerations

With regard to access to testing, specialist referral may limit inappropriate use of the test but may decrease access to testing.

Recommendations

It is recommended that on the strength of evidence pertaining to OAEA, public funding should be supported for this procedure for the detection of PCHI in groups of children at high risk due to the following factors:

- admission to a NICU
- family history of hearing impairment
- perinatal infection (either suspected or confirmed)
- birthweight <1.5kg
- craniofacial deformity
- birth asphyxia
- chromosomal abnormality, including Down syndrome
- exchange transfusion

In addition, it is recommended that OAEA be preceded by a specialist assessment to exclude middle ear pathology.

Appendix A MSAC terms of reference and membership

The terms of reference of MSAC are to advise the Commonwealth Minister for Health and Aged Care on:

- the strength of evidence pertaining to new and emerging medical technologies and procedures in relation to their safety, effectiveness and cost-effectiveness and under what circumstances public funding should be supported;
- which new medical technologies and procedures should be funded on an interim basis to allow data to be assembled to determine their safety, effectiveness and cost-effectiveness; and
- references related either to new and/or existing medical technologies and procedures.

The membership of MSAC comprises a mix of clinical expertise covering pathology, nuclear medicine, surgery, specialist medicine and general practice, plus clinical epidemiology and clinical trials, health economics, consumers, and health administration and planning:

Member	Expertise
Professor David Weedon (Chair)	pathology
Ms Hilda Bastian	consumer health issues
Dr Ross Blair	vascular surgery (New Zealand)
Mr Stephen Blamey	general surgery
Dr Paul Hemming	general practice
Dr Terri Jackson	health economics
Professor Brendon Kearney	health administration and planning
Mr Alan Keith	Assistant Secretary, Diagnostics and Technology Branch, Commonwealth Department of Health and Aged Care (from 3 May 1999)
Dr Richard King	gastroenterology
Dr Michael Kitchener	nuclear medicine
Professor Peter Phelan	paediatrics
Dr David Robinson	plastic surgery
Ms Penny Rogers	Assistant Secretary, Diagnostics and Technology Branch, Commonwealth Department of Health and Aged Care (until 3 May 1999)
Associate Professor John Simes	clinical epidemiology and clinical trials
Dr Bryant Stokes	neurological surgery, representing the Australian Health Ministers' Advisory Council (from 1 January 1999)
Dr Doris Zonta	population health, representing the Australian Health Ministers' Advisory Council (until 31 December 1998)

Appendix B Included and excluded studies

Included Studies

Beppu R, Hattori T, Yanagita N. Comparison of OAEA with play audiometry for screening hearing problems in children. Auris Nasus Larynx 1997; 24: 367-71.

Doyle KJ, Burggraaff B, Fujikawa S et al. Newborn hearing screening by otoacoustic emissions and automated auditory brainstem response. International Journal of Pediatric Otorhinolaryngology 1997; 41: 111-9.

El-Refaie A, Parker DJ, Bamford JM. Otoacoustic emission versus ABR screening: the effect of external and middle ear abnormalities in a group of SCBU neonates. British Journal of Audiology 1996;30: 3-8.

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Appendix C Supporting committee

Supporting committee for MSAC application 1002

Dr John Primrose (Chair) MB BS (Hons), FRACR Senior Medical Adviser, Health Access and Financing Division, Commonwealth Department of Health and Aged Care	Medical Adviser to MSAC
Dr Harvey Coates MB BS, MS (OTOL), DABO, FRACS, FRCS(C), FACS Senior Paediatric ENT Surgeon, Princess Margaret Hospital for Children, Perth	nominee of the Royal Australasian College of Physicians
Mrs Jean Feder Parent of a severe/profound hearing impaired son, Secretary of Parents of Hearing Impaired SA Inc. 16 years, Board Member Deafness Forum representing Parents, Board Member Townsend House for Blind and Deaf Children	nominee of the Deafness Forum
Professor Bill Gibson MD, MB MS, FRCS, FRACS Professor of Otolaryngology, University of Sydney, Director of Children's Cochlea Implant Centre (NSW)	nominee of the Royal Australasian College of Surgeons
Mr Roger Lovegrove BA, MA (Audiology), M Aud Soc Aust, CC Senior Audiologist, Hearing Service Support, Australian Hearing Services	nominee of the Australian Hearing Services
Dr David Starte MB BS, MRCP (UK), FRACP Service Director, Chatswood Assessment Centre, Royal North Shore Hospital	nominee of the Royal Australasian College of Physicians

Abbreviations

ABR	Auditory brainstem response
dB HL	decibel measure of hearing level
DPOAE	Distortion product oto-acoustic emission audiometry
MSAC	Medicare Services Advisory Committee
NICU	Neonatal intensive care unit
NPV	Negative predictive value
OAE	Oto-acoustic emission
OAEA	Oto-acoustic emission audiometry
PCHI	Permanent congenital hearing impairment
PPV	Positive predictive value
SCBU	Special care baby unit
TGA	Therapeutic Goods Administration
TOAEA	Transient evoked oto-acoustic emission audiometry
VRA	Visual reinforcement audiometry

Glossary

Habilitation	Providing people with an ability they never had, as opposed to rehabilitation which is the restoration of an ability
Hearing impairment	This report recognises three grades of impairment: moderate, severe and profound. Moderate hearing impairment is defined as 40dB or greater in the better ear.

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