Review of newborn hearing screening regimes and associated screening devices

for the

National Screening Unit
Ministry of Health
New Zealand

young futures
realising possibilities

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Review team

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Revisions to the *Review of Newborn Hearing Screening and Associated Screening Devices*

This *Review of Newborn Hearing Screening and Associated Screening Devices* was first released in March 2014.

Following the report's release it was identified that a number of corrections and revisions needed to be made. This is a revised version of the report that includes these corrections and revisions.

Details of the corrections and revisions are available on the New Zealand Ministry of Health website at http://www.nsu.govt.nz/health-professionals/4627.aspx

The corrections and revisions do not impact on the report's conclusions or the recommendations made by Young Futures to the New Zealand Ministry of Health.

Acknowledgements

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- The staff of the National Screening Unit (NSU) of the New Zealand Ministry of Health and the project steering group for so promptly and constructively facilitating the completion of each stage of the review.

- The many staff and stakeholders of the UNHSEIP across New Zealand who gave of their time and experiences so generously and demonstrated such commitment to the continued development of newborn hearing screening in New Zealand.

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  o British Columbia Early Hearing Program, Provincial Health Services Authority, British Columbia, Canada

  o Quebec Newborn Hearing Screening Program, Quebec, Canada

  o NHS Newborn Hearing Screening Program, England, United Kingdom

  o Kind en Gezin's Gehoorscreenings Programma, Flanders, Belgium

  o Colorado Early Hearing Detection and Intervention, Colorado, USA

  o Tasmanian Universal Newborn Hearing Screening Program, Department of Health and Human Services, Tasmania, Australia

  o Healthy Hearing, Queensland Health, Queensland, Australia

  o Victorian Infant Hearing Screening Program, Department of Health, Victoria, Australia

  o Newborn and Children's Hearing, Women's and Children's Health Network, South Australia, Australia

- Dr Carlie Driscoll, Senior Lecturer in Audiology at the University of Queensland, and Ms Gwen Carr, Programme Lead of the NHS Newborn Hearing Screening Programme for contributing their expertise to inform the review method and recommendations.

- The manufacturers and suppliers of newborn hearing screening equipment.

Limits of this work

This report is prepared specifically for the use of
the National Screening Unit of the New Zealand Ministry of Health.

Young Futures accepts no duty of care to any other person or entity.

The areas of focus in the report respond to
the specific information needs of the New Zealand Ministry of Health.
As such, some issues and concepts receive greater emphasis than others.

The report does not intend to comprehensively attend to
every issue pertinent to newborn hearing screening.

The report has been prepared for the purpose of making recommendations
regarding the optimal universal newborn hearing screening regime for
implementation by the Universal Newborn Hearing Screening and Early
Intervention Programme, within the New Zealand context.

Conflicts of interest

The authors of this work have no personal or financial conflicts of interest that
have had undue influence on either the method used to undertake the review or
the recommendations made.

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List of abbreviations

A-ABR	Automated auditory brainstem response
ASSR	Auditory steady state evoked response
ABR	Auditory brainstem response
ANSD	Auditory neuropathy spectrum disorder
AOAE	Automated otoacoustic emissions
CE chirp	Claus Elberling chirp
CMV	Cytomegalovirus
dB	Decibel
dBnHL	Decibel normalised hearing level
DHB	District Health Board
DPOAE	Distortion product otoacoustic emissions
ENT	Ear, Nose and Throat
JCIH	Joint Committee on Infant Hearing
MASSR	Multi-frequency auditory steady state evoked response
MSAC	Medical Services Advisory Committee
NHS	National Health Service
NHSP	Newborn hearing screening programme
NICU	Neonatal intensive care unit
NSU	National Screening Unit
NZ	New Zealand
PCHI	Permanent childhood hearing impairment
SCBU	Special care baby unit

TEOAE	Transient evoked otoacoustic emissions
UK	United Kingdom
UNHSEIP	Universal Newborn Hearing Screening and Early Intervention Programme
UNHSI	Universal newborn hearing screening and intervention
USA	United States of America
WAI	Wideband acoustic immittance
WHO	World Health Organization

Executive summary and recommendations

Review objective, method and context

In September 2013, the New Zealand Ministry of Health contracted Young Futures to:

> *examine best practice in newborn hearing screening regimes, including associated equipment options, to assist the National Screening Unit to determine the most appropriate screening regime for the New Zealand Universal Newborn Hearing Screening and Early Intervention Programme.*

The stimulus for the work was the *Quality improvement review of a screening event in the Universal Newborn Hearing Screening and Early Intervention Programme[1],* which recommended that:

> *The NSU must reassess the screening protocol with a view to changing to an A-ABR only protocol.*

The *Quality improvement review* was precipitated in 2012 following the National Screening Unit (NSU) being notified that two newborn hearing screeners, from two District Health Boards (DHBs), had not followed screening protocols.

This report has been formulated based on a rapid review of the literature regarding hearing screening regimes and emerging technologies; a review of nine international programmes; an exploration of the New Zealand Universal Hearing Screening and Early intervention Programme (UNHSEIP); a review of newborn hearing screening devices; and analysis of the variables which impact on regime choice.

The expertise of the review team was supplemented by independent advice provided by Dr Carlie Driscoll, Senior Lecturer in Audiology at the University of

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[1] Ministry of Health. (2012). Quality improvement review of a screening event in the Universal Newborn Hearing Screening and Early Intervention Programme. Retrieved from: http://www.nsu.govt.nz/files/ANNB/Newborn_Screening_Report_FINAL.pdf

Queensland, and Ms Gwen Carr, Programme Lead of the National Health Service, Newborn Hearing Screening Programme in England.

Literature review findings

The literature review considered current evidence regarding the application of genetic testing, multi-frequency and bandwidth automated steady state evoked response, and wide band acoustic immittance to universal newborn hearing screening. Evidence does not suggest that these technologies are viable for stand-alone use in universal newborn hearing screening programmes at this point in time.

Regimes that use automated otoacoustic emissions (AOAE) alone (e.g. AOAE/AOAE[2]) have lower sensitivity and specificity and high false positive rates and therefore a high rate of referral, compared to other regimes. This corresponds to higher costs per infant screened. AOAE-only regimes do not detect auditory neuropathy spectrum disorder (ANSD).

Regimes that use a combination of AOAE and automated auditory brainstem response (A-ABR) (e.g. AOAE/A-ABR or, AOAE, A-ABR/A-ABR) have higher sensitivity and specificity and a much lower rate of referral to audiology compared to regimes that use AOAE alone. This corresponds to lower costs per infant screened and demands on fewer families to attend follow-up screening. Regimes using a combination of AOAE and A-ABR do not detect ANSD if the infant is discharged with a pass result.

Regimes that use A-ABR alone (typically A-ABR/A-ABR) have the highest sensitivity and specificity and the lowest first screen refer rate. This corresponds to lower costs per baby screened and demands on fewer families to attend follow up screening. Regimes that use A-ABR alone can identify ANSD.

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[2] Screening regimes consist of the type of screening, the number of stages, and the number of steps involved in the screening process prior to referral to audiology for diagnostic assessment. Stages are similar to 'appointments' or occasions that a child is seen. Screening regimes typically have either one, two or three stages. The number of steps refers to the number of screens. A screening regime may have two stages, but three steps, if one of the stages involves the use of two different screens.

In this report, to notate different screening regimes a forward slash is used to mark a boundary between screening stages (A-ABR/A-ABR). The second or third stage screen only proceeds when a refer result occurs on the preceding screen. Similarly, where two screens occur within the one stage and the second screen only proceeds when a refer result occurs on the preceding screen, the notation of the two screens within the one stage is separated by a comma (DPOAE, A-ABR/A-ABR). When the regime requires that both screens are completed, regardless of the result on the first screen, the notation of the two screens is separated by a plus sign (TEOAE + A-ABR).

Claus Elberling chirp (CE chirp) ASSR/A-ABR is a validated and reliable screening tool[3, 4, 5, 6, 7, 8, 9].

Evidence from international and UNHSEIP practice

Regime choice

The international programmes studied used a range of regimes. Selection of an A-ABR/A-ABR regime was typically based on a lower first screen refer rate, capacity to identify ANSD, reduced impact of noise on screening, and multiple benefits arising from the simplicity of the regime.

Selection of a regime using a combination of AOAE and A-ABR was typically on the basis that at the time of regime selection AOAE was believed to be quicker and more cost efficient than A-ABR/A-ABR regimes.

For well babies, the UNHSEIP uses a two stage regime with AOAE (specifically, DPOAE) completed in the first stage, followed immediately by A-ABR in the same appointment if a first screen refer result occurs. A second A-ABR occurs in a second stage if required. The decision to use AOAE as a first screen was informed by the speed, cost and ease of use of AOAE. Babies who have spent 48 hours or more in neonatal intensive care (NICU) are screened using a single stage A-ABR, and other babies identified with risk factors are screened with a two stage A-ABR

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[3] Melagrana, A., Casale, S., Calevo, M.G., Tarantino, V. (2007). MB 11 BERAphone and auditory brainstem response in newborns at audiologic risk: Comparison of results. *International Journal of Pediatric Otorhinolaryngology*. *71*(8), 1175-1180.

[4] Cebulla, M., Hofmann, S., Shehata-Dieler, W. (2014). Sensitivity of ABR based newborn screening with the MB 11 BERAphone®. *International Journal of Pediatric Otorhinolaryngology*. *78*(5), 756-761.

[5] White, K., Foust, T., Winston, R., Ditty, K. (2008). A comparative study of the MB 11 BERAphone and ABAER automated auditory brainstem response newborn hearing screening equipment. Retrieved from: http://www.maico-diagnostics.com/eprise/main/_downloads/us_en/Documentation/MB11_study-final.pdf

[6] Soares, M., Nakazawa, M., Ishikawa, K., Sato, T., Honda, K. (2014). Hearing screening for Japanese children and young adults using the automated auditory brainstem response. *Auris, Nasus, Larynx*. *41*(1), 17-21.

[7] Augustine, A.M., Jana, A.K., Danda, S., Lepcha, A., Ebenezer, J., Paul, R.R., Tyagi, A., Balraj, A. (2014). Neonatal hearing screening: Experience from a tertiary care hospital in Southern India. *Indian Pediatrics.15,* 179-183.

[8] Guastini, L., Mora, R., Dellepiane, M., Santomaura, V., Mora, M., Rocca, A., Salami, A. (2010). Evaluation of an automated auditory brainstem response in a multi-stage infant hearing screening. *European Archive of Otolaryngology*. *267*(8), 1199-1205.

[9] Cobb, K.M., Stuart, A. (2014). Test-retest reliability of auditory brainstem responses to chirp stimuli in newborns. *International Journal of Audiology*. *11,* 1-7.

regime. This regime was selected after a review of literature on screening and a review of other programmes.

Screening timing and location

For the international programmes, one programme completes 96.8% of screening through a universal community based preventative health service, therefore the timing of screening is later than other programmes. All other programmes prioritise inpatient screening to maintain high capture and completion rates. Two programmes have no minimum screening age, one programme screens from four hours, and two screen from six hours. All programmes provide mechanisms for community screening of infants who do not complete screening while in hospital.

There is considerable variation in the timing and location of screening within the UNHSEIP. Some DHBs screen within the first few hours of birth and some do not screen before the infant is 24 hours old. Some larger DHBs screen a high proportion of babies as inpatients, while some small DHBs screen the majority of infants in community clinics.

Screening completion and refer rates

Each international programme achieved a screening completion rate of 96.5% or greater. Of the six programmes that provided data on completion of screening within 30 days, five achieved rates between 88.9% and 97.8%. The programme providing universal community screening achieved a rate of 73.3% within 30 days.

First screen refer rates were available from four programmes. The rate of the three programmes using AOAE as the first screen ranged from 8.8% to 23.5%. The rate from one A-ABR/A-ABR programme was 6.3%.

The UNHSEIP achieved a screening completion rate of 83.0%. Preliminary data provided by the NSU for October 2011 to December 2012 indicates a first screen refer rate from AOAE of approximately 15.0% and from the first A-ABR of approximately 5.0%. The refer rate to audiology was 1.7%[10].

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[10] Ministry of Health. (2013). UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012, National Screening Unit, Ministry of Health, New Zealand Government. Retrieved from: http://www.nsu.govt.nz/files/ANNB/UNHSEIP_Monitoring_report_April_to_Dec_2012.pdf

Screening devices

The international programmes reviewed use a range of screening devices. Devices that use Chirp and devices that do not use consumables are gaining the attention of programmes as they consider new device purchases.

Four different screening devices are used across New Zealand DHBs. Three devices use consumables for each screen. One A-ABR-only device does not require consumables, although the headphone pads must be replaced at intervals.

Data management

All international programmes reviewed have one or a combination of databases that is accessible across the jurisdiction. All databases include screening and diagnostic audiology information. Some include medical, early intervention and family support information. Each database is equipped with standard reporting tools to monitor performance. The scope of data auditing varies across programmes. Data management challenges include timely and accurate reporting of audiology data, 'double handling' of data entered into multiple systems, and reducing opportunities for post-screening fraud by protecting screening equipment text files.

In New Zealand, DHBs are required to report screening data to a national data collection. This database enables the collection of diagnostic outcome data from audiology, although compliance is low. Monitoring reports, developed by the NSU, provide a thorough analysis of the available data.

Each DHB has developed mechanisms for data collection, operational management and reporting. These range from sophisticated data systems, data entered into a spreadsheet, through to paper-based systems. Challenges arise from doubling, or tripling of data input effort with potential transcription errors, the use of several data repositories for screening information, use of different data sources for some data items, and delays in providing data back to DHBs. A national data repository is being developed that will record hearing screening events through the Maternity Clinical Information System.

Workforce

Internationally, workforces vary across the programmes reviewed and include nurses, midwives, dedicated screeners, and audiometric technicians.

The UNHSEIP has a dedicated screening workforce, supplemented in some locations by individuals in other roles who undertake screening as one of a number of responsibilities.

Governance

For all international programmes reviewed, jurisdiction-wide programme administration, planning and management occurs within an operational context such as a state-wide service, community health service or a tertiary hospital that is accountable to the state, province or local government.

The NSU of the Ministry of Health is responsible for setting the strategic direction; developing and maintaining policy and standards; national monitoring, auditing, evaluating and quality improvement; funding and contractual management; providing educational resources; and reviewing and overseeing the introduction of new technologies. The day-to-day operations of the UNHSEIP are managed by each DHB. DHBs deliver newborn hearing screening, diagnostic audiology and appropriate medical services.

Variables impacting on regime choice

Delivering optimal clinical efficacy

The clinical efficacy of a screening regime is determined by its capacity to identify moderate or greater permanent childhood hearing impairment and ANSD. The device/s must meet sensitivity and specificity requirements and have minimal impacts from extraneous and physiological noise, and from minor middle and outer ear conditions.

All regimes are able to detect moderate or greater hearing impairment.

A-ABR screening has higher sensitivity and specificity rates than AOAE screening[11, 12, 13].

A-ABR screening is not as susceptible to extraneous noise as AOAE screening. To a lesser extent, high levels of electrical interference and muscle artefact (i.e. large movements of the baby) can impact on the A-ABR screening process.

AOAE screening is more susceptible to outer ear and middle ear status in comparison to A-ABR screening, which results in higher referral rates.

Facilitating diagnostic audiology assessment as early as possible

Early completion of screening facilitates achievement of the benchmark of completion of audiology assessment by three months of age. The main operational advantage of an A-ABR regime is that screening can occur soon after birth and deliver a lower first screen refer rate than a regime with a first screen AOAE. Delays can result in babies being too old for electrophysiological audiology assessment. Babies older than three months typically require multiple audiology appointments to complete assessments due to lighter sleep states.

Achieving optimal screening capture and completion rates

The internationally accepted benchmark for universal newborn hearing screening capture and completion is that 95% of eligible infants complete screening by one month corrected age[14]. This is typically achieved through prioritisation of inpatient screening. A screening regime is required that is conducive to screening very soon after birth, and can be effectively administered in a hospital or birthing centre environment.

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[11] van den Berg, E., Deiman, C., van Straaten, H.L. (2010) MB 11 BERAphone hearing screening compared to ALGO portable in a Dutch NICU: a pilot study. *International Journal of Pediatric Otorhinolaryngology, 74*(10), 1189-92.

[12] Melagrana et al. (2007). MB 11 BERAphone and auditory brainstem response in newborns at audiologic risk: comparison of results.

[13] Cebulla, M., Stürzebecher, E. (2013). Detectability of newborn chirp-evoked ABR in the frequency domain at different stimulus rates. *International Journal of Audiology*, *52*(10), 698-705. doi:10.3109/14992027.2013.804634

[14] Joint Committee on Infant Hearing. (2007). Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs. *Pediatrics*, *120*(4) 898-921. doi: 10.1542/peds.2007-2333

Minimising the impacts on families

When determining a newborn hearing screening regime, the relative impacts on families must be considered. Three systematic literature reviews[15,16,17] have found minimal high quality evidence on issues pertaining to the impacts of newborn hearing screening on parents and their relationship with their infant. Of the available evidence, there is little to support suggestions of undue anxiety arising from newborn hearing screening in general, or more specifically, following a false-positive screening result.

Achieving optimal operational efficiency

Regimes involving two different screening approaches (i.e. those using a combination of AOAE and A-ABR screening) have significantly greater operational complexity than those involving a single screening approach (e.g. A-ABR/A-ABR).

This review has not identified published evidence exploring whether specific newborn hearing screening regimes warrant unique workforce arrangements. The review found that the workforce used to undertake screening is highly varied with at least seven workforce models identified.

For the UNHSEIP, there is a need to improve screening coverage and completion. A simplified regime would enable more flexible use of the broader health workforce. Opportunities that could be considered include incorporating newborn hearing screening into the roles of other health workers, and use of supervised telehealth where available.

A costings analysis considered the relative costs of the two-stage screening regimes identified in the literature review. Primary consideration was given to the regime used by New Zealand AOAE, A-ABR/A-ABR and A-ABR/A-ABR. The

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[15] Picton, N., Hyde, M. (2004). Family anxiety and universal newborn hearing screening (UNHS): A review of current evidence. Mount Sinai Hospital, Toronto, Canada. Retrieved from: http://childhearinggroup.isib.cnr.it/docs/unhs_and_family_anxiety.pdf

[16] Nelson, H. D., Bougatsos, C., Nygren, P. (2008). Universal newborn hearing screening: Systematic review to update the 2001 US Preventive Services Task Force recommendation. Pediatrics, 122(1), e266-276. doi: 122(1),e266-e276

[17] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee reference 17, Assessment report. Retrieved from: http://www.msac.gov.au/internet/msac/publishing.nsf/Content/8FD1D98FE64C8A2FCA2575AD0082FD8F/$File/ref17.pdf

impact of specific screening variables such as inpatient and outpatient screening were also considered.

For annual screening costs, including and excluding diagnostic costs, there were minimal differences between an AOAE, A-ABR/A-ABR regime and an A-ABR/A-ABR regime where the use of disposable ear cups are required. In contrast, where an A-ABR device did not need disposable ear cups, A-ABR/A-ABR showed a considerable cost advantage.

Inpatient screening has clinical and programme advantages in terms of population capture and early referral to audiology. It was also shown to have cost advantages over outpatient screening in a community screening clinic. The additional estimated cost to screen in community clinics is NZ$20 per infant.

Supporting the maintenance of quality standards

In the context of the UNHSEIP screening incident, the most conspicuous risk for a regime using an AOAE as the first screen is the capacity for a screener to screen their own hearing and falsify screening records to suggest that a specific infant's hearing has been screened. Whilst it is not impossible to self-screen utilising an A-ABR, it is more difficult than with an AOAE.

Quality management systems for regimes that combine two screening approaches (namely AOAE and A-ABR), and involve more than two screens, are more demanding and complex than those that use a single approach to screening and a maximum of two screens. Increased complexity arises from additional data management; follow-up and tracking; foundational screener training and updating; optimal maintenance of two different screening devices or modules; and programme auditing.

Regardless of the regime used, effective data systems are the cornerstone of monitoring programme quality and performance. The need to reliably track infants and reduce lost to follow-up rates is critical.

Devices for newborn hearing screening

All screening devices have strengths and limitations. Seven devices were reviewed against 15 criteria. The key criteria included high sensitivity and specificity, low false positive and false negative rates, scientific validation, minimising the risk of falsification, portability, and monitoring and data security. Each device met these criteria to varying degrees.

Three of the seven devices have been cited in published literature more frequently than others, and have a more rigorous foundation of validation. These are the MAICO MB 11 (BERAphone and Classic), the Natus ALGO 3i, and the MADSEN AccuScreen. The clinical advantages and disadvantages of these three devices are presented.

All three devices have similarities. Key differences between the devices include click A-ABR screening only with the Natus ALGO 3i; single ear screening with the MAICO MB 11 BERAphone (MAICO MB 11 Classic is binaural); limited touchscreen life and no data encryption for the MADSEN AccuScreen.

According to manufacturers, consumables are a highly negotiable cost. The MAICO MB 11 BERAphone does not use any consumables which brings a significant cost advantage. The ear cushions on this device require replacement with wear and tear, although this is not a frequent cost.

Ultimately, use of a single device across a jurisdiction holds many operational advantages. Significant financial savings can also be obtained for bulk purchases of devices and consumables, and training and maintenance contracts. The risks involved in using a single device are low and can be managed through rigorous tendering processes and careful contract management.

Every piece of equipment has risks and is open to compromise through inadvertent or deliberate acts.

With specific reference to the falsification of screening results, it is generally more difficult to self-screen using an A-ABR screening device than an AOAE screening device due to electrode placement and infant detection algorithms. However, there are other mechanisms through which falsification of records can occur.

Ultimately, risk mitigation involves minimising the opportunity to self-screen; increasing training including understanding of reasons for standardised protocols and ethical standards; increasing competency assessments; incorporating random spot checks into daily practice; and increasing accountability within the screener role. Ensuring effective monitoring, an accountable work place culture, and an open-door policy to support remedying errors and facilitate knowledge and skill development is essential to all programmes.

All manufacturers are willing to provide extensive training and support. This can be negotiated in any tender process for a new device, and may include yearly training and competency checks.

Recommendations

The review team has made three recommendations. The key factors guiding the formulation of these recommendations include supporting the UNHSEIP to:

- deliver optimal clinical efficacy and efficiency,

- facilitate diagnostic audiology assessment as early as possible,

- achieve optimal screening capture and completion rates,

- minimise impacts on families,

- achieve optimal operational efficiency, and

- meet specified quality standards.

Recommendation 1

Implement a two stage A-ABR screening regime for all neonates, including for neonates who have been under the care of a neonatal intensive care unit. Medical exclusions should continue, with direct referral to audiology.

Recommendation 2

Specify a standard screening device for all screening. The device must:

a. demonstrate sensitivity >95% and specificity >90%,

b. be validated through peer reviewed studies published in international journals, with appropriate sample size and scientific methodology,

c. have a validated detection algorithm,

d. be user friendly/functional for the screener workforce,

e. minimise ease of falsifying a screen,

f. enable data monitoring and data security,

g. be time effective relative to other devices,

h. contribute to cost effective screening outcomes,

i. be portable,

j.   be consumer friendly,

k.   enable efficient data upload and download,

l.   be calibrated to an internationally published reference,

m. have an effective, efficient and flexible computing interface,

n.   incorporate comprehensive support, including training, and

o.   be compatible with current and future technologies.

Recommendation 3

Ensure the following conditions are met to facilitate both a successful change in regime and device, and to optimise the overall effectiveness of the programme:

a.   use a nationally managed organisational change process to facilitate implementation of the new regime and introduction of a nationally consistent device,

b.   standardise the screening regime, device, clinical practice and protocols nationally,

c.   prioritise inpatient screening, including reducing the minimum age of screening,

d.   review targeted follow-up criteria,

e.   increase flexibility of workforce models, particularly in regional and rural areas,

f.   strengthen current UNHSEIP continuous quality improvement processes by linking them to a set of international best practice benchmarks,

g.   as a matter of priority, establish a national information system or data system which fulfils operational needs in real time as well as monitoring and reporting needs at DHB and national levels,

h.   establish a regional system of operational management that transcends DHB boundaries and interfaces with the governance function of the NSU, and

i. build upon and continue existing expert, multidisciplinary, clinical advisory forums and processes to guide the implementation of the change to the regime and device, as well as ongoing feedback and advice regarding programme performance.

CHAPTER 1 – Background

Introduction

Affecting approximately 1.5 per 1,000 live births[18], permanent childhood hearing impairment (PCHI) is recognised as one of the most prevalent congenital childhood conditions[19]. The developmental opportunities afforded by identifying children who have congenital hearing loss in the first months of life, and providing appropriate intervention, have been recognised for decades.[20] Children diagnosed with a PCHI through universal newborn hearing screening, and who commenced early intervention services by six months of age, have been shown to have significantly better communication skills, parental bonding and parental grief resolution than those whose hearing loss is identified at a later age.[21] In this context, universal newborn hearing screening in association with relevant early intervention services, is increasingly becoming accepted practice for the care of infants in the newborn period[22].

The New Zealand UNHSEIP

In New Zealand, the Universal Newborn Hearing Screening and Early Intervention Programme (UNHSEIP) was implemented over a three-year period from 2007 – 2010 with the aim of:

> *early identification of newborns with hearing loss so that they can access*
> *timely and appropriate interventions, inequalities are reduced and the*

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[18] Centers for Disease Control and Prevention. (2013). Summary of 2011 national CDC EHDI data. Retrieved from: http://www.cdc.gov/ncbddd/hearingloss/ehdi-data2011.html

[19] U.S. Preventive Services Task Force. (2008). Universal screening for hearing loss in newborns: Recommendation statement. Retrieved from: http://www.uspreventiveservicestaskforce.org/uspstf08/newbornhear/newbhearrs.htm

[20] White, K. R., Forsman, M. S., Eichwald, J., Munoz, K. (2013). The foundations and evolution of EHDI. In L. R. Schmeltz (Eds) *The National Center for Hearing Assessment and Management e-book: A resource guide for early hearing detection and intervention (EHDI)* (pp. 1.1 – 1.18). Utah State University. Retrieved from: http://www.infanthearing.org/ehdi-ebook/index.html

[21] Yoshinaga-Itano, C. (2003). Early intervention after universal neonatal hearing screening: impact on outcomes. *Mental Retardation and Developmental Disabilities Research Review, 9*(4) 252-66.

[22] World Health Organization. (2010). Newborn and infant hearing screening: Current issues and guiding principles for action – Outcome of a WHO informal consultation held at WHO Headquarters, Geneva, Switzerland, 09 – 10 November 2009. Retrieved from: http://www.who.int/blindness/publications/Newborn_and_Infant_Hearing_Screening_Report.pdf

outcomes for these children, their families and whānau, communities and society are improved.[23]

By 2010, all 20 District Health Boards (DHBs) across New Zealand were providing universal newborn hearing screening. The UNHSEIP works to the specific goals of babies completing hearing screening by one month of age, diagnostic audiology by three months, and commencement of early intervention by six months[24].

The screening services provided by the UNHSEIP are overseen by the National Screening Unit (NSU) of the Ministry of Health.

Falsification of screening records

In 2012, the NSU was notified that two newborn hearing screeners from two DHBs had not followed screening protocols. A subsequent audit, as one element of a review of the incident, identified that between 2009 and 2012, approximately 2,000 babies were not screened according to protocols. At the time of publishing the review report in December 2012, eight screeners, from six DHBs, had been identified to have not complied with screening protocols. The three ways in which screening protocols were not being followed included:

- screening the same ear of a baby twice,

- screening one ear of the baby, and one of the screener's ears, and

- screening both of the screener's ears instead of the baby's ears[25].

The report also identified a similar incident known to have occurred in two different National Health Service (NHS) Newborn Hearing Screening Programmes in the United Kingdom (UK). In this context a screener entered a pass response for infant ears that had not been screened, either through conducting a repeat screen on one ear of the infant and recording a clear response

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[23] Ministry of Health. (2013). Universal Newborn Hearing Screening and Early Intervention Programme (UNHSEIP): National policy and quality standards. Wellington: Ministry of Health. Retrieved from: http://www.nsu.govt.nz/files/UNHSEIP-national_policy_and_quality_standards-jun13.pdf

[24] ibid.

[25] Ministry of Health. (2012). Quality improvement review of a screening event in the Universal Newborn Hearing Screening and Early Intervention Programme. Retrieved from: http://www.nsu.govt.nz/files/ANNB/Newborn_Screening_Report_FINAL.pdf

for both ears, or after a clear response in the baby's first ear, screening their own ear and recording it as a result for the other ear of the baby.

The report identified a number of factors as having contributed to the incident occurring, including i) individual screener factors, ii) training/education, iii) resource constraints, iv) programme management, v) absence of individual screeners monitoring and awareness of monitoring, and vi) an AOAE/A-ABR screening protocol[26].

The review made 21 recommendations in the following areas:

- the screening protocol,

- individual screener monitoring,

- the screener role,

- the coordinator role,

- audiology, and

- programme management.

Recommendation 1 of the review specified that:

> *The NSU must reassess the screening protocol with a view to changing to an A-ABR only protocol.*

To fulfil this recommendation, in September 2013, the New Zealand Ministry of Health contracted Young Futures to:

> *'examine best practice in newborn hearing screening regimes, including associated equipment options, to assist the National Screening Unit to determine the most appropriate screening regime for the New Zealand Universal Newborn Hearing Screening and Early Intervention Programme.'*

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[26] Ministry of Health. (2012). Quality improvement review of a screening event in the Universal Newborn Hearing Screening and Early Intervention Programme.

Overview of the report

This report presents the outcomes of this work:

- Chapter 1 provides an overview of the context of the review.

- Chapter 2 discusses factors recognised internationally to be central to good practice in newborn hearing screening.

- Chapter 3 presents the review method and limitations to the review method.

- Chapters 4, 5 and 6 detail the findings of a rapid review of the literature on emerging technologies in newborn hearing screening and current hearing screening regimes; evidence from nine international newborn hearing screening programmes; and an overview of newborn hearing screening practice in New Zealand.

- Chapter 7 brings these findings together and provides an analysis of variables influencing newborn hearing screening regime choice.

- Chapter 8 presents the key criteria for decision making regarding screening devices, alongside a detailed overview of current screening device options, their key specifications, and clinical advantages and risks.

- Chapter 9 details three recommendations to the NSU regarding the newborn hearing screening regime, hearing screening devise and details regarding conditions that will need to be met to facilitate both a successful change in regime and device, and to optimise the overall effectiveness of the UNHSEIP.

CHAPTER 2 – International standards

In the nearly 45 years since the Joint Committee on Infant Hearing (JCIH) was established in the USA, it has become the recognised international reference point for best practice in newborn hearing screening. The JCIH identifies the goal of early hearing detection and intervention as being 'to maximize linguistic competence and literacy development for children who are deaf and hard of hearing.'[27]

Approaches to newborn hearing screening

The 2007 JCIH Position Statement states a commitment to identifying all degrees and types of hearing loss, including mild permanent hearing impairment and neural hearing impairment (auditory neuropathy spectrum disorder). The JCIH recognises that current screening technologies are most effective at reliably identifying moderate and greater hearing loss. JCIH indicates that all infants should have access to hearing screening using a physiologic measure (including automated otoacoustic emissions (AOAE) and/or automated auditory brainstem response (A-ABR) before one month of age. To detect sensory and/or conductive hearing impairment, most inpatient well-infant protocols provide one screen and, when necessary, a second screen (of both ears) no later than at the time of hospital discharge. It is recognised that infants who do not enter the screening pathway by receiving at least their first screen before hospital discharge may be at a higher risk of not completing hearing screening[28].

When A-ABR is used as the single screening technology for both screens (rather than AOAE followed by A-ABR) auditory neuropathy spectrum disorder (ANSD) can also be detected. Given the known risks for ANSD for infants who spend more than five days in neonatal intensive care (NICU), A-ABR screening is recognised as the only appropriate screening technique for these infants.

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[27] Joint Committee on Infant Hearing. (2007). Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs.

[28] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia. Cancer and Screening Unit Working Paper, Cat. No. CAN 73. Retrieved from http://www.aihw.gov.au/WorkArea/DownloadAsset.aspx?id=60129545175

Screening devices and protocols

To ensure consistency of screening across infants, screening conditions, and screening personnel and to remove the need for screening interpretation and reduce impacts of screener bias or error, it is important to use technologies with automated-response detection. The JCIH 2000 emphasises that screening sensitivity and specificity should be evidence-based[29]. Furthermore, attempts should be made to minimise confounding extrinsic and intrinsic variables that contribute to potential false negative and false positive rates. Particular care should be exercised in the consideration of rescreening outside of specified protocols, which may result in an increase in exposure to confounding variables, hence utilisation of rescreens should be on an exception only basis.

Screening results should be communicated immediately to families and medical professionals to ensure their understanding of the outcome and importance of follow-up when indicated. When indicated, an appointment for follow-up screening should be made before discharge.

Diagnostic audiology and medical assessment

When an infant does not pass screening they should have appropriate audiological and medical evaluation to confirm the presence of a hearing impairment before three months of age. Infants with confirmed permanent hearing impairment should be referred to early intervention services as soon as possible after diagnosis but no later than six months of age. This includes otologic and other medical evaluation to identify the cause of the hearing impairment, related physical conditions, and provide recommendations for treatment and referral to other services.

Targeted audiology follow-up

The JCIH identifies 11 risk indicators associated with congenital or delayed-onset hearing loss to guide targeted surveillance. These risk factors have three roles i) historically, to identify infants who should receive audiological evaluation but who live in locations where universal hearing screening is not yet available, ii) to help identify infants who pass newborn screening but are at risk of developing

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[29] Joint Committee on Infant Hearing. (2000). Special Article, Year 2000 Position Statement: Principles and Guidelines for Early Hearing Detection and Intervention Programs Joint. Retrieved from: http://www.jcih.org/jcih2000.pdf

delayed-onset hearing loss, and iii) to identify infants who pass newborn screening but have mild permanent hearing loss.

Partnerships

The JCIH recognises that programmes need families to work in partnership with professionals as a coordinated team and the responsibilities of team members must be well-defined and understood.

Data management

In the 2000 position statement[30], the JCIH recommended development of uniform registries and national information databases using standardised methodology, reporting, and system evaluation. It was noted that information systems should be designed and implemented to interface with electronic health charts and should be used to measure outcomes and report the effectiveness of early hearing detection and intervention services at the child, practice, community, and national levels.

Quality management and benchmarks

The national information-management system should assist health care professionals and the state health agency measure screening, diagnosis and intervention quality indicators and provide the means to determine the extent to which processes are stable and sustainable and conform to stated benchmarks. The JCIH reinforces that timely and accurate monitoring of quality measures is essential.

The JCIH focuses on routine performance measurement and recommends inter-programme comparison and continuous quality improvement. The use of performance benchmarks, determined by a consensus of expert opinion, that set the minimum standard to be attained by high-quality programmes are emphasised. Undertaking frequent measures of quality enable timely recognition and correction of 'unstable' elements of screening programmes. The JCIH presents the following quality indicators for screening, confirmation of hearing loss and early intervention:

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[30] Joint Committee on Infant Hearing. (2000). Special Article, Year 2000 Position Statement.

- \> 95% of infants complete screening by one month corrected age,

- < 4% of infants are referred from screening to audiology,

- 90% of infants who are referred from screening to audiology complete comprehensive audiological evaluation by three months of age,

- 95% of infants with confirmed bilateral hearing loss whose families elect to use amplification receive amplification devices within one month of hearing loss confirmation,

- 90% of infants with confirmed hearing loss who qualify for early intervention services commence services no later than six months of age,

- 95% of children with acquired or late-identified hearing loss who qualify for early intervention services commence services no later than 45 days after diagnosis, and

- 90% of infants with confirmed hearing loss receive a first developmental assessment with standardised assessment protocols (not criterion reference checklists) for language, speech, and nonverbal cognitive development by no later than 12 months of age.

Meeting these standards contributes not only to optimal outcomes for children and their families, but also the efficient and effective operation of a newborn hearing screening programme.

Individual jurisdictions will typically set local standards that supplement and elaborate on the details of the JCIH guidelines and quality indicators. One example is the *National performance indicators for neonatal hearing screening in Australia*[31]. Examples include, but are not limited to, benchmarks for parental decline of screening (<1%), parental consent for diagnostic audiology (99%), referrals to diagnostic audiology services are made in less than five days (>97%), and families are referred to Australian Hearing (to consider amplification options) within three days of confirmed hearing loss (>97%).

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[31] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia.

CHAPTER 3 – Review method

To fulfil the project objective, the review process was designed to facilitate understanding of the following seven questions:

1. What are the clinical and practical benefits and risks of different newborn hearing screening regimes and associated screening devices?

2. What screening regimes and associated screening devices are used in programmes comparable to the UNHSEIP?

3. What are the advantages and disadvantages of these different screening regimes and the screening devices used?

4. What operational factors might impact on the cost of different newborn hearing screening regimes and choice of associated screening devices? What might the scale of these impacts be?

5. How does current day-to-day practice and the recent screening incident in New Zealand affect the decision to remain with or implement an alternative screening regime using existing or alternative screening devices?

6. What are the features of the specific screening devices that may be suitable for use by the UNHSEIP?

7. What are the relative merits of different screening regimes and associated screening devices, including the status quo?

Rapid review of the literature

A rapid review of the literature was carried out to review evidence regarding the clinical and practical benefits and risks of different hearing screening regimes, including regimes that use AOAE only, regimes that use a combination of AOAE and A-ABR, and regimes that use A-ABR only. Emerging techniques for consideration in the area of newborn hearing screening are also presented.

The specific search terms were formulated in collaboration with the NSU. These are presented in Appendix A.

Both published and unpublished literature, from 2003 to 2013 was included. Publication types included high quality and/or primary studies, systematic reviews, landmark and frequently cited studies, published commentaries, quasi-experimental studies and/or observational studies. Literature excluded from the rapid review included literature published before 2003 (unless recognised as a landmark publication), literature published in languages other than English, opinion pieces, literature pertaining to monitoring and surveillance, and literature regarding diagnostic audiology.

Review of selected international programmes

A review of selected international programmes was carried out to build understanding of the practical experiences of comparable jurisdictions using different newborn hearing screening regimes and devices and to consider the impact of these variables on operational issues.

The jurisdictions invited to contribute were determined in collaboration with the NSU and the independent experts providing advice to the review. Seventeen programmes from the United Kingdom, Europe, Canada, USA and Australia were invited to participate in a semi-structured telephone interview and to provide details of a number of data items. Details of the interview questions asked and data items sought are presented in Appendix C and Appendix D respectively.

Nine programmes participated in the review. Seven programmes participated in an interview and provided programme data, one programme participated in an interview but did not provide programme data, and one programme provided programme data but did not participate in an interview.

Understanding the UNHSEIP and its context

Reviewing UNHSEIP documents

The UNHSEIP provided a comprehensive set of documents relevant to the UNHSEIP, including but not limited to terms of reference; quality standards; monitoring frameworks, protocols and reports; training protocols and manuals; role specifications; audit reports; and programme reports including the review of the screening incident.

These documents were reviewed and analysed by the project team. A full list of documents reviewed is provided at Appendix E.

Interviewing UNHSEIP stakeholders

Telephone interviews were conducted with 12 stakeholders of the UNHSEIP. Interviewees included:

- a consumer representative involved in audits of DHB screening and the UNHSEIP Advisory Group,

- employees of seven DHBs, incorporating diverse roles, including:

    o screening (n = 4)

    o screening team leader (n = 2)

    o screening coordination (n = 4)

    o screener training (n = 2)

    o contributing to audits of DHB screening (n = 2)

    o audiology (n = 3),

- a representative from Child and Youth Health, Ministry of Health, involved as a clinical lead in establishing the UNHSEIP and a member of the UNHSEIP Advisory Group, and

- two representatives from the NSU, whose respective roles included providing:

    o strategic direction to the UNHSEIP, and

    o data analysis for the UNHSEIP.

Interviewees involved in the coordination and delivery of screening represented diverse contexts ranging from tertiary hospitals in large metropolitan centres to regional and rural areas providing screening in multiple small hospitals and birthing centres across a large geographic area. Across the group, experience included use of a range of screening devices, including the MADSEN Classic AccuScreen, MADSEN AccuScreen, MAICO MB 11 BERAphone, and ALGO 3i.

The interviews explored the role of each individual in the UNHSEIP, their perceptions of the strengths and weaknesses of the current screening regime and devices, issues relating to data management, the opportunities likely to arise from a change of regime and/or devices, issues requiring consideration if a change in regime and/or devices was to occur, and reasons for maintaining the status quo.

Understanding the local context

To supplement the review of UNHSEIP documents and interviews with UNHSEIP stakeholders, the review team met with the NSU and visited two DHBs. The two locations were selected on the basis of their contrasting demographics and clinical context. One DHB was in a large metropolitan location and included a tertiary hospital (Auckland DHB) and the other was in a regional community, incorporating a main regional hospital and multiple smaller birthing centres (Northland DHB).

All relevant individuals involved in each of these programmes were invited to participate in the visit. The process involved discussion around the local implications of implementing a range of different possible screening regimes, including the status quo. The focus was on:

- surfacing information about the variables influencing the programme in each of the two contexts, and

- identifying the unique features of the UNHSEIP and the broader New Zealand context against the findings of the literature review and the review of selected international programmes.

Analysis of variables impacting on regime choice

A review of variables impacting on regime choice was undertaken. The variables considered included:

- delivering optimal clinical efficacy and efficiency,

- facilitating diagnostic audiology assessment occurring as early as possible,

- achieving optimal screening capture and completion rates,

- minimising the burden on families,

- achieving optimal operational efficiency, including cost efficiency and effectiveness, and

- supporting the maintenance of quality standards.

Review of devices

Device technical specifications from each manufacturer were reviewed and compared. In addition, a thorough search of available scientific literature pertaining to each device was undertaken on clinical databases, including but not limited to PubMed, MedLine and Ovid. Telephone and/or face-to-face interviews were carried out with the audiological and/or technical representatives of the relevant manufacturers. Some devices were trialled by a member of the review team who has extensive experience in newborn hearing screening and diagnostic audiology for newborns. The objective was to identify screening utility, advantages and disadvantages and identify potential risks of each device.

Expert advisors

The project team sought the input of two independent experts to provide input and additional perspectives at key stages of the review. The independent experts included Dr Carlie Driscoll, Senior Lecturer in Audiology at the University of Queensland, and Ms Gwen Carr, Programme Lead of the NHS Newborn Hearing Screening Programme and Honorary Senior Research Associate of the University College London Ear Institute.

The independent experts contributed by providing feedback and advice regarding:

- the proposed review method,

- the literature review search terms,

- possible programmes to invite to participate in the review of selected international programmes, and

- the draft recommendations to the NSU.

Limitations of the review

Although the review sought to comprehensively consider the full range of evidentiary and practical factors that might inform a decision by the NSU regarding the future screening regime and screening devices, a number of limitations to the review must be acknowledged.

- The literature review undertaken was limited to a rapid review of the literature.

- Since 2012, the UNHSEIP has undertaken extensive programme audits, updated training processes and made changes to monitoring frameworks. The intended programme improvements from many of these processes have not yet had the opportunity to fully manifest within the programme and available programme data.

- Being based in Brisbane, Australia, the review team only had a brief opportunity to engage directly with UNHSEIP services 'on the ground'.

- Although the review team spoke with a wide number of representatives from across the UNHSEIP, it was not possible to develop a full understanding of the circumstances and experiences with newborn screening within each DHB.

- Limitations in current data collection systems and processes have made it difficult to make a fully informed assessment of the performance of current practices within the UNHSEIP and improvements that may have occurred as a result of recent programme changes.

Despite these limitations, the review team are confident that the processes undertaken were both robust and appropriate to inform the evidence based recommendations this report presents to the NSU.

CHAPTER 4 – Findings: Evidence from the literature

Introduction

This rapid review of the literature presents current evidence regarding newborn hearing screening regimes that use AOAE alone, screening regimes that use a combination of AOAE and A-ABR, and screening regimes that use A-ABR alone. The clinical and practical benefits and risks, and the advantages and disadvantages of screening regimes within each of these three categories are presented. Emerging techniques for consideration in the area of newborn hearing screening are also presented.

It must be recognised that literature review findings alone cannot be used to make decisions regarding the most appropriate screening regime in a specific context.

Method

This literature review has been conducted using a rapid review methodology. A rapid review of the literature is designed to identify and explain a topic of interest and provide an organised and structured overview of relevant evidence identified. It does not employ the same level of rigour as that of a systematic review of the literature, and for this reason is subject to an increased bias and/or error. It is a valuable approach for informing decision making about the practicalities of a topic of contention and provides evidence to decision makers within a short timeframe; streamlining the approach to a timely collection of available evidence, collating it into a user-friendly report, and informing decision makers regarding emergent topics.

This rapid review of the literature provides a summary of key evidence published in English between 2003 and 2013 relevant to research questions and search terms formulated in collaboration with the NSU. A full list of search terms is presented in Appendix A.

Both published and unpublished literature was included. Publication types included:

- high quality and/or primary studies (with 'quality' determined based on methodological rigour),

- systematic reviews,

- landmark and frequently cited studies,

- published commentaries (to provide background and contextualise the literature), and

- quasi-experimental studies and/or observational studies, including those with prospective and/or rigorous quantitative analyses, particularly those published in peer reviewed journals.

Literature excluded from the rapid review included:

- literature published before 2003 (unless recognised as a landmark publication),

- literature published in languages other than English,

- opinion pieces,

- literature pertaining to monitoring and surveillance,

- literature regarding diagnostic audiology,

- small sample sized studies including case studies, and

- methodologically poor studies.

This resulted in the exclusion of five articles.

Retrieval of full text documents took place following screening of abstracts. Full text articles were obtained through the journal subscriptions held by the Queensland State Library, including but not limited to, peer reviewed databases such as PubMed, Ovid, Medline, Medline Plus, CINAHL and the Cochrane Library. Non-peer reviewed databases including, e.g., Google, MetaCrawler were also used. Literature not available electronically was excluded due to time and resource constraints.

The rapid review concisely and methodically addresses all components of the agreed questions. The review questions mark out sections of the report, with key findings clearly and concisely presented at the beginning of each section, followed by an explanation and discussion of the literature. Each section is concluded with a practical statement of the 'bottom line' implications of the literature review findings. These 'bottom line' findings from each section are brought together in Appendix B as an easy reference regarding the conclusions drawn from the rapid review of the literature.

Screening stages

Screening regimes consist of the type of screening, the number of stages, and the number of steps involved in the screening process prior to discharge or referral to audiology for diagnostic assessment. For simplicity, stages are similar to 'appointments' or occasions that a child is seen. Screening regimes typically have either one, two or three stages. The number of steps refers to the number of screens. For example, a screening regime may have two stages, but three steps, if one of the stages involves the use of two different screens.

A one stage regime involves a single screening appointment. The regime may utilise an A-ABR and/or a transient evoked otoacoustic emissions screen (TEOAE) or distortion product otoacoustic emissions screen (DPOAE).

When using a one stage, one step regime, after a single refer result on a single screen, the infant is immediately referred to audiology for diagnostic testing. Countries which use the TEOAE screen as a one stage, one step screening regime include Brazil, India and Serbia[32].

A one stage screening regime can include the option of completing two screens in the one appointment. This is known as a one stage, two step regime. If a baby has a first screen and passes, they are discharged. If the baby has a first screen and refers, within the same appointment the screener performs another screen using a different screening method (e.g. AOAE then A-ABR). If the baby has a pass result on the second screen they are discharged, but if they have a pass result they are referred to audiology for diagnostic assessment.

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[32] World Health Organization. (2010). Newborn and infant hearing screening: Current issues and guiding principles for action.

A two stage screening regime may include either one or two screening steps in each of two stages. Each stage may involve use of the same screening type (e.g. AOAE/AOAE or A-ABR/A-ABR) or different screening types (e.g. AOAE/A-ABR or AOAE, A-ABR/A-ABR or AOAE/AOAE, A-ABR). In the majority of two stage regimes, babies are only referred for a second stage screen (or screens) if they do not pass the first stage screen (or screens). Once a baby passes a screen they are discharged from screening. In India and Serbia, having already completed an initial stage with a TEOAE screen, a second stage with TEOAE is performed. In Sweden and the United States, TEOAEs and A-ABR are used; and, A-ABR alone is used in the second stage in Germany, the Netherlands, and Australia.

Rarely, a three stage regime is used. For example, in India, TEOAE is used in the first stage and is then repeated in a second stage. Finally, in a third stage, A-ABR is used.

Table 1 outlines examples of possible screening regimes, including different sessions and steps. In New Zealand, for example, the progression would typically involve a neonate having a first stage DPOAE and A-ABR (if required), followed by a second stage A-ABR at a later time or date (if required). In most States and Territories of Australia, the first stage is a single A-ABR, followed by a second stage single A-ABR.

Table 1. Stages in newborn hearing screening programmes.

Number of stages	Options	Number of steps	Stage 1	Stage 2	Stage 3
1 stage regimes	Option 1	1	AOAE	NA	NA
	Option 2	1	A-ABR		
	Option 3	2	AOAE, A-ABR		
2 stage regimes	Option 1	2	AOAE	A-ABR	NA
	Option 2	2	A-ABR	A-ABR	
	Option 3[A]	3	AOAE, A-ABR	A-ABR	
	Option 4	3	AOAE	AOAE, A-ABR	
3 stage regimes	Option 1	3	AOAE	AOAE	AOAE
	Option 2	3	A-ABR	A-ABR	A-ABR
	Option 3	3	AOAE	AOAE	A-ABR

[A]Regime used by the UNHSEIP.

The first and second stages of screening are typically performed in hospitals (China, Germany, India, Serbia, Australia, and the USA), while third stage

screens are more likely to occur in a range of different locations, including hospitals (India), in hearing health care services (Brazil), paediatric audiological or ENT departments and practices (Germany)[33]. In the majority of situations, provisions are made for community screening where initial screening stages are completed in hospital but follow-up screening is required after discharge.

Literature review findings

Technologies on the horizon

Q.1. What technologies are emerging but are not currently viable for population level screening due to current costs, lack of evidence, or lack of test refinement?

Key findings

Genetic screening

- Genetic screening for hearing impairment is not currently viable. Forty per cent of permanent childhood hearing impairment (PCHI) does not have a genetic cause and can be attributed to environmental factors.

- Two key genes are linked to PCHI; Connexin 26 mutation and Connexin 30 deletion. Genetic screening cannot discern between the two without a hearing screen or assessment.

- The importance of maintaining newborn hearing screening programmes, even in the context of genetic testing, is widely acknowledged given both the environmental causes and the determination of the genetic cause.

Multi frequency Auditory Steady State Responses (MASSRs)

- The technique is designed for threshold estimation only, not as a hearing screening tool.

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[33] World Health Organization. (2010). Newborn and infant hearing screening: Current issues and guiding principles for action.

- Although an important emerging technique for audiological assessment, with good specificity (95%) and sensitivity (100%)[34], literature on MASSR is scant, especially on the relationship to hearing impairment in newborns.

- Cautious interpretation of results is required for newborns with PCHI. This is particularly the case for those with hearing thresholds in the normal to moderate range of hearing impairment due to the lack of evidence correlating findings to behavioural thresholds[35].

Band width chirp auditory steady state responses

- This is a newly emerging area therefore it is in the early stages of validation and research.

Wideband acoustic immittance

- WAI research is in its early stages of development. The technique is designed as a diagnostic tool and the applicability to the newborn population as a screening tool is yet to be determined.

- Normative data is continuing to be collated.

- The technique is not yet adequately developed for application to a universal newborn hearing screening programme.

Background

This section briefly describes new and emerging technology which may one day be useful for universal newborn hearing screening programmes. While these methods are presented for consideration, for reasons such as cost, efficacy, and lack of substantiated evidence, they are not yet adequately developed to be considered for implementation.

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[34] Savio, G., Perez-Abalo, M.C., Gaya, J., Hernandez, O., Mijares, E. (2006). Test accuracy and prognostic validity of multiple auditory steady state responses for targeted hearing screening. *International Journal of Audiology*, *45*(2), 109-120.

[35] Rance, G., Beer, D.E., Cone-Wesson, B., Shepherd, R.K., Dowell, R.C., King, A.M.,…Clark, G.M. Clinical findings for a group of infants and young children with auditory neuropathy. *Ear and Hearing*, *20*(3), 238-52.

Genetic testing

Typically, once a child is diagnosed with a permanent hearing impairment, they undergo genetic testing to assist in determining the cause of the hearing impairment. The most recent evidence suggests that while a genetic cause can be found for approximately 60% of children who have a congenital hearing impairment, the remaining 40% of babies have a hearing impairment with an environmental origin such as cytomegalovirus (CMV), herpes, toxoplasmosis, ototoxicity, syphilis, head trauma, or sub-arachnoid haemorrhage[36,37].

It has been suggested that screening for the three main genes responsible for hearing impairment, in conjunction with a test for CMV within three weeks of birth, would detect almost 60% of late onset hearing impairment. However, the interpretation of even simple molecular tests is not possible without a hearing screening programme. For example, to correctly identify whether a child is a heterozygous carrier of a Connexin 26 mutation or a carrier of a deletion of the Connexin 30 gene, it must be known whether the child's hearing is normal or not. A normal hearing screen indicates the former and differentiates between the two.

Furthermore, mutations in the ACTG1, CDH23, CLDN14, COCH, COL11A2, DFNA5, DFNB31, DFNB59, ESPN, EYA4, GJB2, GJB6, KCNQ4, LHFPL5, MT-TS1, MYO15A, MYO6, MYO7A, OTOF, PCDH15, POU3F4, SLC26A4, STRC, TECTA, TMC1, TMIE, TMPRSS3, TRIOBP, USH1C, and WFS1 genes are all known to cause non-syndromic deafness. GJB2 is linked to Connexin 26 protein, one of the most common non-syndromic causes of PCHI.

It is not within the scope of this review to discuss all possible genetic factors linked to hearing impairment. While genetic testing may become more sophisticated in the future it is currently not able to replace newborn hearing screening as genetic testing is not yet able to detect every genes responsible for hearing impairment. Genetic testing does, however, look like a promising tool when used concurrently with hearing screening[38].

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[36] Kennedy, C., McCann, D. (2004). Universal neonatal hearing screening moving from evidence to practice. *Archives of Disease in Childhood – Fetal and Neonatology, 89*(5), F378-F383. doi: 10.1136/adc.2003.034454

[37] Fortnum, H.M., Summerfield, A.Q., Marshall, D.H., Davis, A.C., Bamford, J.M. (2001). Prevalence of permanent childhood hearing impairment in the United Kingdom and implications for universal neonatal hearing screening: Questionnaire based ascertainment study. *British Medical Journal*, 323, 536-540. doi: http://dx.doi.org/10.1136/bmj.323.7312.536

[38] Zhang, J., Wang, P., Han, B., Ding, Y., Pan, L., Zou, J.,…Wang, Q. (2013). Newborn hearing concurrent genetic screening for hearing impairment – A clinical practice in 58,397 neonates in Tianjin, China.

A significant limitation of genetic testing is that currently only one or a short sequence of genes can be tested. Hearing impairment is often attributed to multiple mutations in multiple genes and therefore the process of selecting appropriate test(s) is challenging. The cost of genetic testing weighs heavily against the responsibility to order the most appropriate test. While next generation genetic testing is moving to the testing of multiple genes in a cost effective manner, research is only in its early infancy[39]. At this stage genetic testing is an excellent adjunct procedure following the diagnosis of hearing impairment as opposed to a universal screen.

Emerging audiology technologies

The following outlines emerging tests which have potential in newborn diagnosis and detection of hearing impairment.

Multi-frequency auditory steady state responses (MASSR)

Auditory steady state responses (ASSR) are currently used in the audiological diagnostic test battery. An ASSR provides specific information about the thresholds and frequencies of a hearing impairment. This is useful for subsequent hearing aid fitting as it can map results onto a behavioural audiogram (i.e. graph of hearing). Multi-frequency auditory steady-state evoked responses (ASSR) are also a measure of hearing across four frequencies using analysis of the evoked potential. The multi-frequency ASSR differs from the ASSR, in that the technique assesses all four frequencies at the same time, providing the same information in a much faster timeframe.

Current literature is cautious about the correlation of the single ASSR result with hearing thresholds, particularly in the mild to moderate range[40]. For this reason, ASSRs are not recommended as a screening tool[41]. They are rarely utilised in

International Journal of Pediatric Otorhinolaryngology. 77(12), 1929-35. doi: 10.1016/j.ijporl.2013.08.038. [Epub ahead of print]

[39] Rao, A., Schnooveld, C., Schimmeti, L.A., Vestal, B.A., Ferrello, M., Ward, J., Friedman, B. (2011). Genetic testing in childhood hearing loss: Review and case studies, *Audiology Online*. Retrieved from: http://www.audiologyonline.com/articles/genetic-testing-in-childhood-hearing-820.

[40] Rance, G.B., David, E., Cone-Wesson, B., Shepherd, R.K., Dowell R.C. (1999). Clinical findings for a group of infants and young children with auditory neuropathy. *Ear Hear, 20*(3) 238-52.

[41] Mahmoudian, S, Farhadi, M, Kadivar, M, Ghalehbaghi, B, Rahimi, F, Hemami, M.R, Mohagheghi, P. (2011). Prognostic validity of dichotic multiple frequencies auditory steady-state responses versus distortion

isolation and usually form a part of a thorough diagnostic audiology test battery. The recommendation to use multi-frequency ASSRs for threshold estimation still stands for diagnostic audiology[42], and the approach ultimately has the potential to provide a frequency specific screen at some stage in the future, permitting device development.

Band width chirp auditory steady state responses

The use of band width chirps (a differing stimulus type that is frequency specific) in ASSRs has not been well validated in the literature at this time. Further research is required on band width chirp ASSR methods before they can be used for newborn hearing screening. The use of band width ASSRs for screening has significant opportunities in terms of the provision of frequency specific information. However, this is a newly emerging area.

Wide-band acoustic immittance

As a new technique to measure middle ear function, wide-band acoustic immittance (WAI) has strengths as a complimentary tool in newborn hearing screening programmes. WAI could specifically assess newborns that refer from AOAE screening. Evidence indicates that these neonates would have lower absorbance for frequencies from 1 to 3 kHz[43] which suggests results consistent with middle ear issues at birth. In newborn hearing screening programmes, it is proposed that WAI may help with the interpretation of hearing screening results, particularly for programmes using AOAE. Despite only having a small amount of evidence on the use of WAI and associated methodological considerations, current results include the presence of normative data[44], which are promising. However, further research is required in order to make conclusions regarding the diagnostic and/or screening accuracy of WAI technologies for neonates.

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product otoacoustic emissions in hearing screening of high risk neonates. *International Journal of Pediatric Otorhinolaryngology*, *75*(9), 1109-16. doi: 10.1016/j.ijporl.2011.05.026

[42] Van Maanaan, A., Stapells, D.R. (2009). Normal multiple auditory steady-state response thresholds to air-conducted stimuli in infants. *Journal of the American Academy of Audiology*, *20,* 196–207.

[43] Hunter, L.L., Prieve, B.A., Kei, J., Sanford, C.A. (2013). Pediatric applications of wideband acoustic immittance measures. *Ear Hear, 34*(Suppl 1) 36S-42S. doi: 10.1097/AUD.0b013e31829d5158

[44] Kei J., Sanford, C.A., Prieve, B.A., Hunter, L.L. (2013) Wideband acoustic immittance measures: developmental characteristics (0 to 12 months). *Ear Hear*, *34*(Suppl 1) 17S-26S. doi: 10.1097/AUD.0b013e31829db914.

Bottom line: Technologies on the horizon

Evidence from current research does not suggest that genetic testing, multi-frequency ASSR, or WAI are ready for implementation in a universal newborn hearing screening programme. Further research is required to validate and provide further information for test development and validation prior to use within a screening regime.

Screening regimes using AOAE only

Q.2. What are AOAE screening regimes?

Q.3. Where are AOAE screening regimes used?

Q.4. What are the clinical risks of AOAE screening regimes?

Q.5. What are the clinical benefits of AOAE screening regimes?

Q.6. What are the practical implications/considerations of an AOAE screening regime?

Q.7. How does this affect the accuracy of screening?

Q.8. What are the costs per child of the AOAE screen?

Key findings

Benefits

- Screening TEOAEs and DPOAEs typically screen at lower decibel levels (30dBHL) than A-ABR (35 – 40dBnHL).

- A mild hearing impairment can be detected, with limitations due to the pass/refer criteria of three out of four frequencies.

- Fast to administer in ideal conditions.

Risks

- Does not detect ANSD.

- High false positive rate in comparison to other regimes.

- Highest refer rate to audiology in comparison to other regimes.

- Lack of well-researched pass criteria for DPOAEs.

- Mild hearing impairments may be missed with a three out of four frequency pass/refer criteria.

Overview of AOAE regimes

An AOAE screening regime can consist of two types of otoacoustic emissions, the TEOAE and the DPOAE. TEOAEs are elicited using a click stimulus that has a broad frequency range, up to 4 kHz. They can also be evoked using a toneburst stimulus, which has a short duration pure tone, and evokes a response from the same region as the pure tone presented. In contrast, the DPOAEs are elicited using a pair of primary tones. Essentially, TEOAEs and DPOAEs assess the same mechanism within the auditory system, namely the outer hair cell function of the cochlear.

Generally, the main advantages of TEOAE or DPOAE screening regimes are that they are a simple and painless screen, and take only a few minutes to complete. A neonate can be awake during the screen, but must remain settled. Feeding noises and heavy breathing or crying precludes the ability to measure the AOAE. The probe fit is essential and affects the validity of the AOAE screen, requiring screeners to determine the correct size and fit for the ear canal of a neonate.

In a European Consensus Statement (1998), a moderate level of validity for the AOAE screen was reported[45]. The range of sensitivity, from nine studies, was between 50.0 – 98.8%, with an average sensitivity of 87.0%. This means that a proportion of children who have a hearing impairment are not identified using this screen approach. The range of specificity was 41.0 – 92.3%, with an average of 78.6%. Relative to other regimes, this results in higher numbers of children being recalled for either rescreening and/or unnecessary referrals to diagnostic audiology.

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[45] European Consensus Statement on Neonatal Hearing Screening. (1999). *Acta Paediatrics*, *88*(1), 107-8. doi: 10.1111/j.1651-2227.1999.tb01282.x

A risk of the AOAE is that there are no currently agreed upon international calibration standards[46]. Manufacturers calibrate equipment according to their own standards and conduct trials and field research to assess the reliability of their equipment.

TEOAE screening

TEOAEs are often employed as a first stage screen as they are able to detect hearing impairment up to 4 kHz. An advantage of the AOAE screen is that the equipment can screen at 30dBHL and can therefore distinguish normal hearing from a refer result.

The two stage TEOAE regime has been shown to accurately identify hearing impairment at a rate of 0.5%, a result which does not significantly differ to other regimes in terms of overall detection[47].

It is often stated that due to the lower decibel level a mild hearing impairment can be detected. However, most screening equipment is programmed to initiate a pass criteria of three out of four frequencies[48]. Essentially this means, that a child may pass a screen but may actually have a hearing impairment at one of the four frequencies.

An advantage of using TEOAEs in a two stage regime (AOAE followed by an AOAE at a later appointment) is the low false-positive rate of 2.0%, when utilised by professionals in quiet conditions[49]. A TEOAE screen has high sensitivity rates of 100%, with specificity at 92.0%, when conducted in very quiet or sound-proofed conditions. However, very quiet conditions are rarely found in screening venues. Administration of a TEOAE in real world environments, such as a

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[46] Johnson, J.L., White, K.R., Widen, J.E., Gravel, J.S., Vohr, B.R., James, M….Meyer, S. (2005). A multisite study to examine the efficacy of the otoacoustic emission/automated auditory brainstem response newborn hearing screening protocol: introduction and overview of the study. *American Journal of Audiology, 14*(2), S178-85.

[47] Lin, H.C., Shu, M.T., Lee, K.S., Lin, H.Y, Lin, G. (2007). Reducing false positives in newborn hearing screening program: how and why. *Otology and Neurotology*, *28*(6), 788-92.

[48] Kurman, B.L., Gravel, J. (2006). Standards for NHS equipment aka What are we measuring in NHS programs? Retrieved from: http://infanthearing.org/meeting/ehdi2006/presentations/index.html

[49] Hatzopoulos, S., Petruccelli, J., Ciorba, A., Martini, A. (2009). Optimising otoacoustic emission protocols for a UNHS program. *Audiology and Neurotology*, *14*(1), 7-16. doi: 10.1159/000148205

neonatal intensive care unit, significantly reduces the sensitivity and specificity of the screen to 50.0%[50].

Documentation of the false positive rate of TEOAEs in a screening regime ranges from 16.0% to 97.0% of all initial screen refer results[51]. The high false positives may be due to the high incidence of outer ear debris and middle ear fluid in the first hours post-delivery. High first screen false positive rates result in unnecessary rescreen rates of 11.5% and testing at diagnostic audiology at rates of 10.0%[52].

The presence of the OAE is adversely affected by incomplete clearance of vernix caseosa (debris) in the external ear. Research indicates that this results in referral rates of 5.0% to 20.0% when screening is performed during the first 24 hours after birth. Refer rates of a TEOAE screen can reach 12.3% up to 30 hours after birth[53,54] and progressively reduce each day in the first week of life to 1.9% on day seven[55]. For this reason, TEOAE is not a good detector of permanent childhood hearing impairment due to how acutely affected it is by the presence of conductive pathology.

The direct costs incurred for a two stage TEOAE regime have been calculated at US$10.04[56]. This value includes pre-discharge screening and post-discharge follow-up and is higher than any other regime. The literature shows high variances in the costs of screening regimes. This is due to the types of variables included to calculate overall costs. There is only one study which compares all three regimes (AOAE/AOAE, AOAE/A-ABR, A-ABR/A-ABR), however the values for each regime type appear lower than other literature. A recurring theme was that an AOAE regime has higher costs than all others due to the high referral

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[50] Jacobsen, J.T., Jacobsen, C.J. (1994). The effects of noise in transient EOAE in newborn hearing screening. *International Journal of Paediatric Otorhinolaryngology*, *29*(3), 235-248.

[51] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee reference 17, Assessment report. Retrieved from: http://www.msac.gov.au/internet/msac/publishing.nsf/Content/8FD1D98FE64C8A2FCA2575AD0082FD8F/$File/ref17.pdf

[52] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

[53] Kok M.R., Zanten G.A. van, Brocaar M.P. (1993) Click-evoked otoacoustic emissions in 1036 ears of healthy newborns. *Audiology*, *32*(4), 213-24.

[54] Maxon, A.B., White, K.R., Culpepper, B., Vohr, B.R. (1997). Maintaining acceptably low referral rates in TEOAE-based newborn hearing screening programs. *Journal of Communication Disorders*, 30(6), 457-475.

[55] Prieve, B.A., Hancur-Bucci, C.A., Preston, J.L. (2009). Changes in transient-evoked otoacoustic emissions in the first month of life. Ear and Hearing, *30*(3), 330-9.

[56] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

rates. Furthermore, most studies do not assess parental anxiety as a consequence of experiencing a refer result. The single intangible cost of parental anxiety levels has been shown to be higher for this regime than any other[57]. Despite this, the evidence indicates that false-positive hearing screen results do not cause long-term general parental anxiety[58] and that mothers who have the knowledge base to understand the context of a refer result experience much less anxiety[59].

DPOAE screening

DPOAEs are used less frequently than TEOAEs in newborn hearing screening. Although DPOAEs span a greater frequency range and are generally more robust in the presence of electrophysiological noise[60], there are problems in specifying a universally acceptable pass/refer criteria for the screen. A single study identifies a pass/fail criteria, with little identifiable data in the area since[61].

While some evidence suggests that DPOAEs have a pass rate of 77.4%[62], other research has found false-positive rates ranging from 11.0% to 35.0%[63]. One study reported implications for both the cost of consumables required for AOAE screening and parental anxiety[64].

The reason that DPOAEs are not widely implemented is the unavailability of research data specifying the pass criteria to be utilised. Variability in pass and

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[57] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

[58] Van der Ploeg, C.P., Lanting, C.I., Kauffman-de Boer, M.A., Uilenburg, N.N., de Ridder-Sluiter, J.G., Verkerk, P.H. (2008). Examination of long-lasting parental concern after false-positive results of neonatal hearing screening. *Archives of the Disordered Child*, *93*(6), 508-11. doi: 10.1136/adc.2007.129320

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[61] Pelosi, G., Hatzopoulos, S., Chierici, R., Vigi, V., Martini, A. (2000) Distortion product otoacoustic emission (DPOAEs) and newborn hearing screening: a feasibility and performance study. *Acta Otorhinolaryngology Italica*. 20(4), 237-44.

[62] Abdul Wahid, S.N., Daud, M.K., Sidek, D., Abd Rahman, N., Mansor, S., Zakaria, M.N. (2012). The performance of distortion product otoacoustic emissions and automated auditory brainstem response in the same ear of the babies in neonatal unit. *International Journal of Pediatric Otorhinolaryngology*, 76(9), 1366-9. doi: 10.1016/j.ijporl.2012.06.008

[63] Barker, S.E., Lesperance, M.M., Kileny, P.R. (2000). Outcome of newborn hearing screening by ABR compared with four different DPOAE pass criteria. *American Journal of Audiology, 9*(2)*,* 142-148.

[64] Iley, K.L., Addis, R.J. (2000). Impact of technology choice on service provision for universal newborn hearing screening within a busy district hospital. *Journal of Perinatology*, *20*(8 Pt 2), S122-7.

refer rates with differing pass criteria indicates the need for standardisation, particularly when implemented in newborn hearing screening programmes. A renowned expert in OAEs noted that DPOAEs are collected over a much wider range of stimulus intensities and a range of stimulus intensity ratios[65]. This strongly affects sensitivity and specificity, so the volume of data on any particular stimulus combination is small. While there is validity in performing DPOAEs, it has been stated that 'There are no sufficiently well-established criteria for DPOAEs that would allow DPOAEs to replace TEOAEs in Newborn Hearing Screening Programs without significant risk of a change in current specificity and sensitivity'[66]. For this reason, this technique is not ideal for newborn hearing screening programmes until such data is available. The interpretation of DPOAE results for a neonate should include age-related differences in DPOAE pass rates for low-frequency and high-frequency information[67].

Implications for detection of ANSD

Detection of ANSD disorders utilising AOAE regimes is not possible. The OAE indicates cochlear function only, at the level of the outer hair cell. The OAE does not assess auditory function beyond the level of the cochlear and for this reason ANSD cannot be detected. Approximately 10.0% of all children diagnosed with a permanent hearing impairment have ANSD[68, 69]. While NICU admission is a risk factor for ANSD, ANSD can also occur within the well-baby population and in neonates who do not have a hearing impairment, this is possibly due to an autosomal recessive inherited genetic mutation[70, 71].

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[65] Kemp, D. (2011). Use of DPOAEs in assessment following screening. NHSP Clinical Group. Retrieved from: http://hearing.screening.nhs.uk/searchwebsite.php?searchstring=tag:audiology

[66] ibid.

[67] Zhang Z., Jiang, Z.D. (2007). Distortion product otoacoustic emissions during the first year in term infants: a longitudinal study. *Brain Development*, *29*(6), 346-51.

[68] Sininger, Y.S. (2002). Identification of auditory neuropathy in infants and children. *Seminars in Hearing*, *23*(3), 193-200. doi: 10.1055/s-2002-34456

[69] Uus, K., Bamford, J. (2006). Ages of interventions for identified babies and profile of cases. *Pediatrics*, *117*(5), e887-e893. doi:10.1542/peds.2005-1064

[70] Varga, R., Kelley, P.M., Keats, B.J., Starr, A., Leal, S.M., Cohn, E., Kimberling, W.J. (2003). Non-syndromic recessive auditory neuropathy is the result of mutations in the otoferlin (OTOF) gene. *Journal of Medical Genetics*, *40*(1), 45-50.

[71] Delmaghani, S., del Castillo, F.J., Michel, V., Leibovici, M., Aghaie, A., Ron, U., Petit, C. (2006) Mutations in the gene encoding pejvakin, a newly identified protein of the afferent auditory pathway, cause DFNB59 auditory neuropathy. *National Genetics, 38*(7), 770-8.

ANSD accounts for approximately 1.0% of newborn hearing impairment, affecting approximately 0.5% of neonates with sensorineural hearing impairment[72]. One researcher found that 24.0% of a cohort of NICU children who had a hearing impairment had ANSD profiles[73]. Another found an ANSD prevalence of 0.2% in infants at risk of hearing impairment, as defined by NICU admission, and 11.0% in children with permanent hearing impairment[74]. Detection for this condition relies on a screen of the neural pathway.

Many international hearing screening programmes, including England, New Zealand and the Netherlands, are not able to detect ANSD in healthy babies due to the initial AOAE screen. Sole reliance on an initial AOAE screen, with discharge based on a pass result, means that well babies with ANSD are not able to be identified. The consequences of ANSD include poor auditory cortical development and function, hence early identification is important for supporting optimal child development[75].

Bottom line: Screening regimes using AOAE only

AOAE regimes have a high false positive rate and therefore a high rate of referral, in comparison to other regimes. AOAE regimes do not detect ANSD.

Screening regimes using a combination of AOAE and A-ABR

Q.9. What are AOAE/A-ABR screening regimes?

Q.10. Where are AOAE/A-ABR screening regimes used?

Q.11. What are the clinical risks of AOAE/A-ABR screening regimes?

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[72] Davis, H. (1976). Principles of electric response audiometry. *Annals of Otology, Rhinology and Laryngology*, *85* Suppl (3 Pt3), 1-96.

[73] Berg, A.L., Spitzer, J.B., Towers, H.M., Bartosiewicz, C., & Diamond, B.E. (2005). Newborn hearing screening in the NICU: profile of failed auditory brainstem response/passed otoacoustic emission. *Pediatrics*, *116*(4), 933-8.

[74] Rance, G., Beer, D.E., Cone-Wesson, B., Shepherd, R.K., Dowell, R.C., King, A.M., Clark, G.M. (1999). Clinical findings for a group of infants and young children with auditory neuropathy. *Ear and Hearing*, *20*(3), 238-52.

[75] Sharma, A., Cardon, G., Henion, K., Roland, P. (2011). Cortical maturation and behavioral outcomes in children with auditory neuropathy spectrum disorder. *International Journal of Audiology*, 50: 98-106. doi: 10.3109/14992027.2010.542492

Q.12. What are the clinical benefits of AOAE/A-ABR screening regimes?

Q.13. What are the practical implications/considerations of an AOAE/A-ABR screening regime?

Q.14. How does this affect the accuracy of screening?

Q.15. What are the costs per child of the AOAE/A-ABR screen?

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Key findings

Benefits

- High rates of specificity and sensitivity compared to AOAE regimes.

Risks

- The high refer rate of the AOAE screen requires babies to return for an A-ABR, which the baby may have initially passed if an A-ABR/A-ABR regime was implemented.

- A lower refer rate to audiology compared to the AOAE/AOAE regime.

- Does not detect ANSD.

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Overview

This section addresses the use of screening regimes using a combination of AOAE and A-ABR, not the use of each screen type in isolation. Given overlaps in information relevant to the AOAE regime and the AOAE/A-ABR regime, there is some duplication of information within this section and/or reference to the AOAE regime discussed above.

An AOAE/A-ABR hearing screening regime commences with an AOAE screen. The OAE is usually a TEOAE, but may be either a TEOAE or DPOAE. According to the literature it is completed between day one and day 24 post birth.

There are three different approaches to the AOAE/A-ABR regime. A one-step regime involves an initial AOAE screen immediately followed by an A-ABR within a single screening appointment. A two-step regime involves an AOAE screen, and in the event of a refer result the baby is screened with A-ABR at a later date. A three step regime involves an AOAE immediately followed by an A-ABR within

the same screening appointment. If a refer result is obtained on both screens then a second A-ABR is completed at a later date. In the UK, NZ and the Netherlands, a three-step process is used. For example, if both the baby's ears receive a pass result on the initial TEOAE screen the baby is discharged. If the baby refers on one or both ears, the second TEOAE screen is repeated within seven days. For those who refer on the second TEOAE, an A-ABR screen follows within five weeks of the second TEOAE screen. A refer result on the A-ABR results in immediate referral for audiological assessment.

For the most part, the UK have implemented hearing screening regimes, involving TEOAE/A-ABR. A large retrospective analysis of the programme shows high rates of specificity and sensitivity, utilising this regime, with respective rates as high as 92.0% and 98.0%[76]. The refer rate for a TEOAE/A-ABR regime is reported at 1.6%[77]. The high rate of referrals from the initial AOAE screening requires a large portion of babies to subsequently attend A-ABR screening, which they may have passed had they been screened with A-ABR at the outset[78]. The TEOAE/A-ABR screening regime has a detection rate of 0.3%, in the context of accurate identification of congenital hearing impairment[79]. The same study which detailed TEOAE screening programmes as costing US $10.04 per child, identified a lower cost of US $8.60 for TEOAE/A-ABR screening[80]. This difference is due to the lower referral rate achieved by combining the two screen types.

Clinically there are many variables of this regime which have already been addressed within the OAE section. For example, this regime is also susceptible to vernix in the ear canal, external noise and physiological noise of the neonate (sucking, heavy breathing etc.).

Combining AOAE and A-ABR technology in a single device enables an easy switch from one screen type to the other[81]. This is particularly useful if the protocol requires an immediate transition to A-ABR following an AOAE refer result. This

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[76] Bamford, J., Uus, K., Davis, A. (2005). Screening for hearing loss in childhood: Issues, evidence and current approaches in the UK. *Journal of Medical Screening, 12*(3), 119-124.

[77] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

[78] ibid.

[79] ibid.

[80] ibid.

[81] Meier, S., Narabayashi, O., Probst, R., Schmuziger, N. (2004). Comparison of currently available devices designed for newborn hearing screening using automated auditory brainstem and/or otoacoustic emission measurements. *International Journal of Pediatric Otorhinolaryngology, 68*(7), 927-34.

quickly identifies children who pass screening and can therefore be discharged or immediately identifies children requiring referral to diagnostic audiology without the need for further screening stages. The result is reduced non-attendance rates and increased capture and completion rates. This can be particularly beneficial in regional and rural areas.

A-ABR screening

The A-ABR is a measure of total auditory function. The screen is painless and fast. The screen requires the attachment of electrodes to the infant's scalp to obtain a response (waveform). Attaching electrodes needs to be performed carefully, with screeners preparing the newborn's skin with cleaning. Poor electrode attachment can lead to high impedances, which contaminate screen results. Neonates are best assessed sleeping or very settled. A wakeful baby may be assessed in the mother's arms, if settled. A crying baby affects the EEG trace on the A-ABR, and in this circumstance the screen will time out with no result. Screen times vary based on the state of the infant and the type of equipment used.

Most newborn hearing screening programmes work to the target of identifying children who have a moderate hearing impairment or greater. An A-ABR in most instances is set at the 35 – 40dBnHL and therefore aims to identify impairments greater than 35 – 40dBnHL. A possible consequence of the AOAE/A-ABR regime is that when a mild impairment is detected by an AOAE refer result, the infant is likely to pass the subsequent A-ABR screen[82, 83].

Implications for detection of ANSD

An AOAE/A-ABR regime, similar to the AOAE screen alone, will not identify babies who present with ANSD. This is because the AOAE screen at the commencement of this regime will not detect neonates with normal hearing who have ANSD (see OAEs and ANSD). Neonates who pass the AOAE screen do not receive further audiological diagnostic assessment that would enable the identification of ANSD.

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[82] Kurman, B.L., Gravel, J. (2006). Standards for NHS equipment aka What are we measuring in NHS programs? Retrieved from: http://infanthearing.org/meeting/ehdi2006/presentations/index.html

[83] Johnson, J.L., White, K.R., Widen, J.E., Gravel, J.S., James, M., Kennalley, T., Maxon, A.B. … Holstrum, J. (2005). A multi-center evaluation of how many infants with permanent hearing loss pass a two-stage otoacoustic emissions/automated auditory brainstem response newborn hearing screening protocol. *Pediatrics. 116*(3), 663-72.

Bottom line: Screening regimes using a combination of AOAE and A-ABR

AOAE/A-ABR regimes have a much lower false positive rate and therefore a lower rate of referral, in comparison to AOAE regimes. AOAE/A-ABR regimes do not detect ANSD, and may not detect mild hearing impairments.

Screening regimes using A-ABR only

Q.16. What are A-ABR/A-ABR screening regimes?

Q.17. Where are A-ABR/A-ABR screening regimes used?

Q.18. What are the clinical risks of A-ABR/A-ABR screening regimes?

Q.19. What are the clinical benefits of A-ABR/A-ABR screening regimes?

Q.20. What are the practical implications/considerations of an A-ABR/A-ABR screening regime?

Q.21. How does this affect the accuracy of screening?

Q.22. What are the costs per child of the A-ABR/A-ABR screen?

Key findings

Benefits

- Low refer rates (0.8%) compared to AOAE and AOAE/A-ABR regimes.
- Capacity to detect ANSD.
- Highest rates of sensitivity and specificity compared to other regimes.

Risks

- Mild impairments may remain undetected (this can be rectified with an alternate manufacturer setting).

Overview

For completeness, this section duplicates some previous information.

In most programmes, A-ABR regimes involve two stages. A neonate with a refer result on the first A-ABR receives a second A-ABR screen at a later time. The advantage of the A-ABR/A-ABR screening regime is the reduction in refer rates in comparison to the AOAE and the AOAE/A-ABR regimes. Protocols for the timing of the first and second screen vary across programmes and are typically informed by local logistics and optimising screening capture and completion rates for a programme as a whole and to respond to the circumstances of individual babies and their families. For babies born in hospital, in keeping with manufacturer guidelines, the first A-ABR typically occurs prior to discharge at a minimum of four hours of age. If necessary a second screen can occur within 24 hours, but sometimes occurs days or up to a couple of weeks later. In some locations first and second screens occur sooner than these timelines to maximise screening commencement and completion for infants who are likely to be discharged and may experience challenges returning for community screening. It is optimal for screening to be completed by one month corrected age to enhance the ease of completing the screen. Another reason is to allow referral for audiological assessment and diagnosis within the first three months of life when a neonate has longer and deeper episodes of sleep, which is necessary for effective diagnostic assessment.

A-ABR regimes, where A-ABR is the only screening approach used, are able to identify the presence of ANSD more effectively than any other regime[84].

Sensitivity and specificity rates for A-ABR vary slightly across equipment but are essentially much higher than AOAEs. Sensitivity is currently reported at 100% (most likely in ideal conditions) with specificity at 97.0%[85, 86, 87]. The low false

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[84] Tognola, G., Paglioalonga, A., Grandori, F. (2010). Improved newborn hearing screening: Future directions. C. Driscoll, B. McPherson (Eds.), *Newborn hearing screening: The complete perspective*. USA: Plural Publishing.

[85] van den Berg et al. (2010). MB 11 BERAphone hearing screening compared to ALGO portable in a Dutch NICU: A pilot study.

[86] Melagrana et al. (2007). MB 11 BERAphone and auditory brainstem response in newborns at audiologic risk: Comparison of results.

[87] Cebulla, Stürzebecher, (2013). Detectability of newborn chirp-evoked ABR in the frequency domain at different stimulus rates.

positive rate (0.3%) in A-ABR regimes, is a distinct advantage[88, 89], as is the lower refer rate (0.8%) than an AOAE alone or AOAE/A-ABR[90].

A-ABR is reportedly the gold standard for universal newborn hearing screening[91, 92]. This is due to lower costs per infant screened from post-discharge screening to diagnostic evaluation (US$45.85 for A-ABR versus US$58.07 for TEOAE), lower referral rates (4.0% for A-ABR compared to 15.0% for TEOAE), and the capacity to detect ANSD[93]. A more recent study identified A-ABR/A-ABR regimes as the least expensive with a total direct cost of US$7.33[94]. The reason for the large disparity between the costs identified between the two studies mentioned above is unknown. It may reflect differing methodologies, cheaper costs of consumables, allocation of equipment costs over the equipment's life span, or lower labour costs in one study compared to the other. A consistent finding is that overall costs of an A-ABR/A-ABR programme are lower.

It is usual for A-ABR/A-ABR regimes to work to the target of identifying children who have a moderate permanent hearing impairment. For this reason the A-ABR screening equipment is set at a 35 – 40dBnHL screen. This means that neonates with a mild hearing impairment in either ear may not be detected by A-ABR/A-ABR regimes. On some equipment, the decibel level is changeable (either on the equipment or by the distributor). Hence, for programmes wishing to detect mild hearing impairment a lower decibel stimulus level can be utilised. Such a change would probably increase refer rates to audiology and increase the identification of children with a permanent hearing impairment. Anecdotally, children who have a mild hearing impairment at birth may experience progression in their hearing impairment at a later stage. It can be speculated that earlier identification of a hearing impairment for these children may have developmental advantages. In effect, the lowering of the stimulus level may lead to less late identified

[88] Korres, S., Nikolopoulos, T.P., Peraki, E.E., Tsiakou, M., Karakitsou, M., Apostolopoulos, N….Ferekidis, E. (2008). Outcomes and efficacy of newborn hearing screening: Strengths and weaknesses (success or failure?). *Laryngoscope*, *118*, 1253-1256. doi: 10.1097/MLG.0b013e31816d726c

[89] Guastini et al. (2010). Evaluation of an automated auditory brainstem response in a multi-stage infant hearing screen.

[90] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

[91] Lemons, J., Fanaroff, A., Stewart, E.J., Bentkover, J.D., Murray, G., Diefendorf, A. (2002). Newborn hearing screening: Costs of establishing a program. *Journal of Perinatalogy*, *22*(2), 120-124.

[92] Joint Committee on Infant Hearing. (2007). Year 2007 position statement.

[93] Lemons et al. (2002). Newborn hearing screening: Costs of establishing a program.

[94] Lin et al. (2007). Reducing false positives in newborn hearing screening program: how and why.

permanent childhood hearing impairment arising in the context of progression of an initially mild congenital hearing impairment.

A risk of the A-ABR/A-ABR regime is that there are no currently agreed upon international calibration standards for the A-ABR[95], this is not dissimilar to the AOAE, and is currently being addressed by the UK newborn hearing screening programme. Currently, manufacturers calibrate equipment according to their own standards utilising clinical research and field trials.

Another risk is that the delivery of the A-ABR click stimulus (usually at 35 – 40dBnHL) may vary at the level of the tympanic membrane (ear drum) due to differing physical volumes of the neonate's closed ear canal. Essentially the actual sound pressure level may vary significantly between neonates and may create slight fluctuations in the sound level presented to the ear[96]. This may be a plausible explanation as to why some neonates may pass the A-ABR while referring on an OAE, despite having the same stimulus level of presentation. Variations in stimulus levels are reported with regard to the use of insert earphones[97]. It is important to note that when a level is reported in dBnHL, the reference is often an adult ear. This level means that impairments at or slightly worse than 35dBnHL may not be detected, this is due to a variation of 5 – 10dB to account for the smaller size in ear canal volume of the infant. Not all A-ABR devices utilise insert earphones, reverting to ear cups and/or muffs to overcome this issue.

The A-ABR has specific requirements for skin preparation in order to adhere the electrodes to the neonate's head. Should electrode placement not be optimal the screen performance can be significantly affected, resulting in a refer result. Another practical consideration, is that some equipment will be affected by closely located electrical equipment. While the A-ABR will have filter settings to this electrical interference, sometimes it is not possible.

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[95] Johnson et al. (2005). A multisite study to examine the efficacy of the otoacoustic emission/automated auditory brainstem response newborn hearing screening protocol: introduction and overview of the study.

[96] Sutton, G., Lightfoot, G. (2013). Newborn hearing screening and assessment: Guidance for auditory brainstem response testing in babies, v2.1. *NHS Antenatal Newborn Hearing Screening Programmes*. Retrieved from: http://hearing.screening.nhs.uk/getdata.php?id=19345

[97] Sutton, G., & Lightfoot, G. (2013). Newborn hearing screening and assessment: Guidance for auditory brainstem response testing in babies, v2.1. NHS Antenatal Newborn Hearing Screening Programmes. Retrieved from: http://hearing.screening.nhs.uk/getdata.php?id=19345

Research suggests that the accuracy of the A-ABR is influenced by the experience of the assessor[98]. This outcome potentially relates to the ability of an experienced assessor to set up, initiate and trouble shoot all aspects of the screen. Evidence suggests that similar results would also arise for an AOAE[99].

Auditory brainstem response screening and auditory neuropathy spectrum disorder

In 2007, the JCIH recommended the exclusive use of A-ABR screening in NICU populations[100]. The reason for this recommendation relates exclusively to the detection of ANSD. Babies who refer from A-ABR screening are referred for audiological assessment. At this point they then receive more detailed assessment to determine the presence and/or absence of ANSD.

Bottom line: Screening regimes using A-ABR alone

A-ABR/A-ABR regimes have the lowest false positive rate and the lowest rate of referral, in comparison to AOAE and AOAE/A-ABR regimes. A-ABR/A-ABR regimes also identify ANSD. A-ABR/A-ABR regimes typically do not identify mild impairments due to the target condition of moderate or greater hearing impairment, but this can be adjusted in most equipment in consultation with manufacturers, if desired.

CE chirp stimulus

Q.23. How does the CE chirp A-ABR screening regime differ clinically from the A-ABR screening regime?

Q.24. How does screening technology utilise the CE chirp A-ABR?

Q.25. What clinical implications does emerging screening technology have for the screening regime?

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[98] Guastini et al. (2010). Evaluation of an automated auditory brainstem response in a multi-stage infant hearing screen.

[99] Dort, J.C., Tobolski, C., Brown, D. (2000). Screening strategies for neonatal hearing loss: which test is best? *Journal of Otolaryngology, 29*(4), 206-10.

[100] Joint Committee on Infant Hearing. (2007). Year 2007 position statement: Principles and guidelines for early hearing detection and intervention programs.

Q.26. What clinical implications does the CE chirp A-ABR have on follow-up services such as audiology?

> **Key findings**
>
> In the literature, the CE chirp stimuli has emerged as an effective stimulus for screening, given:
>
> - the speed at which it collects information,
>
> - the potential cost-savings in the context of no consumables being required with some CE chirp A-ABR equipment, and
>
> - the potential future use of the equipment for detection of frequency specific information (multi-frequency ASSRs). Further research in this area is warranted.

While the CE chirp is sometimes perceived as emerging technology clinically, it has been well researched, evidenced and validated since 2010. The outcomes of a CE chirp ASSR correlate well with behavioural audiograms, supporting reliability. For this reason, the CE chirp is no longer an emerging technique and will be presented in this review as an established stimulus, now utilised in newborn screening equipment. The following discussion addresses the CE chirp in comparison to the regular click A-ABR.

Typically an A-ABR screen utilises a 'click' stimulus. This stimulus is broad spectrum and presented at a slow repetition rate. The CE chirp is designed to enhance neural synchronicity. The CE chirp uses an input compensation method. This means that the stimulus for high frequencies is delayed relative to the low frequencies, which models the traveling wave of sound through the basilar membrane of the cochlear. This maintains a larger activated quantity of hair cells in the response. In creating the delay, all the components arrive at the cochlear at the same time. This creates a significantly larger amplitude and/or waveform in the ABR, which allows easier identification of hearing thresholds.

Simply, the CE chirp optimises the stimulus which is delivered. This means that the energy elicited from the stimulus reaches all regions of the cochlea at approximately the same time, creating much larger amplitude response.

There are several diagnostic clinical benefits that the CE chirp stimuli provides that renders it different from other ASSRs. Firstly, CE chirps have a significantly

larger amplitude in comparison to ABRs (typically double the response) which makes the test time decrease and the response much easier to detect. Secondly, when performing screening, because of the larger amplitude, thresholds can be detected at much lower stimulus intensity levels. Typically the responses are especially close to behavioural thresholds[101]. Thirdly, the CE chirp is faster to recruit information and easier to detect waveform at thresholds, particularly at levels that are low in intensity (i.e. quieter), compared to click ABR. Fourthly, it is suggested that the CE chirp ABR also has a higher specificity rate compared to click ABR[102]. To date, screening shows an average detection rate of 15-30 seconds when recording CE chirp A-ABR bilaterally, significantly faster than click A-ABR systems which are approximately 3-5 minutes.

Although the CE chirp is a type of ASSR, manufacturers consistently refer to it as an A-ABR. This may be due to the fact that, historically, some aspect of ASSRs have poor correlation to behavioural audiograms. This has given the ASSR a poor reputation. The CE chirp though, due to the large amplitudes that it elicits, has excellent correlation to behavioural audiograms[103]. Given the short screen time, ease of use, and high sensitivity and specificity, it is a good tool for newborn hearing screening[104, 105, 106, 107, 108, 109, 110].

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[101] Maloff, E.S., Hood, L.J. (2014). A comparison of auditory brain stem responses elicited by click and chirp stimuli in adults with normal hearing and sensory hearing loss. *Ear Hear*, *35*(2), 271-82. doi: 10.1097/AUD.0b013e3182a99cf2

[102] Cebulla, Sturzebecher, (2013). Detectability of newborn chirp evoked ABR in the frequency domain at different stimulus rates.

[103] Elberling, C., Don, M. (2008). Auditory brainstem responses to a chirp stimulus designed from derived-band latencies in normal-hearing subjects. *Journal of the Acoustical Society of America, 124*(5), 3022-37. doi: 10.1121/1.2990709

[104] Melagrana et al. (2007). MB 11 BERAphone and auditory brainstem response in newborns at audiologic risk: Comparison of results.

[105] Cebulla et al. (2014). Sensitivity of ABR based newborn screening with the MB 11 BERAphone®.

[106] White et al. (2008). A comparative study of the MB 11 BERAphone and ABAER automated auditory brainstem response newborn hearing screening equipment.

[107] Soares et al. (2014). Hearing screening for Japanese children and young adults using the automated auditory brainstem response.

[108] Augustine et al. (2014). Neonatal hearing screening: Experience from a tertiary care hospital in Southern India.

[109] Guastini et al. (2010). Evaluation of an automated auditory brainstem response in a multi-stage infant hearing screening.

[110] Cobb, Stuart (2014). Test-retest reliability of auditory brainstem responses to chirp stimuli in newborns.

The implementation of a programme utilising the CE chirp, would not differ to that of an A-ABR regime. Ultimately, the CE chirp elicits an auditory brainstem response. However, the high rate of the stimulus presentation and the algorithmic analysis of the response is what differentiates it from an A-ABR and defines it as an ASSR.

There are a growing number of devices using the CE chirp technology, with one device containing no consumables.

Bottom line: CE chirp A-ABR

CE chirp ASSR/A-ABR is a reliable and validated screening tool.

CE chirp A-ABR regimes have similarities to A-ABR regimes.

CHAPTER 5 – Findings: Evidence from international practice

Introduction

Nine programmes contributed to the review of selected international programmes. Seven programmes contributed through an interview as well as the provision of data, one programme contributed to an interview only, and a final programme contributed data only.

A summary of findings are presented in Table 2 on the following page. Data for the UNHSEIP is included in the final column for purposes of comparison. A discussion of interview and data collection findings follow;

*Table 2.*Programme data provided by participating international programmes.

Details	Programme 1	Programme 2	Programme 3	Programme 4	Programme 5	Programme 6	Programme 7	Programme 8	Programme 9	UNHSEIP
Data collection start date	01/01/2012	01/01/2012	01/01/2012	22/11/2011	01/01/2012	01/04/2012	01/01/2012	01/01/2012		01/04/2012
Data collection end date	31/12/2012	31/12/2012	31/12/2012	31/01/2013	31/12/2012	31/03/2013	31/12/2012	31/03/2012	not provided	31/12/2012
Regime – well baby	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR / A-ABR	Not designated Largely TEOAE / TEOAE / TEOAE Some tertiary hospitals use A-ABR / A-ABR	TEOAE / TEOAE / A-ABR	TEOAE / TEOAE, A-ABR	TEOAE / A-ABR	DPOAE, A-ABR / A-ABR	DPOAE, A-ABR / A-ABR
Regime – NICU	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR	AOAE + A-ABR	A-ABR	A-ABR / A-ABR	A-ABR / A-ABR	A-ABR
Minimum time for screening after birth	6 hours	4 hours	No minimum age	(Community screening)	Variable	Not provided	No minimum age	6 hours	Not provided	Variable – up to 24 hours
Workforce	Nurses with dedicated shifts	Dedicated screeners	Dedicated screeners	Nurses within existing preventative health programme	Nurses Some privately contracted dedicated screeners	Dedicated screeners	Midwives Dedicated screeners Community nurses in existing service	Dedicated screeners Audiometric technicians	Dedicated screeners Audiometric technicians and midwives in remote areas	Dedicated screeners
Device/s used at time of interview	ALGO 3, ALGO 3i	ALGO 3i	MAICO MB 11 BERAphone	MAICO MB 11 Classic	AuDx[A], Biologic[B], ABaer[B]	not provided	Echocheck[A], AccuScreen[A], Otoport[A], ALGO 3i[B]	AccuScreen[A] Biologic[B]	AccuScreen[A,B]	AccuScreen[A,B], MAICO MB 11 BERAphone[B], Natus ALGO 3i [B]
Device/s used during period of data collection	as above	as above	AccuScreen	ALGO 3i	as above	not provided	as above	as above	as above	as above
Live births	63,441	77,499	6,078	68,533	65,600	684,768	20,928	44,061		46,957
% eligible	99.5%	99.5%	97.9%	99.8%	99. 7%	99.6%	99.8%	99.7%		not available
% declined	0.2%	0.3%	2.5%	1.0%	0.2%	0.1%	0.1%	0.4%		incomplete data
% completed	99.5%	99.4%	97.6%	96.8%	98.2%	99.1%	96.5%	96.8%		83.0%[F]
% completed <30 days	96.5%	97.8%	88.9%	73.3%	not available	97.2%	92.1%	not available	not provided	91.9%

Details	Programme 1	Programme 2	Programme 3	Programme 4	Programme 5	Programme 6	Programme 7	Programme 8	Programme 9	UNHSEIP
% inpatient screening	92.7%	90.0%	~ 90.0%	3.2%	not provided	67.8%	92.5%	80.5%		not available
% 1st screen refer	6.3%	not available[C]	not available[C]	1.3%	not available[D]	23.5%	18.4%	8.8%		~ 15.0%
% audiology refer	0.9%	0.9%	1.4%	0.5%	0.9%	2.4%	1.1%	1.6%		1.7%
% targeted f/u refer	2.3%	not available	5.6%	not available	not available	1.1%	0.5%	3.7%		5.4%
% PCHI[E] – bilat mod or > in better ear	0.1%	0.1%	0.1%	0.1%	not provided	0.1%	0.1%	0.1%		incomplete data
% PCHI – bilat or unilat, mod or > in better ear	0.2%	0.1%	0.1%	0.2%	0.2%	0.2%	0.2%	0.2%		incomplete data

[A] For AOAE screening, [B] For A-ABR screening, [C] Programme data systems do not provide first screen refer data, data, [D] Refer rate at discharge from hospital: 3.7% (includes some children who have had a single AOAE and some who have had two AOAEs, [E] PCHI = permanent childhood hearing impairment, [F] Calculated on live births rather than eligible births

Regime selection

As detailed in Table 2 above, of the nine programmes that participated, for well babies four programmes used an A-ABR/A-ABR regime and four others used a combination of AOAE and A-ABR, either as:

- TEOAE/TEOAE/A-ABR,

- TEOAE/TEOAE, A-ABR,

- DPOAE, A-ABR/A-ABR, or

- TEOAE/A-ABR.

Programme 5 had no prescribed protocol for well babies, however, TEOAE/TEOAE/+ or - TEOAE was the regime used for well babies in most hospitals. Three screens were completed if two of the AOAEs had occurred prior to discharge. If only one AOAE was completed prior to discharge the regime involved referral to diagnostic audiology immediately after the second AOAE.

On the most part, programmes using a regime combining AOAE and A-ABR selected this regime on the assumption AOAE screening is quicker and therefore more cost efficient than A-ABR. These programmes all expressed the opinion current evidence regarding the incidence of ANSD does not warrant changing to an A-ABR/A-ABR regime given the cost and change process involved for an as yet ill-defined outcome.

Programmes using a two stage A-ABR regime for well babies made this selection based on the lower first screen refer rate; the capacity to identify ANSD; the simplicity of the regime and the benefits this brings to many elements of the programme; and the reduced impact noise has on the screening process.

Given the large rural population served by Programme 5 (TEOAE/TEOAE/+ or - TEOAE), with many hospitals having small birth numbers and long distances to diagnostic audiology services, an AOAE-only regime is used. This reduces capital outlay on equipment given the lower cost of AOAE devices and that only a single device is needed.

None of the programmes using a two stage A-ABR regime for well babies were considering changing the use of A-ABR as the basis of their programme. Programme 3 (A-ABR/A-ABR) is considering use of a third A-ABR screen for children in rural areas to reduce the amount of travel for families and audiology services.

Regime changes being considered by programmes using AOAE and A-ABR include:

- A possible change to A-ABR/A-ABR by Programme 7 (TEOAE/TEOAE, A-ABR). This is being considered in the context of the recent introduction of national standards for newborn hearing screening specifying A-ABR/A-ABR screening as best practice in newborn hearing screening and to align with other programmes across the country. Although mild hearing loss is acknowledged to be outside the target condition of the programme, Programme 7 currently carries out diagnostic assessment at 12 – 15 months for infants who refer on two AOAEs. This results in identification of mild hearing loss in approximately seven children each year. A change to a two stage A-ABR regime will remove this opportunity to identify these children.

- A possible change to a single screening approach for low volume screeners by Programme 8 (TEOAE/A-ABR).

- Programme 9 (DPOAE, A-ABR/A-ABR), where universal screening is only just about to begin, noted that if the cost of screening using A-ABR/A-ABR had been less than or equivalent to AOAE/A-ABR/A-ABR at the time they decided upon their regime, an A-ABR/A-ABR regime would have been selected.

All programmes use A-ABR to screen NICU and other at risk infants:

- Programme 5 (TEOAE/TEOAE/+ or - TEOAE) and Programme 7 (TEOAE/TEOAE, A-ABR) use a single A-ABR,

- Programme 6 (TEOAE/TEOAE/A-ABR) use A-ABR in combination with AOAE, and

- all other programmes use a two stage A-ABR.

Screening timing and protocols

There is significant variability across the programmes regarding the timing of first and second screens as well as other screening processes. Rationales for procedures are informed by local context, perspectives and experiences.

All programmes recognised the value of screening infants after 24 hours of age. However, this was balanced with the knowledge that screening infants while they are still in hospital reduces the burden on families to return for outpatient

appointments; is the most efficient use of screening resources to achieve optimal coverage rates; reduces the age at which infants are referred to audiology and therefore facilitates more efficient diagnostic processes and earlier engagement with early intervention services; and supports achievement of programme targets.

In this context, Programme 3 (A-ABR/A-ABR) and Programme 7 (TEOAE/TEOAE, A-ABR) have no minimum screening age. Programme 2 (A-ABR/A-ABR) screens from four hours and Programme 1 (A-ABR/A-ABR) screens from six hours, although opportunistic screening is also carried out to capture infants who may be difficult to engage for screening at a later time (e.g. infants returning to remote communities). Programme 8 (TEOAE/A-ABR) does not screen infants under six hours and for infants over 16 hours the AOAE and A-ABR are undertaken back-to-back to remove the need for an additional screening stage. This review identified no literature regarding the minimum age to screen a newborn baby. It is acknowledged that family preferences, and operational considerations, play a role in the completion of a screen soon after birth.

Information was not collected from all programmes regarding the approach to 'rescreens'. In Programme 8 (TEOAE/A-ABR), every screen is fully completed, with a result obtained from both ears, and counted as a screen, and in Programme 2 (A-ABR/A-ABR) one 'technical fail' is allowed for each screen attempt, but any refer result is considered valid and the screen is not repeated.

Screening devices

A range of screening devices are used by different programmes. In some programmes, different sites use different devices. In others, the same device is used universally. Details are presented in Table 2 above.

Two programmes have recently made changes to their devices. Both Programme 4 (A-ABR/A-ABR) and Programme 3 (A-ABR/A-ABR) now use a MAICO MB 11 device, using a CE chirp stimulus.

Programme 4 is using the MAICO MB 11 Classic. This choice was made on the basis that the device screens both ears simultaneously, in contrast to the MAICO MB 11 BERAphone which requires screening of one ear at a time. Programme 4 expressed concerned about babies waking when changing from one ear to the other. Programme 4 indicated that the need to use consumables was a disadvantage.

Programme 3 began using the MAICO BERAphone in early 2013. This decision was made based on the savings that would be realised through not needing consumables, whilst retaining the same level of screening rigour. At the time of transitioning to the MAICO MB 11 BERAphone there was a temporary increase in refer rates for a few months and some screeners experienced some initial difficulties with the change. These issues have now resolved and the programme sees it as having been a worthwhile change.

Inpatient, outpatient and community screening

Seven of the eight programmes interviewed focussed on achieving as much inpatient screening as possible and had specific mechanisms in place to contribute to this, including:

- early screening, particularly in the context of early discharge (including screening sooner after birth than manufacturer's recommendations in some circumstances)

- inclusion of screening on the maternity pathway

- inclusion of screening on the discharge plan

- ensuring screening occurs seven days a week and on public holidays, at least in larger hospitals

- planning of the length of screening shifts to achieve daily coverage of inpatient facilities within available resources (e.g. four hour and six hour shifts).

Programmes tended to have more community screening (and typically lower capture rates) in regional areas where it is less viable to have screeners and equipment located at each birthing centre. In some small communities, outpatient clinics are the only screening option.

In Programme 1 (A-ABR/A-ABR), some community screening is carried out as part of extended midwifery services, in collaboration with postnatal check-up appointments. This provides a more coordinated service and reduces the burden on families.

Programme 3 (A-ABR/A-ABR) noted that the younger a baby is at the time of a scheduled outpatient screening appointment the more likely families are to attend. Programme 5 (TEOAE/TEOAE/+ or - TEOAE) indicated that families

leave hospital with an outpatient screening appointment they are more likely to attend than if this is coordinated later.

Programme 4 (A-ABR/A-ABR) was the one programme using a universal community screening approach. In the context of a comprehensive community based preventative health care service for 0 – 3 year olds, 96.8% of screening occurs in the community with a screening completion rate of 96.8%. Programme 4's first refer rate of 1.3% and refer rate to audiology of 0.5% are the lowest of all programmes that provided data. In Programme 7 (TEOAE/TEOAE, A-ABR), although the focus is on initial inpatient screening by midwives and dedicated screening staff, where necessary, community based child health nurses provide follow-up screening post-discharge.

Time and cost

Although specific data on cost was not provided by programmes, variables relating to cost influenced the current design or are likely to influence future plans of programmes. Examples include:

- Programme 3's (A-ABR/A-ABR) transition to the MAICO BERAphone given the cost savings resulting from no longer needing consumables would cover the cost of the device within two years and evidence indicated equivalent performance.

- Use of CE chirp A-ABR by Programme 3 (A-ABR/A-ABR) and Programme 4 (A-ABR/A-ABR) significantly reduced the time to carry out an A-ABR screen.

- Programme 5's (TEOAE/TEOAE/+ or - TEOAE) use of an AOAE-only screening regime to reduce the upfront capital outlay on screening devices.

- Use of an AOAE and A-ABR regime by Programme 8 (TEOAE/A-ABR) and Programme 9 (DPOAE, A-ABR/A-ABR) on the assumption that A-ABR screening takes longer and would add significantly to the cost of the programme if all babies were screened using A-ABR.

- For infants in Programme 3's (A-ABR/A-ABR) regional and rural areas, possible introduction of a third screen prior to referral to audiology to reduce costs of time and travel for families and professional staff.

Screening refer rates

Given the variability in data systems and definitions used by different programmes, comparable data for first refer rates is only available for four programmes. Of these four programmes three use TEOAE as the first screen and one uses A-ABR.

The first refer rate of the three programmes using TEOAE as the first screen vary widely, ranging from 8.8% for Programme 8 to 23.5% for Programme 6. Programme 1, which uses A-ABR as the first screen had a first refer rate of 6.3%.

Data from Programme 5 (TEOAE/TEOAE/+ or -TEOAE) reflects the refer rate at the time the infant is discharged from hospital and includes a mix of infants who have had either one or two AOAE screens, rather than simply a first refer rate following a single screen.

Programme 2 (A-ABR/A-ABR) and Programme 3 (A-ABR/A-ABR) both use the same data system which does not provide this information.

Rate of referral to audiology

Rates of referral to audiology ranged from 0.5% to 2.4% (Programme 4 (A-ABR/A-ABR) and Programme 6 (TEOAE/TEOAE/A-ABR) respectively). The rate achieved in Programme 4 (A-ABR/A-ABR) is significantly lower than other programmes, with the next lowest rates being clustered together, including Programme 2 (A-ABR/A-ABR) at 0.9%, Programme 5 (TEOAE/TEOAE/+ or -TEOAE) at 0.9% and Programme 1 (A-ABR/A-ABR) at 0.9%. The reason for the lower rate in Programme 4 (A-ABR/A-ABR) is not entirely clear, but could relate to the community based programme resulting in an older screening age which in turn supports a lower rate of transient hearing issues influencing screening outcomes.

It should be noted that although Programme 5's AOAE-only screening regime (TEOAE/TEOAE/+ or -TEOAE) achieves a rate of referral to audiology similar to the rate of A-ABR/A-ABR programmes, this outcome is in the context of completing three AOAE screens.

Given the wide variability across the programmes reviewed, and not having access to details on every aspect of each programme, it is not possible to speculate further on the reasons for the differences between the referral rates to audiology.

Data management

All programmes have a database, or in some cases more than one database, to manage newborn hearing screening data. A summary of the features of these data systems is presented below in Table 3 on the following page. Details of the data system for Programme 6 were not accessed as part of the review.

All of these programmes, at least for screening data, have either web-based data systems or availability of data for community screening (Programme 7) which enable wide access to the system within the jurisdiction.

All the databases include diagnostic audiology information. Several programmes include medical assessments and early intervention and one programme (Programme 1 (A-ABR/A-ABR)) also includes integrated information relating to family support. Programmes with these more extensive modules report this as being a significant strength.

Auditing of data varies across the programmes in terms of the scope of auditing undertaken. Resources to undertake these audits were often cited as limited.

Standardised reporting tools are available from all data systems to report on key performance indicators for newborn hearing screening. However, the need for improvements in standard reporting was also highlighted by several programmes.

A common challenge identified is the timely return of accurate and useable diagnostic audiology data. Other challenges included 'double handling' of data which is input, usually manually, to two or more systems (e.g. a hospital system and the newborn hearing screening programme database). Reducing the opportunity for fraud by protecting text files from screening equipment was also noted as a current challenge.

Table 3. Profile of data management systems of participating international programmes[A].

Details	Programme 1	Programme 2	Programme 3	Programme 4	Programme 5	Programme 7	Programme 8	Programme 9
Access to demographic data?	yes	yes	yes	Yes	yes	yes	yes	yes
Demographic data uploaded electronically or manually input to screening device?	manual	manual	manual	manual	manual	manual	manual	manual
Interface, electronic upload, or manual entry of demographic data to database?	electronic	manual	interface	electronic	electronic	manual	electronic	electronic
Data from screening device uploaded electronically or manually input to database?	electronic	electronic and manual	electronic	electronic	electronic	manual	manual	electronic and manual
Raw files from screening device saved to network and reconciled against database?	raw data files saved and reconciled consistently	raw data files saved and reconciled consistently	raw data files saved and reconciled occasionally			raw data files saved and reconciled consistently	raw data files saved and reconciled occasionally	raw data files saved and reconciled consistently
Unique identifier?	no	no	yes	yes	yes	yes	yes	yes
Referral to audiology electronic from database or manual?	electronic	manual	manual	electronic		electronic	electronic	electronic
Scope of data collection?	screening, AU, medical, EI, family support	screening, AU	screening, AU	screening, AU, medical, medical, EI	screening, AU, medical, EI	screening, AU, medical, EI, family support	screening, AU, medical, EI	screening, AU, medical
Number of databases (e.g. district/national; screening/audiology)?	1	2	2	1	2	2	1	1
Primary database web-based and available remotely and in major screening sites?	web-based, available remotely and in hospitals	web-based, available remotely and in hospitals	web-based, available remotely and in hospitals	web-based, available remotely and in hospitals	web-based, available remotely and in hospitals	not web-based, but accessible remotely, although not in hospitals	web-based, available remotely and in hospitals	web-based, available remotely and in hospitals

Details	Programme 1	Programme 2	Programme 3	Programme 4	Programme 5	Programme 7	Programme 8	Programme 9
Frequency of audit of screening and diagnostic data?	regular and frequent audits at all levels	regular and frequent audits at all levels	some auditing, not scheduled and consistent	regular and frequent audits at all levels	some auditing, not scheduled and consistent	regular and frequent audits at all levels	regular and frequent audits at all levels	regular and frequent audits at all levels
Reporting from database meet requirements for reporting against newborn hearing screening KPIs?	reports meet all requirements for KPI reporting	reports meet some requirements for KPI reporting	reports meet some requirements for KPI reporting	reports meet all requirements for KPI reporting	reports meet most requirements for KPI reporting	reports meet all requirements for KPI reporting	reports meet all requirements for KPI reporting	reports meet all requirements for KPI reporting
Key challenges	encouraging full use of all database features	exploring ways to ensure txt files from devices cannot be edited	double entry of audiology data from ESP to audiology Access database		entering data twice – once for hospital and once for database	manual entry to Guthrie card, then to pathology, no access to community data by hospitals, different identifier in community and hospitals	timely return of diagnostic data	implementation of whole programme, inclusion of EI
Future developments	building modules for related areas, e.g. Indigenous ear health	app for audiologists for ease of data entry	resolution of issues with standard reports in ESP, e.g. to report first refer rates		extend newborn hearing screening / audiology database to meet hospital needs.	downloads from equipment to audit individual screener or device performance	make system the database of choice for audiologists to encourage compliance	not all hospitals can provide direct access to demographic data

A Details of the data system used by Programme 6 were not accessed.

Workforce

The workforce across the eight programmes interviewed was variable, including nurses and midwives, a dedicated screening workforce and audiometric technicians. Details for each programme are provided in Table 2 above. Each programme expressed satisfaction with the workforce arrangements in place.

Programme 3 (A-ABR/A-ABR) noted a particular emphasis on employing screeners for their personality and attitude rather than their skills and past experience. It was noted that on occasions when a health professional (e.g. a nurse) had been recruited to a screening position within this programme they did not tend to stay for long.

Programme 1 (A-ABR/A-ABR), Programme 2 (A-ABR/A-ABR), Programme 3 (A-ABR/A-ABR), Programme 7 (TEOAE/TEOAE, A-ABR), and Programme 9 (DPOAE, A-ABR/A-ABR) all provide screening seven days per week. Programme 8 (TEOAE/A-ABR) provides seven day a week screening in larger urban hospitals.

Programme governance

Although oversight of each of the programmes occurs within a corporate health context, at the state/provincial/local government level, Programme 1 (A-ABR/A-ABR) is the only programme where the majority of jurisdiction-wide programme administration, planning and management occurs at this level. Even so, in Programme 1 (A-ABR/A-ABR), the central administrative systems have a strong interface with three coordinating roles that inform and monitor operational management across each of the local health services. In all other programmes, oversight of the programme is centralised but occurs within an operational context such as a community health service or a tertiary hospital that is accountable to the state/province/local government. Although some programmes expressed a desire for greater interest from central administrators, essentially the arrangements in place were noted to operate effectively.

CHAPTER 6 – Findings: Evidence from UNHSEIP practice

Introduction

The systems, quality standards, protocols and practices of the UNHSEIP were reviewed in detail through:

- analysis of a comprehensive set of key documents provided by the UNHSEIP (see Appendix E),

- a series of interviews with key stakeholders of the UNHSEIP, and

- visits to one metropolitan DHB (Auckland DHB) and one regional DHB (Northland DHB).

This process focussed on:

- exploring the features of the UNHSEIP against the background of published evidence and the systems and practices of other programmes, and

- identifying local contextual features that need to be taken into account when making recommendations specific to the UNHSEIP.

Details regarding the following processes and features of the UNHSEIP are presented in this chapter:

- programme inception

- programme governance

- regime and rationale

- screening timing and protocols

- screening devices

- inpatient and community screening

- workforce

- screening refer rates

- audiology

- data management

- the screening incident

Programme inception

The New Zealand UNHSEIP commenced in 2007 with a three year phased implementation. By 2010, universal screening was available in each of the 20 DHBs across New Zealand.

Essential elements of the UNHSEIP are described as including:

- ensuring coordination of all components of the programme,

- providing an organised invitation for screening to families and whānau of all newborns,

- delivering a multidisciplinary approach to screening, diagnosis and follow-up,

- building close links with treatment and early intervention services,

- establishing operational policies and quality standards and ongoing monitoring, and

- maintaining a focus on continuous quality improvement.

Programme governance

The UNHSEIP is jointly overseen by the Ministry of Health and the Ministry of Education. The Ministry of Health has oversight of screening, as well as audiology services and medical early intervention for babies diagnosed with hearing loss. The Ministry of Education is accountable for the early intervention services for babies diagnosed with hearing loss.

The NSU of the Ministry of Health is responsible for setting the strategic direction of the screening programme; developing and maintaining policy and standards; national monitoring, auditing, evaluating and quality improvement;

funding and contractual management; providing educational resources; and reviewing and overseeing the introduction of new technologies.[111]

The work of the NSU is supported by the UNHSEIP Advisory Group which includes representatives from professional and tertiary education bodies; Māori and Pacific populations; and consumer groups. The group provides advice to the NSU on matters relating to the monitoring and strategic direction of the programme. Additionally, working groups are established as needed to provide advice on specific matters.

Additional forums that guide and facilitate the work of the UNHSEIP include the:

- Joint Ministries Group, which includes representation from the Ministry of Health and the Ministry of Education and meets several times each year,

- NSU Advisory Group, which includes representation from public health experts and technical experts and meets several times each year, and

- Paediatric Technical Advisory Group of the New Zealand Audiological Society, which meets with the NSU approximately every two months.

The day-to-day operations of the UNHSEIP are managed by each DHB. DHBs deliver newborn hearing screening, diagnostic audiology and appropriate medical services. DHBs are responsible for providing leadership and oversight of the multidisciplinary team; internal quality assurance; quality improvement; clinical leadership; and executive support. The Ministry of Education Group Special Education is responsible for providing advisors of deaf children.

In 2012 the Ministry of Health contracted Deloitte to lead a three year audit programme for the UNHSEIP. The audits assess the compliance of DHB's with the Ministry's contractual agreement with the DHB for newborn hearing screening services and the UNHSEIP National Policy and Quality Standards (July 2013). The audits are also intended to assist DHBs to identify areas of focus for quality improvement and inform the NSU of areas for development of the UNHSEIP from a national perspective.

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[111] Ministry of Health. (2013). UNHSEIP National policy and quality standards.

Regime and rationale

The UNHSEIP has the core goals of:

- completion of screening by one month of age,

- completion of diagnostic audiology by three months of age, and

- initiation of appropriate medical and early intervention services by six months of age.

The programme reports on identification of both bilateral and unilateral permanent childhood hearing impairment.

The screening regime was determined through consideration of literature on screening and review of other international programmes at the time of programme establishment. The decision was informed by the speed, cost and ease of use of DPOAE as a first screen, paired with the opportunities afforded by A-ABR for infants cared for in NICU or special care baby unit (SCBU) and well babies with risk factors.

The well-baby screening regime is detailed in Figure 1 on the following page.

Figure 1. UNHSEIP well-baby newborn hearing screening regime.
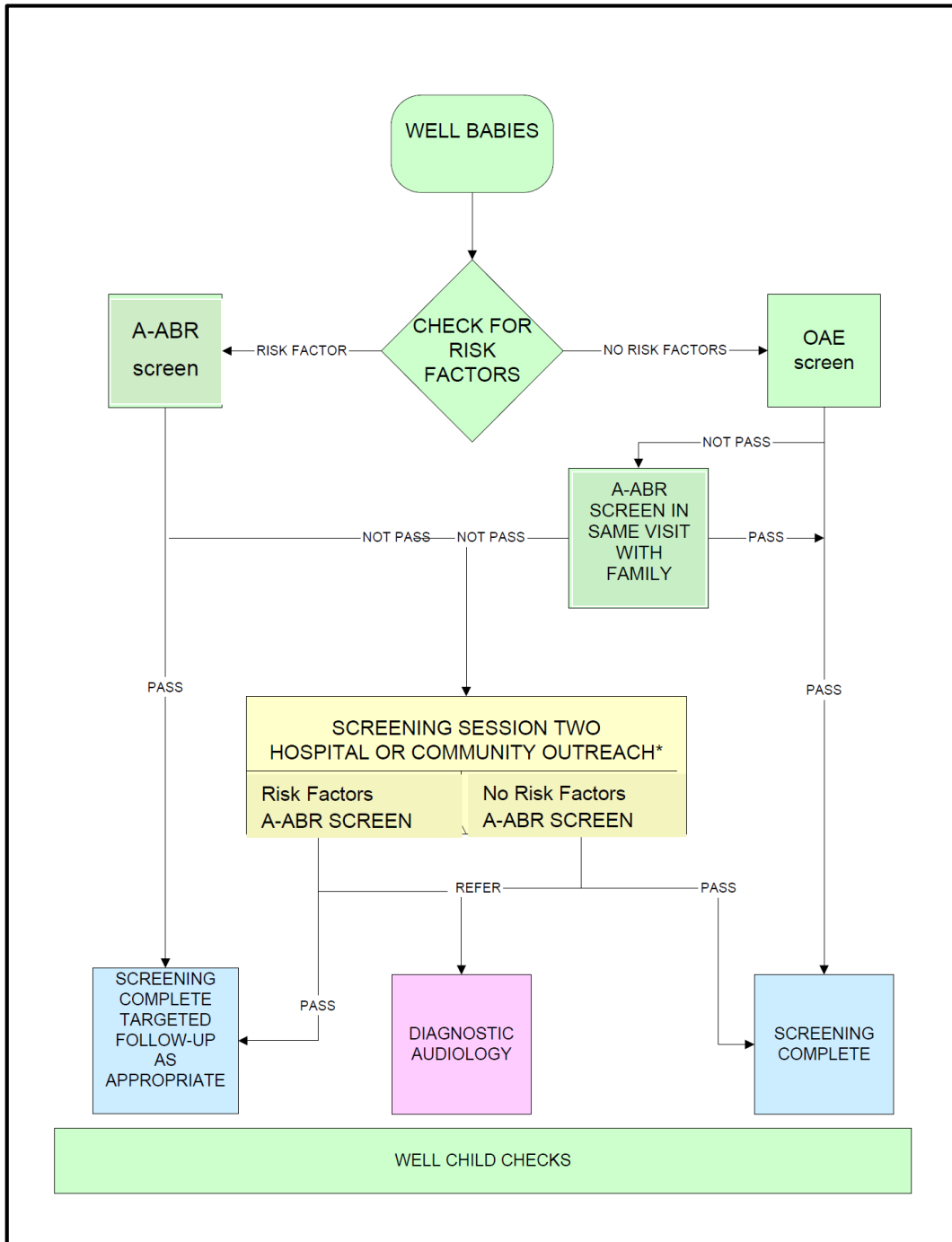
In exceptional circumstances, well babies who are screened at less than 72 hours and receive a refer result have the option of a third A-ABR screen prior to referral to audiology. Decisions regarding this option are made in collaboration with the family, screener and screening coordinator.

In contrast to the well-baby regime, well-babies who have risk factors have a two stage A-ABR screening regime. When a refer result occurs on the first A-ABR a second A-ABR is performed a minimum of five hours after the previous A-ABR. Following a screening refer result on a second A-ABR, the infant is referred to audiology for diagnostic assessment.

Infants who have spent more than 48 continuous hours in NICU or the SCBU have a single A-ABR screen followed by referral to audiology for diagnostic assessment in the event of a refer result.

All babies who pass newborn hearing screening but have defined risk factors for permanent hearing impairment are referred to audiology for targeted follow-up at 18 months of age.

Up to a total of three screening attempts per ear in each screening step are permitted. An 'attempt' is defined as occurring when a screen is started but stopped before a result is registered on the device. If a result is registered it is treated as a completed screen.

A number of screeners, coordinators and audiologists noted that the multiple variables influencing the screening and referral pathways cause confusion in the screening process. It was suggested that the complexity of the regime and the recent experience of the screening incident has resulted in too much focus on the screener and the protocol rather than on the baby, the parents and ensuring an optimal screening process.

A number of screeners, coordinators and audiologists, from across different DHBs noted that having two screening approaches, and in some situations two screening devices, significantly increases the length of screening scripts and the process of gaining consent from parents. It was reported that for some parents this causes confusion and anxiety, while others simply tune out to the explanations. An additional observation from one screener was that some families, when presented with the two screening approaches and devices, want to choose the approach they think might be best or the equipment they prefer.

Despite concern about the complexity of the screening regime, screeners noted that typically the AOAE screen is simple, quick and effective. Even so, it was noted that screening using DPOAE on the ward can be difficult due to noise interference.

One screener observed that screeners can develop a preference for AOAE screening over A-ABR screening because when they first begin screening they are learning many new things and do not carry out A-ABR screens as frequently.

Screeners, coordinators and audiologists reported that there is much confusion about the concept of 'screening restarts', including when a 'restart' should occur and when it should not. Specific concerns expressed about 'restarts' included:

- variability in the documentation of 'restarts',

- lack of standard application of the protocol detailed in the *UNHSEIP Screener Manual* regarding repeat screening attempts,

- the possible use of use 'restarts' as a means of 'A-ABR avoidance', and

- concerns about impacts on the willingness of families to attend diagnostic audiology following the experience of multiple rescreens.

Although most interviewees indicated a significant mood for a change from the current screening regime, many (but not all) also expressed the opinion that if a change to a two stage A-ABR regime occurs, each screening stage will take longer than is the case for the current regime. This was seen as particularly significant for DHBs undertaking a high proportion of community screening where babies are older and potentially more difficult to settle. These individuals were also concerned that a higher refer rate to audiology might be a potential risk arising from such a change.

Two individuals interviewed noted that with more effective systems the current screening regime has the potential to be efficient and effective.

Screening timing and protocols

The timing of the first screen is variable across and within DHBs. The timing of screening is influenced by diverse factors, including delivery in small hospitals and birthing centres without dedicated screeners; high rates of early discharge to home or birthing centres prior to screening; relatively high rates of home births compared to some other jurisdictions; and DHB-specific arrangements regarding workforce use, including variations in provision of screening seven days a week.

Some DHBs screen within the first few hours of birth and some do not screen before the infant is 24 hours old for vaginal deliveries and 48 hours for births by Caesarean section.

The UNHSEIP protocol is sensitive to issues of infant-parent bonding in the early postnatal period. A small number of interviewees expressed concern about the impact of newborn hearing screening on the bonding process.

In some locations, consistent with New Zealand practice guidelines, there is a separation of the offer of screening and the consent process for the first screen, in other locations these processes are combined.

Screening devices

The predominant devices used by the UNHSEIP are the MADSEN Classic AccuScreen, the MADSEN AccuScreen, and the MAICO MB 11 BERAphone. The Natus ALGO 3i is used in two DHBs.

The AccuScreen and ALGO 3i devices use consumables for each individual screen. The MAICO MB 11 BERAphone does not, although the headphone pads must be replaced at intervals.

Stakeholders consistently raised concerns about the MADSEN AccuScreen given the lack of capacity to access data on calibration levels for the AOAE which removes a powerful tool for identification of self-screening. Concern about this is heightened in the context of the UNHSEIP screening incident and similar incidents that occurred in the NHS Newborn Hearing Screening Programme in the UK[112].

DHBs noted that many devices are approaching their 'use-by date' and maintenance costs are increasing. Devices and consumables are funded by DHBs and decisions on their replacement must be made in the near future.

There is a belief amongst some UNHSEIP stakeholders that the nature of the screening equipment, which enabled screeners to screen their own hearing, was a contributing factor to the screening incident.

A number of screeners reported that parents can be uncomfortable about the use of probes for DPOAE screening. Not needing to stick electrodes to a baby's skin when using the BERAphone compared to the AccuScreen for an A-ABR is favoured by parents.

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[112] Ministry of Health. (2012). Quality improvement review of a screening event in the Universal Newborn Hearing Screening and Early Intervention Programme.

All screeners interviewed who use the MAICO MB 11 BERAphone expressed a strong commitment to the benefits it offers in terms of time to screen, ease of use and acceptability to parents. Some screeners indicated that devices would need to be changed immediately from the AccuScreen if the regime was to change, while others indicated that given the cost of devices and consumables it would be appropriate for current devices to be used to the end of their life.

Given the current use of the MAICO MB 11 BERAphone in a number of DHBs, many screeners interpreted the idea of an A-ABR/A-ABR regime to mean a 'BERAphone regime'. There was evidence of potential resistance to change to an A-ABR/A-ABR regime if an alternate device is recommended.

Screening coordinators in DHBs that use the AccuScreen and the MAICO MB 11 BERAphone noted the current costs of maintaining and calibrating two different pieces of equipment.

Inpatient and community screening

Rather than having a particular focus on the location of screening, the UNHSEIP emphasises screening by one month of age, regardless of location.

Larger or tertiary hospitals screen a high proportion of babies as inpatients. Possible reasons for this are that high birth numbers and a critical mass of screeners support more efficient use of screener time within the inpatient context.

Early discharge, the presence of many small maternity centres in some regions, homebirths, site-specific protocols that delay screening until 24 hours of age, and variable workforce capacity to provide screening seven days a week make inpatient screening challenging in some locations. In a minority of DHBs more babies are screened as outpatients than inpatients. Hospitals and birthing centres with fewer births noted that it is not a viable or effective use of resources for small hospitals or birthing centres to have screeners onsite and available at all times given that their time would not be fully occupied. In these contexts, community screening is seen to be more efficient. Even so, DHBs recognised community screening requires resource intensive activities, such as appointment scheduling and confirmation with families, travel time, clinic setup, management of non-attendance, and additional time to settle older babies.

Some families need to travel up to one and a half hours each way to attend community screening. This can present significant challenges for people who do

not have access to transport or experience financial challenges. In some DHBs travel assistance is available to families.

Some interviewees noted that local processes that reduce opportunities for inpatient screening (such as protocols for the timing of screening) can impact significantly on the possibility of some babies accessing screening at all. Contributing factors include social issues, competing family priorities, inadequate transport, and financial difficulties.

Workforce

The UNHSEIP has a dedicated screening workforce (not all with health backgrounds), supplemented in some locations by individuals in other roles who undertake screening as one of a number of responsibilities. The workforce is based on one full time equivalent screener per 1,250 live births.

Some screening services are structurally located with the DHB audiology service, others are part of maternity, and others are part of community services. Regardless of service structures, many noted a recent increase in integration between screening and audiology.

In numerous locations screeners routinely travel long distances to undertake community screening in regional and rural communities.

Training requirements, protocols and quality standards are prescribed by the NSU through contractual arrangements with DHBs. All screeners are required to participate in two aspects of training for their roles:

- DHB-based training provided by screener trainers that includes theoretical and practical instruction, and

- completion of NZQA 1623: National Certificate in Health, Disability, and Aged Support (Newborn Hearing Screening) within 12 months of employment.

It is an employment condition that the NZQA 1623 qualification is completed within 12 months of employment. All screeners must undertake competency assessment either annually or after a break from screening of six months or more.

Each DHB is responsible for a set of coordination functions defined by the NSU. These tasks may be carried out by a mix of a screening coordinator, a DHB manager, a senior screener and/or an audiologist. Interview feedback indicated

that the screening regime complexity, use of two different devices in some DHBs, and the extent of monitoring requirements places high demands on the coordination function.

Screeners noted that the complexity of the screening regime and the use of two different devices requires more extensive and complex training and monitoring than might otherwise be required. One screening coordinator noted that if the regime is changed to A-ABR/A-ABR, and appropriate support and monitoring is put in place, the simpler regime may enable more flexible workforce arrangements to be used in locations with low birthing numbers or where there are challenges achieving optimal capture rates. There are already examples of screening being carried out by individuals in a range of roles, including a midwife in a rural town, nurses in a NICU, and nurse assistants in a hospital.

Screening capture, completion and refer rates

National monitoring data for the period April – December 2012 indicates that 83% of all babies completed screening. Of the babies who started screening, 98.5% completed screening. Completion of screening occurred within the national target and international benchmark of one month corrected age for 91.9% of babies who completed screening.[113]. However, 2.7% (1,058) of those babies who were screened did not complete screening until after eight weeks and up to 44 weeks, with a further 17.0% of infants not receiving a newborn hearing screen at all[114].

Preliminary data provided by the NSU for the period October 2011 to December 2012 indicates a first refer rate from DPOAE to A-ABR of approximately 15.0% and from A-ABR to A-ABR of approximately 5.0% (email communication, 14 January 2014).

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[113] Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012, National Screening Unit, Ministry of Health, New Zealand Government. Retrieved from: http://www.nsu.govt.nz/files/ANNB/UNHSEIP_Monitoring_report_April_to_Dec_2012.pdf

[114] ibid.

Audiology

The UNHSEIP has a 1.7% refer rate from screening to audiology. At a national level this is within the 4.0% benchmark. When considered at an individual DHB level, all but one DHB is currently meeting this benchmark.

Although most DHBs provide infant diagnostic audiology services, audiology services are also contracted between DHBs and with private providers in some locations.

The most recent UNHSEIP Monitoring Report includes audiology data on 381 of 672 referrals for the reporting period April – December 2012[115]. Although from current data it is not possible to confirm the audiology lost to follow-up rates, the Monitoring Report notes that rather than indicating that 43.0% of babies have not been seen by audiology, some DHBs have not submitted audiology data to the NSU.

Delays in audiology diagnostic assessment are reported in some locations. Of those that data is available for, 78.0% completed audiology assessment within the target of three months of age[116]. Audiologists in a regional location reported that some infants do not attend their first audiology appointment until after three months of age.

Audiologists from two DHBs reported that there appears to be a higher proportion of Maori children with a sloping high frequency hearing loss than is found in the non-Maori population. In response to this observation, the Deafness Notification Database began collecting data on children diagnosed with very high frequency hearing loss in 2011. The extent of data currently available on this phenomenon is not adequate to inform or influence this review[117].

Data management

Demographic data relating to newborns is available to the UNHSEIP from a variety of local and national sources and key demographic data items are input

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[115] Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012

[116] ibid

[117] New Zealand Audiological Society. (2012). Deafness notification report 2011. Retrieved from: http://www.audiology.org.nz/deafness-notification-database.aspx

manually to devices by screening staff. Demographic data from patient management systems is uploaded to local databases in some DHBs but is not uploaded to a national database as a precursor to input and matching of screening data.

There is a unique patient identifier in New Zealand, the National Health Index (NHI) number.

DHBs are required to report screening data nationally. Each DHB has developed its own mechanisms for data collection, operational management and reporting following screening. Some DHBs have sophisticated data systems available across a DHB, others manually enter data into an Excel spreadsheet, and some small DHBs rely on paper-based systems.

Raw data from screening equipment is saved to the local DHB network for quality monitoring and future reference. There is little, though increasing, reconciliation of raw data with local databases following the review of the incident.

The local and national data requirements and the manual and electronic systems results in doubling, or in some cases, tripling of data input effort and the creation of several data repositories for screening information (including the raw data from screening devices). This makes identification of accurate information problematic.

The scope of data collected varies across the local databases and the NSU database. The NSU database enables the collection of diagnostic outcome data from audiology, although compliance is low.

Extensive data auditing is occurring, particularly in relation to screener performance – although many coordinators reported that the current protocols place excessive demands on resources.

Monitoring reports, developed by the NSU using data collated using the NSU database, provide a thorough analysis of the available data back to DHBs. However, coordinators and audiologists indicated that the reports are not available in a timely fashion so do not offer value for local review and audit of performance. This is in part because a time lag of six months which is allowed before data extraction occurs in order to maximise capture of diagnostic audiology data. Different data sources are used for some data items which makes overall comparisons challenging.

Every interview surfaced significant concern about the lack of a national database that enables effective identification of all births, provides real time data that can be verified, facilitates effective tracking of children, particularly across DHBs, and enables effective monitoring of the screening programme.

A national data repository is currently being developed that will record hearing screening events through the Maternity Clinical Information System.

Screening incident

In the context of the screening incident, a perceived lack of trust in the programme was noted to be challenging for some in the screener workforce. Whilst acknowledging the importance of comprehensive performance monitoring and auditing, interviewees commented that screeners feel a constant sense of scrutiny which in turn impacts upon their confidence and work satisfaction.

Despite these perceptions, one screening coordinator noted that the constructive, 'no-blame' approach the NSU has taken to managing the screening incident has been a significant support to positive change in the programme. Further, all individuals interviewed demonstrated a very strong commitment to doing whatever is necessary to deliver an effective and efficient programme that internal and external stakeholders have confidence in.

There was a perception by some that for as long as the current regime remains, the programme's reputation will continue to be affected and the risk of the same issue arising again will remain. The need for a chance to 'refresh' the programme through a new regime was expressed by some. However, a clear message was conveyed by a number of people that a regime change alone would not be the 'silver bullet'. They stressed that if other programme issues were not addressed, or a well-designed, comprehensive implementation plan was not used to make any recommended changes, the desired outcomes would not be achieved.

CHAPTER 7 – Integrating the findings: Variables impacting on regime choice

Determining the optimal screening regime for a particular context requires consideration of numerous variables, all of which interact with each other, often in complex ways.

This review seeks to recommend a newborn hearing screening regime to the UNHSEIP that:

- delivers optimal clinical efficacy and efficiency,

- facilitates diagnostic audiology assessment occurring as early as possible,

- achieves optimal screening capture and completion rates,

- minimises the impacts on families,

- achieves to optimal operational efficiency, and

- supports maintenance of quality standards.

This chapter will explore each of these issues in turn.

Delivering optimal clinical efficacy

The clinical efficacy of a screening regime is determined by:

- capacity to identify moderate or greater permanent childhood hearing impairment hearing loss,

- capacity to identify ANSD,

- sensitivity and specificity for detection of moderate of greater hearing impairment, indicating the regime's false positive rate (referral to audiology when no hearing impairment is present) and false negative rate (a pass on screening when there is in fact a hearing loss),

- extent to which the screen is effected by extraneous and physiological noise, and

- the extent to which the screen is effected by minor middle and outer ear conditions.

Identifying moderate or greater PCHI

Integral to a universal hearing screening programme is a clearly defined target condition. Most programmes use a target condition of moderate or greater PCHI. This is due to the fact that this degree of hearing impairment is likely to significantly impact on the development of speech and language. Given this, and equipment settings, all regimes are able to detect moderate or greater hearing impairment.

Identifying ANSD and/or neural disorders

ANSD can occur alone or in conjunction with a sensorineural hearing loss. In the NICU population, prevalence rates for ANSD have been found to be 0.2%, or one baby per 605 births, with 69.0% demonstrating a bilateral presentation and 31.0% unilateral. The well-baby population, with no known risk factors for hearing impairment or indicators of central nervous system pathology, is reported to have a lesser rate of 0.06 to 0.3 per 1,000 babies[118], including 14.0% demonstrating a bilateral presentation and 86.0% a unilateral presentation. Despite the lower incidence of ANSD in the well-baby population, early detection is important to optimise opportunities for early intervention.

The detection of ANSD can only occur using a regime that uses A-ABR alone. Any regime using an AOAE will be unable to detect ANSD or neural conditions in babies with normal cochlear function. Therefore the baby will pass the AOAE screening and be discharged without referral to audiology.

Neonates who have ANSD in the presence of a moderate or greater sensorineural or conductive hearing loss will not pass an AOAE screen and will refer on for an additional screen (or screens) at which point a refer result will occur and precipitate a referral for audiological investigation. .

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[118] Korver, A.M.H., van Zanten, G.H., Meuwese-Jongejeugd, A., van Straaten, H.L.M., Oudesluys-Murphy, A.M. (2012). Auditory neuropathy in a low-risk population: A review of the literature. *International Journal of Pediatric Otorhinolaryngology*, *76*(12), 1708–1711.

Sensitivity and specificity, false positives and false negatives

The sensitivity and specificity of a regime is reliant upon the screening device to a significant degree. Equipment utilised in any health or medical profession has rates of sensitivity and specificity (and false positive and negative rates).

Every screen, piece of medical equipment/device, and regime is subject to confounding variables, including human error, and will not be 100% accurate. As identified through the literature review, A-ABR screening has higher sensitivity and specificity rates than AOAE screening.

Low false positive rates are also important in order to minimise excessive referral to audiology of babies who do not have a permanent hearing loss. On the same token, a minimal level of false negatives ensures the screen is not allowing children who have a hearing impairment to pass the screen. Overall, A-ABR screening has higher rates of statistical validity.

Minimising confounding variables impacting on the regime

It is recognised that AOAE screening can be significantly affected by extraneous and physiological noise (i.e. sucking and breathing). These confounding variables make AOAE screening in a ward environment challenging which can impact negatively on screening sensitivity and specificity rates, and the incidence of false positive and negative referrals. A-ABR screening is not as susceptible to extraneous noise, enabling screens to be easily conducted in a ward environment. To a lesser extent, high levels of electrical interference, noise and muscle artefact (i.e. large movements of the baby) can impact on the A-ABR screening process.

There are a wide range of consequences from a false negative screen result[119]. For this reason, regimes need to employ a rigorous and standardised process in order to attempt to minimise confounding variables, and false negative results, as much as possible. This includes strictly limiting the use of additional screens, except in in exceptional circumstances.

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[119] Petticrew, M.P., Sowden, A. J., Lister-Sharp, D., Wright, K. (2000). False-negative results in screening programmes: systematic review of impact and implications. Health Technology Assessment. *Health Technology Assessment*, *4*(5). Retrieved from: http://www.hta.ac.uk/fullmono/mon405.pdf

Minimising the effect of middle and outer ear status

AOAE screening is more susceptible to outer ear and middle ear status in comparison to A-ABR screening.

Neonates are often born with significant amounts of vernix in their ear canals, or with significant middle ear fluid. Vernix in the outer ear can persist for a period of time, as shown in research demonstrating that approximately 14.3% of neonates have vernix occluding the outer ear space within the first 24 hours of life and 11.5% between 24 and 48 hours of life[120].

The DPOAE screen is highly affected by outer and middle ear conditions. This results in a high referral rate following the first DPOAE. A study in the United Kingdom newborn hearing screening programme showed that screening pass rates for ears containing occluding vernix were 38.0% with AOAE and 66.0% with A-ABR[121]. This difference results from the fact that the A-ABR screens the neural auditory system and is less affected by outer and mild middle ear conditions.

Facilitating diagnostic audiology assessment as early as possible

Early completion of screening provides the foundation upon which to meet the international benchmark of completion of audiology assessment by three months of age.

The main operational advantage of an A-ABR regime over a regime that uses a combination of AOAE and A-ABR is that screening can occur very soon after birth and deliver a lower first screen refer rate than a regime using AOAE as the first screen. The two screens of a two stage A-ABR regime can often be completed within 24 hours and prior to discharge.

In addition to early screening, meeting the benchmark of completion of audiology assessment by three months of age can be further supported through:

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[120] Keefe, D. (2007). Otoacoustic emissions: Clinical applications. In Martin S., Robinette, Theodore J. Glattke (Eds). *Otoacoustic emissions: Clinical applications.* Thieme: New York.

[121] Shoup, A.G., Owen, K., Jackson. (2010). 10 years of UNHS: Quality improvement is perpetual for Parkland Universal Hearing Screening Program. Retrieved from: http://www.infanthearing.org/meeting/ehdi2010/ehdi_2010_presentations/10%20Years%20of%20UNHS.pdf

- application of standard audiology benchmarks to commence audiology assessment within two weeks of a bilateral refer result, and six weeks of a unilateral refer result[122], and

- establishment of mechanisms to support flexible use of audiology resources within and between DHBs to ensure benchmarks are met for all children in all locations.

Delays in the opportunity for early diagnostic audiology assessment exist for nearly 20.0% of all New Zealand infants, with 2.7% (1,058) of those who were screened not completing screening until after eight weeks and up to 44 weeks, and a further 17.0% of infants not receiving a newborn hearing screen at all[123].

Such delays can result in babies being too old for electrophysiological assessment. Babies older than three months typically require multiple appointments to complete assessments due to lighter sleep states. In some circumstances, electrophysiological assessment may not be possible and behavioural assessments may have to be utilised. The result creates two issues. The first is that reliance on behavioural assessments on young babies is not as robust and often less frequency specific and ear specific information may be gained from an audiology appointment. This compromises optimal hearing aid fitting due to the need to estimate frequency specific or threshold information. The second is a delay in hearing aid fitting which has significant implications for early communication development.

For these reasons, a two stage A-ABR regime with a focus on high capture rates prior to hospital discharge, has the potential to significantly reduce the timeframe for completing screening and commencing and completing diagnostic audiology within optimal timeframes.

One study has also shown that parents who receive an audiology appointment prior to discharge, have increased attendance and follow through at audiology[124]. Receiving an appointment prior to discharge, provides families with confidence

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[122] Paediatric and Neonatal Diagnostic Audiology (PANDA), Australia. (2013). National minimum assessment standards for newborn hearing screening.

[123] Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012.

[124] Shoup et al. (2010). 10 years of UNHS: Quality improvement is perpetual for Parkland Universal Hearing Screening Program.

that their questions will be resolved quickly, which contributes to reduced anxiety.

The efficacy and impact of a screening regime on audiological resources is important to consider. The profession is small and the skills required in infant diagnostics are highly specialised. Most audiologists require significant experience to develop competence in interpreting infant diagnostics. In the context of resource challenges, minimising false positive referrals to audiology is essential to minimising audiology waiting times. Use of a two stage A-ABR regime prior to discharge, for both well-babies and those who have spent time in NICU[125], supports the achievement of this outcome.

When contrasting the audiology resource requirements needed for an A-ABR/A-ABR regime with the current UNHSEIP regime of AOAE, A-ABR/A-ABR, there is no expectation that a change in audiology requirements would be likely to occur.

It is worth noting that with any change of regime there may be a temporary increase in referrals to audiology while the screening workforce gains proficiency with new protocols and devices.

Achieving optimal screening capture and completion rates

The internationally accepted benchmark for universal newborn hearing screening capture and completion is that 95% of eligible infants complete screening by one month corrected age[126]. As demonstrated in eight out of the nine programmes that contributed to this review, this is typically achieved through prioritisation of inpatient screening. As a result, a screening regime is required that:

• is conducive to screening very soon after birth, and

• can be effectively administered in a hospital or birthing centre environment.

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[125] Although the screening regimes of the UNHSEIP and two programmes that contributed to this review refer directly to diagnostic audiology following a refer result on a single A-ABR screen, this review did not identify any published evidence detailing the clinical merits, or otherwise, of this approach.

[126] Joint Committee on Infant Hearing. (2007). Executive summary of joint committee on infant hearing year 2007 position statement: Principles and Guidelines for Early Hearing Detection and Intervention Programs. http://www.jcih.org/ExecSummFINAL.pdf

As detailed previously, in contrast to AOAE screening, A-ABR screening is less likely to be affected by extraneous noise in a ward, and debris in the outer ear and middle ear fluid that is frequently present soon after birth. Additionally, in these very early hours after birth, infants are likely to have more frequent and longer periods of settled sleep,

Manufacturer guidelines indicate that A-ABR screening can commence within four hours of birth, with a second screen within 24 hours. In some locations first and second screens are undertaken sooner to maximise screening commencement and completion for infants who are likely to be discharged and may experience challenges returning for community screening. Screening in the early hours after birth is not possible for regimes including AOAE as a first screen, without incurring a high refer rate[127, 128].

The Australian Government, MSAC report on universal newborn hearing screening concluded that:

- community based screening is effective when 'piggy-backed' onto other health checks and immunisation programmes, and

- losses to follow-up are more likely when there are long delays between rescreening or screening in the community after early discharge[129].

Interestingly, the one programme that contributed to this review that provides universal community screening completed only 73.3% of all screening under one month of age, despite achieving a screening completion rate of 96.8%. These delays in screening in turn result in the potential risk of compromised timing of diagnostic audiology assessment for a significant proportion of children.

The relatively low proportion of inpatient screening in some DHBs may well be a significant contributor to the low screening completion rate of 83.0% in New Zealand.

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[127] Maxon, A.B., White, K.R., Culpepper, B., Vohr, B.R. (1997). Maintaining acceptably low referral rates in TEOAE-based newborn hearing screening programs. Journal of Communication Disorders, 30, 457-475.

[128] Kok M.R., Zanten G.A., van Brocaar M.P. (1993) Click-evoked 0toacoustic emissions in 1036 ears of healthy newborns. Audiology, *32*, 213-24.

[129] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee reference 17, Assessment report. Retrieved from: http://www.msac.gov.au/internet/msac/publishing.nsf/Content/8FD1D98FE64C8A2FCA2575AD0082FD8F/ $File/ref17.pdf

Recognising A-ABR as being ideally suited to screening in the immediate newborn period, some screeners expressed concern that screening infants in the community, who are by definition older than those screened as inpatients, would take more time using a two stage A-ABR regime rather than a regime that uses an AOAE as the first screen. However, other programs interviewed successfully deliver community screening using a two stage A-ABR regime. Nonetheless, a different regime will require different areas of emphasis in training. Namely, for a two stage A-ABR regime, training will require a greater focus on techniques for effective infant settling.

Minimising the impacts on families

When determining a newborn hearing screening regime, the relative impacts on families of different regimes must considered. The specific circumstances and needs of different community groups must be assessed and responded to appropriately, including Maori, Pacific, Asian and remote and rural populations all require detailed consideration.

Emotional impacts of the screening process and false positive results

Three systematic literature reviews have found minimal high quality evidence on issues pertaining to the impacts of newborn hearing screening on parents and their relationship with their infant[130,131,132]. Of the evidence that is available, there is little to support suggestions of undue anxiety arising from either newborn hearing screening in general, or more specifically, following a false-positive screening result. The review by the Mt Sinai Hospital, Canada stated that 'When standard, validated measurement tools for anxiety are used, there appear to be no substantive differences between affected families and the general population, or between groups of families in different screening outcome situations.' While some parents report a degree of anxiety before, during and/or after screening, anxiety levels have been found to be within normal limits. No significant differences have

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[130] Picton, Hyde (2004). Family anxiety and universal newborn hearing screening (UNHS): A review of current evidence.

[131] Nelson, Bougatsos (2008). Universal newborn hearing screening: Systematic review to update the 2001 US Preventive Services Task Force recommendation.

[132] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee.

been found when studies have compared the anxiety of parents whose child received a refer result with those who received a pass result or those who were not screened at all. The Mt Sinai Hospital review noted that in the context of parents feeling anxiety following screening it is possible that the anxiety relates to issues unrelated to screening.

One study considered by the US Preventive Services Task Force identified that anxiety, worry and uncertainty were increasingly negatively affected as the number of screens increased.

Current evidence does not differentiate between the impacts of different screening regimes on families. Even so, despite the lack of evidence supporting significant impacts on parental anxiety, the Mt Sinai review reflected on the likely value of minimising any potential impacts by reducing false-positive results to the extent that is possible. Given the well-recognised high refer rates resulting from AOAE screening, a two stage A-ABR regime, paired with optimal protocols for screening timing, responds to this suggestion most effectively whilst simultaneously reducing the number of screens prior to referral to audiology.

Notwithstanding the findings of published literature, when considering feedback from UNHSEIP screeners, perceptions of parental confusion and anxiety are not uncommon. Screeners interviewed reported that they believe this is associated with the use of two different approaches to screening, two different devices (in some DHBs), and associated lengthy processes of explanation and consent. A screening regime utilising a single screening approach has the potential to significantly simplify this process for families.

Demands of community screening

As detailed above, a number of variables compromise the clinical efficacy of AOAE as a first screen in the immediate postnatal period and in an inpatient context when contrasted to A-ABR. In the context of early postnatal discharge, when programmes use a regime combining AOAE and A-ABR screening, the need for family to attend community screening is likely to be significantly greater than when programmes use a two stage A-ABR regime. This is an important consideration where distances to travel to community screening can be significant, and for some families the challenges of transport, finances and other family priorities can be significant, particularly in the period following the birth of a baby.

Demands of false positive referrals to audiology

The current UNHSEIP regime for well babies AOAE, A-ABR/A-ABR should not produce a higher rate of false positive referrals to audiology than a standard A-ABR/A-ABR regime. In contrast, the single A-ABR regime used by the UNHSEIP for infants who have spent 48 hours or more in NICU will result in a higher proportion of referrals to audiology than if a two stage A-ABR regime is used. For families whose infants may have already experienced a difficult start, reducing the possibility of this additional demand to the extent that it is possible warrants consideration. This is particularly the case in the context of this review not identifying any published evidence detailing the clinical merits, or otherwise, of this approach.

Even so, the option of referring any child directly to audiology either without screening or following a single screen must always remain available in circumstances such as significant parental anxiety, specific medical conditions, or other extenuating circumstances.

Achieving optimal operational efficiency

Different screening regimes place different operational demands on the screening programme and the broader health service. The review team proposes that ideally, a regime should:

- minimise complexity in relation to training, parental consent, screening, device use and maintenance, and quality management,

- enable flexible use of the available workforce in diverse contexts, and

- deliver cost efficient and effective screening.

Minimising complexity

Regimes involving two different screening approaches, namely those using a combination of AOAE and A-ABR screening, have significantly greater operational complexity than regimes involving a single approach to screening, such as a two stage A-ABR regime.

The current UNHSEIP screening regime utilises two different screening approaches, and three different screening pathways, with a fourth pathway for targeted follow-up of children with defined risk factors. This level of regime complexity was reported to have a significant impact on screener training and

monitoring of screener performance. Some screeners interviewed suggest that use of a simplified screening regime would provide the opportunity for screeners to invest in optimising screening technique rather than needing to give significant focus to the nuances of the screening regime.

In relation to screening devices, currently in some DHBs the same screening device is used for both the AOAE screen and the A-ABR screen. In contrast, in other DHBs, two different devices are used, one for the AOAE and one for the A-ABR.

Consequences of using different screening approaches, and in some locations different screening devices, include:

- the need for screeners to have knowledge of, be proficient in and maintain competency standards in relation to:

  o the daily checks and operational use of two devices,

  o two screening scripts, or one lengthier script with reference to two screens and devices,

  o learn techniques regarding probe tips and electrode placement, screening re-starts, trouble shooting and maintenance which are unique to each device,

- challenges maintaining competency in the use of two screening approaches and devices for screeners working in isolation or where fewer babies are born,

- greater demands on performance monitoring and broader quality management,

- impacts on the ease of travel to community clinics, with two devices being more cumbersome and requiring time for set up,

- greater administrative demands involved in ordering consumables at different rates, times and costs for two devices, and

- yearly calibration of two different devices, with two different manufacturers, rather than one device, with one manufacturer.

Enabling flexible use of the workforce

This review has not identified published evidence exploring whether specific newborn hearing screening regimes warrant unique workforce arrangements.

Independent of the screening regime being used, the review found that the workforce used to undertake screening is highly varied. Different jurisdictions included in the review use one or a combination of:

- a dedicated screener workforce, not necessarily with a health professional background

- a non-professional workforce with individuals undertaking screening as one of a number of roles,

- volunteers,

- students training in audiology and/or nursing,

- a health professional workforce with individuals undertaking screening as one of a number of roles,

- a health professional workforce with individuals rostered onto dedicated screening shifts, and

- a non-professional and/or professional workforce that are drawn upon 'as-needed' and with appropriate supervision to support screening in locations and circumstances where screening coverage is difficult.

As noted above, demands on the screening workforce and its management can be minimised through using a regime that minimises complexity, both in terms of the screening process and the screening device used. For the UNHSEIP, in locations where there is a need to find ways to improve screening coverage and completion rates to meet international benchmarks, a simplified regime would open up possibilities for more flexible use of the broader health workforce to contribute to this goal.

Specific opportunities that could be considered include, but are not limited to incorporating newborn hearing screening into the roles of:

- kaiāwhina (community health workers),

- vision hearing technicians employed through the B4 School Check,

- nurses and midwives from small hospitals and birthing centres, and

- other health technician roles.

In circumstances where screening volume is particularly low and it is not possible for individuals to maintain screening competency, telehealth supervised by a skilled screener/audiologist provides an additional opportunity for effective and efficient workforce use.

The Australian Government, Medical Services Advisory Committee[133] identified that invalid screen results can occur when the screener:

- is unfamiliar with the screening device and inexperienced at determining the validity of a screen result,

- is inexperienced at handling infants,

- positions the insert probe or couplers inadequately or places the electrodes poorly, or

- allows insufficient time for screening.

The demands involved in responding effectively to these factors will vary in the context of different regimes.

Analysing screening costs

Programme costs are key in decision making regarding a newborn hearing screening regime. Principle 9 of the World Health Organization guidelines in relation to screening states that 'The total cost of finding a case should be economically balanced in relation to medical expenditure as a whole.'[134] As one element of this review a cost analysis was undertaken, with the following aims:

- to enable consideration of cost comparisons for varying regimes, and

- to examine where variations in some aspects of a regime, or of screening practice, will affect the overall costs.

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[133] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee.

[134] World Health Organization. (1968). Principles and practice of screening for disease. Retrieved from: http://whqlibdoc.who.int/php/WHO_PHP_34.pdf

The standard regimes[135] included in the literature review were considered and are presented in a table in Appendix F. Particular emphasis was placed on two regimes, namely, AOAE, A-ABR/A-ABR (the regime used by the UNHSEIP) and A-ABR/A-ABR[136].

There are a few important points to note about this costing analysis:

- it provides relative costs only and is not a budgeting exercise,

- it does not comprehensively cover all screening costs (e.g. infrastructure costs),

- it should not be used as the sole basis for decision-making regarding a newborn hearing screening regime, and

- it should not be used to formulate a programme budget.

Cost categories and analysis parameters and variations

A standard set of notional costs were calculated for five key cost categories:

- **Initial setup costs:** office setup, initial training, and equipment purchase (conservatively assuming a five year equipment lifespan).

- **Equipment maintenance costs:** annual supplier maintenance, local screener and co-ordinator maintenance – daily, weekly, monthly and annual.

- **Family costs where screening is not completed in hospital:** mileage and attendance costs (e.g. parking, babysitting).

- **Direct screening costs:** consumables, labour for each screen, additional labour for refer rescreens, co-ordination for refer results, management, additional costs for community screening (assuming 10% community screening rate), tracking of lost to follow-up or referred infants, screener training and competency assessment, additional hearing assessment and

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[135] Screening regimes that only use a single screen are not examined due to the inherently high refer rates and consequent diagnostic costs associated with them.

[136] Analyses for all regimes and detailed assumptions and costs used to inform the analysis are included at Appendix F.

therapy for infants not diagnosed with ANSD in the context of a regime using AOAE as the first screen.

- **Diagnostic costs:** assessment of false positives (two appointments), assessment of permanent bilateral moderate or greater hearing loss, and other hearing losses (four appointments), data entry and report writing.

Data management costs were not included in the screening estimates due the difficulty of establishing a realistic average cost given the wide variety of data management processes used

The parameters for the analysis of notional annual costs are set out below. Annual costs for each regime were based on:

- full capture of 1,000 eligible well-baby births[137],

- use of the same: screening workforce; A-ABR first, second and/or third refer rates; AOAE first, second and/or third refer rates; lost to follow-up rate; and outpatient screening rates,

- use of consumables,

- current costs expressed in (NZ$),

- additional time being allocated to A-ABR screening (30 minutes) compared to AOAE (20 minutes), and

- incorporation of some conservative estimates of: costs to families[138] and additional costs associated with non-detection of auditory neuropathy for regimes which begin with AOAE.

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[137] The review team acknowledges that international best practice requires the use of A-ABR screening for infants who have spent five days or more in NICU, however for this exercise the cost impacts of this difference were not assessed to be of a sufficient scale to make this distinction.

[138] An evaluation carried out in 2006 by the UK NHS considered family costs associated with non-inpatient screening and found that 'An average family cost for NHSP, when the screen had not been completed in the maternity unit, was £20.10, consisting of £9.58 in direct costs (travel, car parking, child minding arrangements, etc.) and £10.52 in lost parental wage costs.'

Uus, K., Bamford, J., Taylor, R. (2006). An analysis of the costs of implementing the National Newborn Hearing Screening Programme in England. *Journal of Medical Screening, 13*(1), 14-9.

Variations to these standard costs are examined, including:

- the cost of screening alone, in contrast to the cost of screening and audiology combined,

- the cost of A-ABR screening with consumables, in contrast to the cost of A-ABR screening without consumables[139], and

- the additional cost of screening infants in community clinics, rather than as inpatients.

Limitations

It is important to note a number of important limitations of this cost analysis exercise:

- reported refer rates and costs for a range of variables vary widely in the literature, as do costs affected by exchange rates and pricing based on bulk purchasing,

- some factors and/or flow on effects are unknown and vary in different situations, and

- the analysis includes immediate and direct expenses only and does not attempt to deal with complex issues such as lost productivity, long term life impacts, costs associated with remoteness or cultural factors.

Standard programme costs

Total standard programme costs for 1,000 infants were calculated for each cost category, based on the parameters above.

Figure 2 presents a comparison of annual costs of three screening regimes:

- AOAE, A-ABR/A-ABR,

- A-ABR/A-ABR with consumables,

- A-ABR/A-ABR without consumables.

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[139] All AOAE screening requires the use of consumables.

The first five sets of columns present a comparison of each of the five cost categories across the three regimes. The final two sets of columns combine these five categories. The first of the combined columns presents screening costs alone and the second presents screening costs combined with audiology costs.

Figure 2. Comparison of screening and audiology costs (NZ$) per 1,000 infants screened with AOAE, A-ABR/A-ABR and A-ABR/A-ABR with and without consumables.
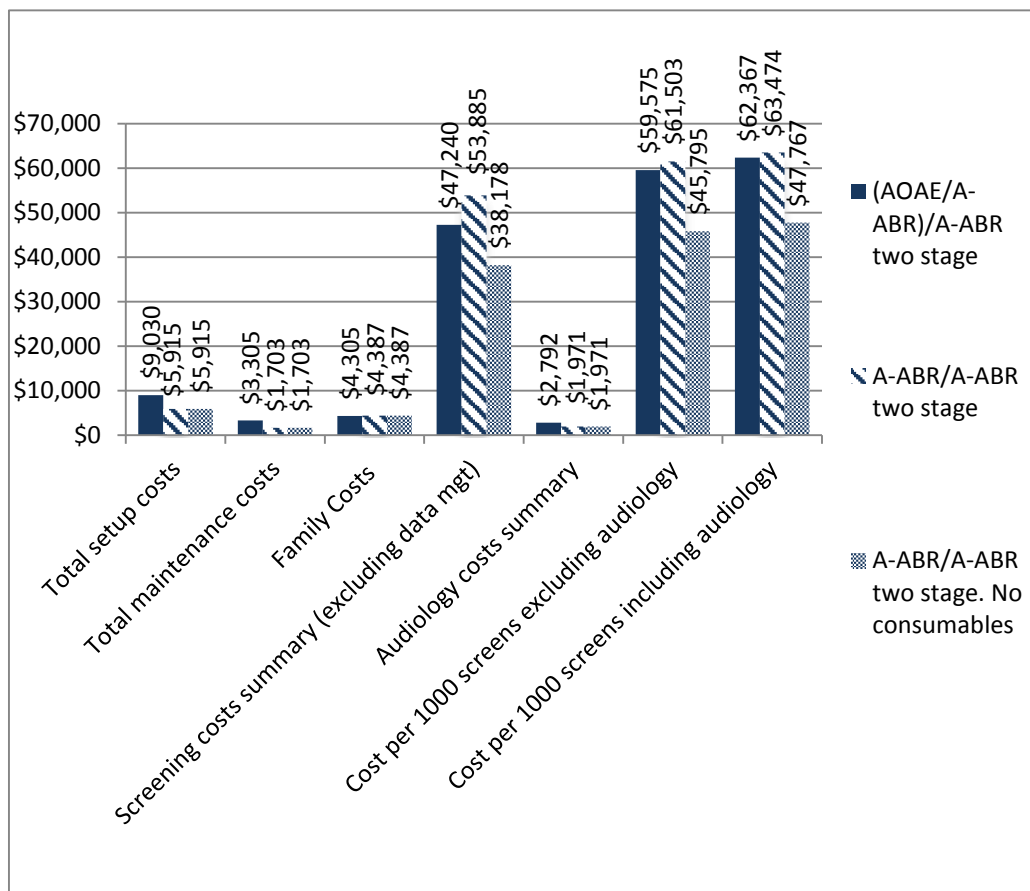
When comparing the three regimes, costs are lowest for the A-ABR/A-ABR regime using no consumables. This is the case for both screening alone ($45,795) as well as when screening and audiology costs are combined ($47,767). There are minimal differences between the overall costs for the AOAE, A-ABR/A-ABR regime ($62,367) and the A-ABR/A-ABR regime using consumables ($63,474).

Costs for an AOAE, A-ABR/A-ABR regime could not be significantly reduced through a change to an A-ABR device which does not require consumables, as a relatively small number of infants are screened using A-ABR under this regime.

Key observations that can be made from these comparisons include:

- Setup and maintenance costs are higher for the AOAE, A-ABR/A-ABR regime because this regime requires purchase of either two devices or two modules within the one device, compared to the A-ABR/A-ABR regime which only requires a single device.

- Although the AOAE, A-ABR/A-ABR regime has a high first refer rate, AOAE, A-ABR/A-ABR screening costs are slightly lower than screenings costs for the A-ABR/A-ABR regime using consumables. This is largely due to the direct impact of the cost of A-ABR consumables[140].

- The slightly higher audiology costs for the AOAE, A-ABR/A-ABR regime are due to the higher refer rate to audiology[141].

Refer rates for A-ABR screening

The A-ABR refer rates used in the cost analysis are estimates based on a combination of refer rates reported in the literature and refer rates from the programmes interviewed. As noted in the limitations section of the cost analysis, refer rates vary widely. An increase in a first or second refer rate will incur additional screening, co-ordination and family expenses, and for second refers, additional diagnostic expenses will be incurred.

Given that this analysis has identified a cost advantage for screening equipment which does not use consumables, a further analysis was done to examine the effects of higher refer rates, and whether this cost advantage remains in the presence of higher refer rates. For an A-ABR programme which uses consumables, for every 1% increase in first refer rate, an additional cost of $369 per 1,000 infants would be incurred. In contrast, for an A-ABR programme which does not use consumables, for every 1% increase in first refer rate, an additional cost of $223 per 1,000 infants would be incurred. For either type of A-ABR

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[140] This analysis has used averaged standard costs for A-ABR consumables – without the benefit of bulk pricing. Competitive tendering and purchasing arrangements, however, could significantly lower these costs.

[141] A refer rate of 1.52% from the AOAE, A-ABR/A-ABR regime to audiology was used. The refer rate for the UNHSEIP is 1.7%.

Ministry of Health. (2013). UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012

A refer rate of 0.95% from an A-ABR/A-ABR programmes was used. This figure is based on an average of the two-screen A-ABR/A-ABR programmes interviewed.

programme, where the second refer rate is increased by 1%, an additional cost of $1,915 per 1,000 infants would be incurred.

As a theoretical example, where both first and second refer rates are elevated – for example, 10% for the first refer rate and 4% for the second refer rate[142] – a cost advantage remains for A-ABR devices which do not use consumables. A likely or actual cost advantage for equipment which does not use disposable ear couplers has also been suggested by others[143, 144, 145].

Inpatient screening contrasted to community screening

All programmes require some outpatient screening and the standard costs in this analysis assume 10% of screening occurs post-discharge. However, UNHSEIP community screening rates in some DHBs are much higher than this. In some DHBs the majority of infants are screened in the community.

The additional cost to screen an infant in a dedicated community screening clinic, rather than as an inpatient is approximately $20 per infant. This additional cost includes screener travel and setup time, screener mileage, time associated with client non-attendance and rescheduling based on a 20% non-attendance rate.

The Australian Government, Medical Services Advisory Committee report on universal neonatal hearing screening concluded that community screening is effective when 'piggy-backed' with other health checks and immunisation. This report also noted that losses to follow-up are more likely when long delays occur between rescreening or screening in the community after early discharge[146].

It is important to note that reducing community screening and increasing inpatient screening would not result in programme savings. For such a change to be achieved successfully, the additional cost required to undertake community

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[142] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia.

[143] White et al. (2008). A comparative study of the MB 11 BERAphone and ABAER automated auditory brainstem response newborn hearing screening equipment.

[144] van den Berg et al. (2010) MB 11 BERAphone hearing screening compared to ALGO portable in a Dutch NICU: a pilot study.

[145] Cebulla, Shehata-Dieler. (2012). ABR-based newborn hearing screening with MB 11 BERAphone® using an optimized chirp for acoustical stimulation.

[146] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee.

screening would need to be redirected to the inpatient context to establish mechanisms for achieving optimal inpatient coverage and meeting programme benchmarks and outcomes.

Capture rates

While, for this exercise, a 100% capture rate has been assumed, all programmes report some infants as lost to follow-up.

The average capture rate from the international programmes reviewed was 98.0% (with a range of 96.5% to 99.5%). The reported capture rate for the UNHSEIP is 83.0%.[147]

If an annual birth rate of 60,000 is assumed, a capture rate of 83% would mean that 10,200 infants would not be screened. Further, if it is assumed that one child per thousand would normally be diagnosed with the target condition (permanent bilateral hearing loss of moderate or greater degree) through the screening programme, then approximately 10 infants with the target condition might not be detected early, and a further 20 infants' mild or unilateral hearing losses could also remain undetected at birth.

Although some direct costs associated with the non-detection of auditory neuropathy have been estimated for comparisons between AOAE and A-ABR regimes, the cost of low capture rates and subsequent late detection of permanent hearing loss have not been estimated in this exercise. Unlike auditory neuropathy which is not detected by AOAE programmes, low capture rates are not necessarily linked with any particular type of regime. Also as noted above, the costing of late detection involves complex issues such as lost productivity, and long term life impacts which are beyond the scope of this exercise.

Supporting the maintenance of quality standards

Risks affected by regime choice

A range of potential risks can impact upon a universal hearing screening programme. These risks can arise out of the screening regime, specific screening

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[147] Ministry of Health. (2013). UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012.

protocols, the screening workforce, and data management systems, just to name a few. As just one of these many variables, the capacity for regime choice to mitigate risk can only reach so far.

In the context of the UNHSEIP screening incident, the most conspicuous risk influenced by the current regime using an AOAE as the first screen, is the capacity for a screener to screen their own hearing and falsify screening records to suggest that a specific infant's hearing has been screened.

Performing an AOAE screen on one's self is a simple task. The process simply requires inserting the probe into the ear canal and running the screen. Many AOAE devices do not measure the ear canal volume to enable differentiation of adult and child responses. Whilst it is not impossible to self-screen utilising as A-ABR, it is more difficult than with an AOAE. The reasons include:

- some A-ABRs require electrodes to be adhered to the skin, requiring time and correct placement,

- some A-ABRs utilise a headphone which comfortably fits a child's ear but would not comfortably fit an adult ear, and

- some A-ABR devices contain algorithms specifically designed for a newborn baby, therefore self-assessment would significantly prolong the screen and most likely end with a null result.

These factors are likely to deter self-screening, particularly given the time and effort required to undertake the process.

Although a regime that uses A-ABR alone will significantly reduce the risk of self-screening, the following risks will persist, regardless of the chosen regime:

- failure to seek parental consent for screening,

- recording parental decline of a screen, without offering screening,

- following a unilateral refer result on a first screen, deliberately or inadvertently screening the ear that received a pass result instead of the ear that referred,

- repeating screens on a baby who has passed screening, but recording the results as multiple different babies, and

- acts of human error, such as screening the same ear twice when using a piece of equipment that screens one ear at a time.

Quality management

Decisions about a screening regime must take into account regime-specific issues of operational quality management; systems level programme monitoring, management, and quality improvement; and impacts on achievement of international benchmarks.

Quality management systems for regimes that combine two screening approaches (namely AOAE and A-ABR), and involve anything more than two screens, are inherently more demanding and complex than those that use a single approach to screening and a maximum of two screens. Specific domains of increased complexity include:

- data management for two screening approaches,

- greater follow-up and tracking in the context of a higher first screen refer rates and community screening result from AOAE screening,

- foundational screener training, training updates and scheduled and random competency checks and assessments for two different screening approaches and in some circumstances two different screening devices,

- optimal maintenance of two different screening devices or modules, and

- programme auditing.

Regardless of the regime used, effective data systems are key to being able to monitor programme quality and performance. The need to track infants and reduce rates of infants who are lost to follow-up is critical in all programmes and these factors have been a major driver for screening programmes around the world establishing or improving data systems.

CHAPTER 8 – Devices for newborn hearing screening

Introduction

Given that both published literature and the review of international programmes demonstrates that the lowest rate of referrals to audiology and false positive refer results is through an A-ABR/A-ABR regime, it was considered prudent to particularly examine devices that provide this capacity. In total, seven devices were reviewed. Some provide the option of AOAE as well as A-ABR screening. One device also provides additional screening capabilities. While this may seem like a positive attribute, extending the role of screeners would increase screening complexity, training, performance monitoring and data management.

Criteria for reviewing screening devices

All screening devices have strengths and limitations. The criteria listed below are considered important to the selection of a device for A-ABR screening. Several of the criteria are considered to be particularly important, these include high sensitivity and specificity, low false positive and false negative rates, scientific validation, minimised risk of falsification, portability, and monitoring and data security. Each of the seven devices reviewed meet these criteria to varying degrees.

Ideally a screening device should:

- have sensitivity rates of >95% and specificity rates of >90%,

- be validated through peer reviewed studies published in international journals, with appropriate sample size and scientific methodology,

- have a validated detection algorithm,

- be user friendly/functional for the screener workforce,

- minimise ease of falsifying screening results,

- enable data monitoring and data security,

- be time effective relative to other devices,

- contribute to cost effective screening outcomes,

- be portable,

- be consumer friendly,

- enable efficient data upload and download,

- be calibrated to an internationally published reference,

- have an effective, efficient and flexible computing interface,

- incorporate comprehensive support, including training, and

- be compatible with current and future technologies.

Device comparisons

Table 4 at the end of this section presents a summary of the key attributes of a variety of screening devices that provide A-ABR screening, with or without the capacity to also provide AOAE screening. Not every device attribute is included in the table. Technical specifications are detailed and the table provides a summary of key features and points of differences between devices. Information was obtained from manufacturer discussions, telephone and face-to-face meeting, journal articles, technical specifications and grey literature. Should there be a change of device used by the UNHSEIP, a tender process for supply of the same device to every DHB is recommended.

Ultimately, a single device holds many clinical advantages. Significant financial savings can also be obtained for bulk purchases of devices and consumables, and training and maintenance contracts. This could significantly assist in the planning of change, particularly with regard to the provision of ongoing training and competency assessment. While any potential risks are perceived as small, there are a few rare but possible risks including product recall, discontinued support of older products, effect of a monopoly, being locked into a specific type of technology for the term of the agreement, dealing with a new business owner or executive officer if the business is sold or taken over. Most of these risks can be managed through effective tendering processes and good contract management.

While the scope of information in Table 4 is extensive, three main devices have been cited in current published evidence more frequently than others, and therefore have a more rigorous validation process than other devices available on

the market. These are the two versions of the MAICO MB 11 (two devices using the same technology, with the MAICO MB 11 BERAphone using a monaural headphone and integrated electrodes and the MAICO MB 11 Classic using binaural headphones), the Natus ALGO 3i, and the MADSEN AccuScreen. It is important to keep in mind that devices meet the criteria to varying degrees. It is anticipated that these subtleties would be thoroughly explored in a formal tender process, should additional equipment be purchased. The advantages and disadvantages of equipment are discussed below. This information has been acquired from interviews, trials, discussions and technical specifications from manufacturers/suppliers of the equipment.

Table 4. Comparison of device features.

	Sentiero	GSI AUDIOscreener	MAICO MB 11 BERAphone	MAICO MB 11 Classic	ALGO 5	ALGO 3i	Titan	MADSEN AccuScreen
Manufacturer and/or supplier	Path Medical Solutions	Grason-Stadler	MAICO Sonic Innovations		Natus Medical Inc. Scanmedics	Natus Medical Inc. Scanmedics	Interacoustics	GN Otometrics
Screening capabilities	A-ABR click and CE chirp DPOAE ASSR	CE chirp A-ABR TEOAE DPOAE Click A-ABR	CE chirp A-ABR		Click A-ABR	Click A-ABR	1 kHz probe tone tympanometry DP or TE OAE screen CE chirp A-ABR High/low CE chirp A-ABR	DPOAE screen Click A-ABR screen Ultrashort chirp
Screen time		84 seconds (A-ABR only)	15 – 112 seconds		3 – 5 minutes	3 – 5 minutes	3 minutes	60 – 80 seconds with chirp
Stimulus level (dB)	35dBnHL	35dBnHL	35dBnHL	35dBnHL	35dBnHL	35dBnHL	35, 40 or 45 dBnHL	35, 40 or 45 dBnHL
Sensitivity	Unknown		99.3%	>99.0%	>99.0%	99.9%	99.7%	
Specificity	Unknown		97.9%	96.0 – 100%	96.0 – 100%	96.0%	98.3 – 99.5%	
First refer rate	Unknown	Unknown	2.1%	1.6%	1.6%	Unknown	2.6%	
Specified age range	Infant to adult	Infant to adult	Infant	Infant	Infant	Infant to adult (for diagnostic modules)	Infant	

	Sentiero	GSI AUDIOscreener	MAICO MB 11 BERAphone	MAICO MB 11 Classic	ALGO 5	ALGO 3i	Titan	MADSEN AccuScreen
Validation	Nil in literature	Nil as a combination unit (A-ABR and AOAE). One study for AOAEs, and adults only.	Large studies		Large studies	Large studies	Nil	Large studies
Portable	YES	YES	YES		NO	YES	YES	YES
Complex capabilities	Device has some complexities. Provides automated screening result. Additional features available for purchase (i.e. other assessments / capabilities).	Screener device. Provides automated screening result. Requires no interpretation of data.	Screener and clinical device. Provides automated screening results. Requires no interpretation of data. Diagnostic options depending on model selected.		Screener device. Provides automated screening results requiring no interpretation of data.	Screener device. Provides automated screening results requiring no interpretation of data.	Screener and clinical device. Provides automated results requiring no interpretation of data for AOAE and A-ABR. Some interpretation required for tympanometry.	Screener device. Provides automated screening results requiring no interpretation of data.
Power source	AC and re-chargeable battery	AC and re-chargeable battery	AA batteries (screening device) Internal rechargeable battery or power source (laptop or tablet)		Power source required	Internal rechargeable battery	Internal rechargeable battery	Internal rechargeable battery
Type of ear cups	Disposable ear cups.	Disposable ear cups – newborn to adult	BERAphone: Nil	Classic: Disposable ear cups	Disposable ear cups	Disposable ear cups	Disposable ear cups	Disposable ear cups
Screen memory	1,000 records on device	300 records on device	Infinite on PC/tablet		Infinite on PC	100 on device	Infinite on PC	250 on device
Warranty	1 year	1 year	1 year		1 year	1 year	1 year	1 year

	Sentiero	GSI AUDIOscreener	MAICO MB 11 BERAphone	MAICO MB 11 Classic	ALGO 5	ALGO 3i	Titan	MADSEN AccuScreen
Clinical benefits	Easy screen to navigate. Binaural screening.	Configurable AOAE pass criteria.	BERAphone & Classic: Data encryption Upgradeable software Small Light Portable / BERAphone: No consumables	Classic: Binaural screening	Binaural screening. Algorithm would slow significantly for an adult therefore deter from falsifying records. Upgradeable software.	Binaural screening. Hand held, portable device. Algorithm would slow significantly for an adult therefore deter from falsifying records. Upgradeable software.	Binaural screening. Full suite of testing. Possibility to add diagnostic modules and utilise through teleaudiology. Can be A-ABR only.	Intuitive touch screen. Slower screen time for an adult.
Clinical risks specific to the UNHSEIP	Not used in any other screening programme. Not designed specifically for newborn screening.	Not an intuitive screen.	BERAphone: Unusual placement on baby's head – additional training required. Can only screen one ear at a time – risk of using results of one ear as the second ear.	Classic: More pieces to carry in comparison to BERAphone (but still compact)	No flexibility for outpatient screening – on large trolley.		Complex due to other tests. No ear canal volumes on AOAEs. Tympanometry requires interpretation. Not used as a screening device in any other country.	No ear canal volumes on AOAE module. Data security.
Disadvantages specific to each device	No scientific literature.	No scientific literature. No upgradeable software.	BERAphone: Unusual placement.	Classic: Consumable costs (in contrast to BERAphone).	Large cart-mounted device. Suitable for hospital screening only.		Additional training due to larger suite of tests, and inclusion of tympanometry. High complexity.	Data security.

[A] Costs are subject to vary and should be used as a guide one.

Device clinical features and risks

The following information specifically focusses on the clinical advantages and disadvantages of the MAICO MB 11 BERAphone/MAICO MB 11 Classic, the Natus ALGO 3i and the MADSEN AccuScreen. These three devices have been selected on the basis that they are more rigorously validated in the available scientific literature, with regard to newborn hearing screening, in comparison to other devices.

MAICO MB 11 BERAphone/MAICO MB 11 Classic

The key benefits of the MAICO MB 11 BERAphone/MAICO MB 11 Classic include:

- portability,

- ease of use,

- encrypted data,

- zero consumables (MAICO MB 11 BERAphone only),

- fast CE chirp algorithm,

- validation,

- upgradeable software,

- use with a tablet, and

- instant upload of results.

Key disadvantages specific to the MAICO MB 11 BERAphone include:

- Restriction to single ear screening, which may increase the possibility of the baby waking between ears.

- Potential for confusion regarding which ear has been screened and which ear should be screened next. It should however be more difficult to confuse than with an AOAE given the preparation required for each ear, such as the application of gel to the baby's ear and subsequent removal of the gel. Protocols regarding the sequence of screening each ear are required to minimise this risk.

- Potential to deliberately or inadvertently screen a single ear rather than both ears. Following a unilateral refer result this opens the opportunity to rescreen the ear that passed rather than the one that referred

The first two disadvantages do not exist with the MAICO MB 11 Classic. However this device brings the disadvantage of the cost of consumables.

Natus ALGO 3i

The key benefits of the ALGO 3i include:

- portability,

- ease of use,

- encrypted data,

- specific algorithm designed for infant click A-ABR,

- validation,

- ease of use, and

- binaural screening.

The key disadvantages are:

- restriction to click A-ABR, therefore slower screen time

MADSEN AccuScreen

The key benefits of the AccuScreen include:

- portability,

- ease of use with a touchscreen,

- CE chirp A-ABR,

- validation, and

- binaural screening.

The key disadvantages are:

- the limited life of the touch screen, and

- no data encryption.

Cost of device and associated consumables

According to manufacturers, consumables are a highly negotiable cost. The MAICO MB 11 BERAphone does not use any consumables which brings a significant cost advantage. The ear cushions do require replacement with wear and tear, although this is not a frequent cost.

Infection control

Measures for infection control should be standard regardless of the device. Most devices recommend detergent wipes over any of the areas that come into contact with the neonate. While disposable ear cups can be thrown away, there is still a need to wipe down leads and any areas coming into contact with the baby. For babies or parents with MRSA, specific hospital guidelines need to be consulted. In these circumstances, some equipment specifies wiping down with 1,000ppm available chlorine. Consultation with the manufacturer and local infection control department is recommended prior to initiating any infection control method.

While individual hospitals may have differing requirements, a standardised approach to cleaning the device is recommended to ensure equipment is maintained appropriately, consistently and in keeping with infection control guidelines.

Risks

Unfortunately, every piece of equipment or protocol has risks and is open to compromise through inadvertent or deliberate acts.

With specific reference to the falsification of screening results, it is more difficult to self-screen using an A-ABR screening device than an AOAE screening device due to electrode placement and infant detection algorithms. However, there are other mechanisms through which falsification of records can occur. Screeners could enter patient data for multiple babies onto the device and commence screening a restful, sleeping baby, repeating the screen many times over, and labelling each screen as a different baby. Additionally, without making an offer or

a screen or seeking parental consent, it is possible to indicate parental decline of a screen.

Minimising this risk may include signed statements, including time and date, from parents indicating they have witnessed the screen and been advised of the results and subsequent actions required. However, this record could also be falsified. Ultimately, risk mitigation involves minimising the opportunity to self-screen; increasing training including understanding of reasons for standardised protocols and ethical standards; increasing competency assessments; incorporating random spot checks into daily practice; and increasing accountability within the screener role.

Whilst the above issues are pertinent to achieving intended behaviour, other issues such as protocol non-compliance will also challenge the integrity of the screen. For example, repeated screening attempts have the potential to increase false negative rates resulting in infants passing when in fact they have a permanent hearing impairment[148]. The more times a baby is screened the greater the likelihood of a confounding variable occurring (human error, noise etc.). This will interfere with a true screening result being obtained. False negatives are possible in all medical assessments and equipment. For this reason, standardisation of practice and a streamlined, consistent protocol is imperative. Poor adherence to protocols, including scripts, allows screeners to make decisions for which they are ultimately untrained, and are therefore unaware of the negative clinical impact.

Regardless of the extent to which attempts are made to mitigate risk, there will always be room for human error, such as inadvertently screening the same ear twice. Ensuring effective monitoring, an accountable work place culture, and an open-door policy to support remedying errors and facilitate knowledge and skill development is essential to all programmes.

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[148] Petticrew, M.P., Sowden, A. J., Lister-Sharp, D., Wright, K. (2000). False-negative results in screening programmes: systematic review of impact and implications. Health Technology Assessment. Health Technology Assessment, 4(5). Retrieved from: http://www.hta.ac.uk/fullmono/mon405.pdf

Training

All manufacturers are willing to provide extensive training and support. This could be negotiated in any tender process for a new device, and may include yearly training and competency checks.

CHAPTER 9 – Recommendations

Having reviewed current literature, a range of international programmes, the systems of the UNHSEIP and the New Zealand context the newborn hearing screening programme is delivered within, the review team has formulated three recommendations. The key factors guiding the formulation of these recommendations include supporting the UNHSEIP to:

- deliver optimal clinical efficacy and efficiency,

- facilitate diagnostic audiology assessment as early as possible,

- achieve optimal screening capture and completion rates,

- minimise impacts on families,

- achieve optimal operational efficiency, and

- meet specified quality standards.

Recommendations 1 and 2 speak directly to the review objective of 'examining best practice in newborn hearing screening regimes, including associated equipment options, to assist the National Screening Unit to determine the most appropriate screening regime for the New Zealand Universal Newborn Hearing Screening and Early Intervention Programme.'

Recommendation 3 details the conditions for success for achieving effective implementation of Recommendations 1 and 2.

Recommendation 1

1. Implement a two stage A-ABR screening regime for all neonates, including for neonates who have been under the care of a neonatal intensive care unit. Medical exclusions should continue, with direct referral to audiology.

Recommendation 2

Specify a standard screening device for all screening. The device must:

a. demonstrate sensitivity >95% and specificity >90%,

    b. be validated through peer reviewed studies published in international journals, with appropriate sample size and scientific methodology,

    c. have a validated detection algorithm,

    d. be user friendly/functional for the screener workforce,

    e. minimise ease of falsifying a screen,

    f. enable data monitoring and data security,

    g. be time effective relative to other devices,

    h. contribute to cost effective screening outcomes,

    i. be portable,

    j. be consumer friendly,

    k. enable efficient data upload and download,

    l. be calibrated to an internationally published reference,

    m. have an effective, efficient and flexible computing interface,

    n. incorporate comprehensive support, including training, and

    o. be compatible with current and future technologies.

Recommendation 3

Ensure the following conditions are met to facilitate both a successful change in regime and device, and to optimise the overall effectiveness of the programme:

    a. use a nationally managed organisational change process to facilitate implementation of the new regime and introduction of a nationally consistent device,

    b. standardise the screening regime, device, clinical practice and protocols nationally,

    c. prioritise inpatient screening, including reducing the minimum age of screening,

    d. review targeted follow-up criteria,

e. increase flexibility of workforce models, particularly in regional and rural areas,

f. strengthen current UNHSEIP continuous quality improvement processes by linking them to a set of international best practice benchmarks,

g. as a matter of priority, establish a national information system or data system which fulfils operational needs in real time as well as monitoring and reporting needs at DHB and national levels,

h. establish a regional system of operational management that transcends DHB boundaries and interfaces with the governance function of the NSU, and

i. build upon and continue existing expert, multidisciplinary, clinical advisory forums and processes to guide the implementation of the change to the regime and device, as well as provide ongoing feedback and advice regarding programme performance.

Examining the conditions for success

As noted above, Recommendation 3 details the conditions for success for achieving effective implementation of Recommendations 1 and 2. The discussion that follows elaborates on each of these points and discusses issues relevant to their application to the UNHSEIP.

Management of a national change process

To enable the successful nationwide implementation of the recommendations in this report, a consistent, comprehensive and nationally managed change process is required. Sponsorship from the NSU that is both visible and active in engaging with DHBs will be necessary. A change team will be required that includes representatives from all stakeholder groups. A change management plan that comprehensively details the objectives of the change; the specific change outcomes; the tasks required to achieve the change – including a detailed work programme and timelines; communication processes; an equipment selection process; and a risk assessment and risk management strategies will need to be developed. For each element of the change process, there will need to be clarity on who is accountable for the outcome being worked towards, who is responsible

for the component stage, who should be consulted and who needs to be informed of the work being undertaken and its outcomes.

Beyond preparing for change, the change plan must give detailed consideration to effectively implementing the changes. This includes monitoring their effectiveness in real time, and establishing mechanisms to reflexively respond to issues in a timely and systemic way.

It is critical that the change management plan attends to the varying needs of individuals and the impacts of change on their work experiences and practice. This is particularly significant given the impacts of the UNHSEIP screening event that precipitated this review; the extensive audit and review processes that have resulted; and the public scrutiny UNHSEIP staff have experienced for more than 18 months.

National standardisation of screening regime, device and protocols

Standardising the UNHSEIP screening regime, device and protocols is fundamental to achieving quality, ensuring cost effectiveness and meeting recognised international benchmarks. An important element to inform the standardisation of screening protocols is the adoption of a defined target condition. In most programmes across the globe the target condition is defined as moderate, bilateral permanent childhood hearing loss. Some programmes include the identification of ANSD and/or neural disorders.

Standardisation provides screeners with clarity regarding their role and its processes, assurance of the quality of their work when they comply with specified standards, and confidence in the appropriate path for seeking resolution to issues and challenges that arise which fall outside specified protocols.

Areas of the UNHSEIP that would benefit from greater definition and ensuring compliance with standardised protocols include:

- establishing a minimum screening age,

- prioritising inpatient screening,

- discontinuing the current protocol exception of allowing a third A-ABR screen in certain circumstances where an infant's screen has been completed under 72 hours of age,

- establishing clear standards detailing the exceptional circumstances in which a decision might be approved by a co-ordinator or audiologist for a screener to undertake a third A-ABR based on an assessment that the risk of an individual child not completing screening outweighs the risk of a false negative screening result,

- defining an 'incomplete' screen/'technical fail', and

- ensuring bilateral rescreening following a unilateral refer result.

Prioritisation of inpatient screening

Although the UNHSEIP has the target of infants completing screening by one month of age, no particular emphasis is placed on whether screening occurs immediately after birth in hospital or a birthing centre, or in a community clinic. There is great variability in inpatient screening rates, with some DHBs as low as 20.0% and others as high 90.0%. The absolute majority of universal newborn hearing screening programmes emphasise the importance of inpatient hearing screening if the international benchmark of 95.0% of infants being screened before one month of age is to be achieved. Infants who do not enter the screening pathway by receiving at least one screen before hospital discharge are recognised to be at higher risk of not completing hearing screening[149]. The exception to this is in circumstances where screening is incorporated as one component of a comprehensive universal community primary health care service.

If the UNHSEIP is to efficiently meet the 95% international screening benchmark, inpatient screening will need to be prioritised in every DHB, to the extent that it is possible. This will require review of the minimum screening age and introduction of more flexible workforce models.

Beyond the achievement of improved programme efficiency and success in meeting international benchmarks, inpatient screening significantly reduces the burden on families by removing the need to attend community screening.

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[149] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia.

Review of criteria for targeted follow-up

The rates of referral from screening to targeted follow-up were variable across the five international programmes that provided data to this review. Even so, at 5.6%, only one programme demonstrated a targeted follow-up rate as high as the 5.5% rate for the UNHSEIP[150]. The targeted follow-up rates of the remaining four programmes were 0.5%, 1.1%, 2.3%, and 3.7%.

Reviewing and implementing revised criteria for targeted follow-up and monitoring the consistent use of the criteria is recommended. Changes to these criteria have the potential to significantly reduce current demands on audiology services and enable delivery of timely audiology services to the community.

The current JCIH recommendations, recent literature, and the standards of other international programmes provide a valuable resource for this process.

Flexible workforce models

Workforce models for universal newborn hearing screening vary across the world with no clear evidence seeming to be available on the application of one model over another to specific screening regimes or contexts. Within different jurisdictions often a mix of approaches is needed to respond to varying demographic and resource variables; including varying birthing numbers in metropolitan, regional and remote contexts; different cultural contexts; and the models and location of maternity and postnatal services.

The UNHSEIP model of a dedicated screening workforce serves the programme well in many, but not all, contexts. The programme's capacity to meet international screening benchmarks, and the efficiency of achieving this, would be enhanced significantly if more flexible workforce models, including appropriate competency management systems and standards, were introduced that facilitated inpatient screening, seven days each week, in all but the smallest hospitals and birthing centres.

Specific opportunities for consideration include:

- incorporating newborn hearing screening into the roles of kaiāwhina and other community/health care workers,

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[150] Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012.

- vision hearing technicians employed through the B4 School Check,

- hospital and birthing centre nurses and midwives, and

- screening via telehealth where available, supervised by a skilled and experienced screener in situations of very low volume screening.

Quality improvement based on international benchmarks

The UNHSEIP specifies the internationally accepted standards of completion of screening by one month, completion of diagnostic audiology assessment by three months, and commencement of early intervention by six months, as its key goals. Although the UNHSEIP makes mention of some benchmarks regarding the proportion of infants meeting particular standards, it is essential that a more comprehensive set of accepted international benchmarks, in combination with local standards, are set for the UNHSEIP and routinely used as the measure against which the performance of the UNHSEIP is assessed and further developed.

More specifically, the UNHSEIP should focus its efforts on achieving the JCIH benchmarks[151] of:

- 95% of infants complete screening by one month corrected age.

- < 4% of infants are referred from screening to audiology.

- 90% of infants who are referred from screening to audiology complete comprehensive audiological evaluation by three months of age.

- 95% of infants with confirmed bilateral hearing loss whose families elect to use amplification receive amplification devices within one month of hearing loss confirmation.

- 90% of infants with confirmed hearing loss who qualify for early intervention services commence services no later than six months of age.

- 95% of children with acquired or late-identified hearing loss who qualify for early intervention services commence services no later than 45 days after diagnosis.

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[151] Joint Committee on Infant Hearing. (2000). Special Article, Year 2000 Position Statement.

- 90% of infants with confirmed hearing loss receive a first developmental assessment with standardised assessment protocols (not criterion reference checklists) for language, speech, and nonverbal cognitive development by no later than 12 months of age.

Benchmarks at a more detailed level (and in some areas, more stringent level) are also presented in the recently developed *National performance indicators for neonatal hearing screening in Australia*[152].

National database

Several international newborn hearing screening programmes interviewed noted that improving data systems is a key area for improvement. Existence of a data system per se does not guarantee availability and use of good information.

An evaluation of universal newborn hearing screening and intervention (UNHSI) programs operating in all states and many territories of the USA found that 'one of the most urgent challenges of UNHSI programs involves loss to follow-up among families whose infants screen positive for hearing loss'. They concluded that there were five key areas for future programme improvements and improving data systems to support surveillance and follow-up activities was the first key area[153].

Similarly, when considering screening programmes in the USA it has been noted that 'Although substantial progress has been made in the percentage of infants screened for hearing loss before hospital discharge, significant improvement is needed with respect to the availability of pediatric audiologists, implementation of effective tracking and data management systems, programme evaluation and quality assurance, availability of appropriate early intervention programs, and linkages with medical home providers.'[154]

The UNHSEIP review raised issues and concerns in relation to data. Changes to the regime and/or screening devices used for newborn hearing screening should

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[152] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia.

[153] Shulman, S., Besculides, M., Saltzman, A., Ireys, H., White, K. R., Forsman, I., Evaluation of the universal newborn hearing screening and intervention program, *Pediatrics, 126*(S1), S19-S27.

[154] White, K. R., Forsman, I., Eichwald, J., Munoz, K., The evolution of early hearing detection and intervention programs in the United States, *Seminars in Perinatology, 34*(2), 170–179.

not be undertaken independently of at least a thorough review, revision and redevelopment of data collection procedures.

A system implemented by the UNHSEIP must:

- Provide essential information to underpin the tracking of infants through what is usually a complex and multi-departmental pathway from birth, through screening, diagnostic audiology, medical assessment, family support, hearing augmentation and early intervention.

- Enable uploading of demographic and screening data, and reconciliation of the two sets of information based on a unique identifier.

- Minimise the requirement and opportunity for screeners to amend Excel or text files exported by the equipment as this would present an opportunity for falsification of results. Ideally, the data would not be able to be corrected or amended until the file is imported and available in the data system where any changes would be automatically audited. Procedures also need to be in place for thorough data auditing against raw data from the screening device.

- Ensure real time access to appropriate levels of information to all relevant stakeholders.

- Provide a standard set of reporting and auditing processes. These processes should be constructed so as to provide automatic alerts when protocols are not adhered to or benchmarks are not achieved.

- Ensure diagnostic audiology results can be input and matched to screening data to enable tracking of infants from screening and beyond audiology, including determination of performance against benchmarks for the timing of screening, diagnostic audiology, referral to medical services, and commencement of early intervention for each individual child.

Operational management

Beyond programme governance and the setting of strategic direction, all but one of the international programmes reviewed has a system of regional operational oversight, monitoring and management that extends across geographic and local service boundaries. These regional mechanisms play a critical role in maintaining service standards; identifying and responding to common challenges and needs in a consistent way; and achieving an optimal interface with the policy, funding, monitoring and quality improvement functions.

The UNHSEIP would be well served by such a regional operational management function that transcends DHB boundaries and is undertaken by individuals with high levels of knowledge and skill in universal newborn hearing screening as well as expertise in quality improvement and systems management.

Expert clinical advisory forums and processes

As detailed previously, the NSU both convenes and works with a number of forums to guide and facilitate the work of the UNHSEIP. These forums include the UNHSEIP Advisory Group, the Joint Ministries Group, the NSU Advisory Group, and the Paediatric Technical Advisory Group of the New Zealand Audiological Society.

These forums, their functions, and relationships to each other should be strengthened and further developed where gaps in expertise and contributions exist.

GLOSSARY

Audiologist: A university-trained professional who is specially qualified to measure hearing, diagnose the degree, configuration and type of hearing loss, advise on the non-medical management of hearing disorders, and supply and fit hearing aids and other hearing devices to suit[155].

Audiology: A field of research and clinical practice devoted to the study of hearing disorders, assessment of hearing, hearing conservation, and aural rehabilitation[156].

Auditory neuropathy spectrum disorder (ANSD): A relatively complex type of hearing loss that is believed to be due to abnormalities at the synapse of the inner hair cell and auditory nerve, and/or the auditory nerve itself[157].

Auditory brainstem response (ABR): An auditory brainstem response (ABR) is a physiological measure of the brainstem's response to sound. It tests the integrity of the hearing system from the ear to the level of the brainstem utilising click or tone stimulus. The rate of the stimulus presentation is typically much slower than the rates presented in an ASSR.

Automated auditory brainstem response (A-ABR): Recordings of the ABR performed with a highly automated and standardised procedure for data collection for the purpose of screening for hearing impairment. The presence of a response (pass) or absence (refer) at the screening intensity level of the stimulus is determined primarily by a clinically proven machine scoring algorithm operating on-line[158].

The A-ABR is an automated version of the auditory brainstem response test (also sometimes referred to as the Brainstem Evoked Response Audiometry BERA-test). The A-ABR uses an auditory stimulus, known as a 'click'. The A-ABR assesses the entire auditory pathway, including the retrocochlear pathway,

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155 Australian Government. (2013). National framework for neonatal hearing screening. Retrieved from http://www.health.gov.au/internet/main/publishing.nsf/Content/neonatal-hearing-screening

156 Audiology Society of Australia. (2013). Retrieved from www.audiology.asn.au

157 National Acoustics Laboratory. (2013). About hearing loss: Hearing problems. Retrieved from: http://www.nal.gov.au/hearing-loss_tab_auditory.shtml

158 NHS Newborn Hearing Screening Programme. (2013). Screening information. Retrieved from: www.nhsp.info/workbook.shtml

making it sensitive to the detection of auditory neuropathy spectrum disorder (ANSD).

Automated brainstem response compared to auditory steady-state response (ASSR): The following is an excerpt contrasting ASSR and ABR[159].

ASSR and ABR use many of the same basic tools and protocols; thus, it is reasonable to compare and contrast the two. The similarities between ABR and ASSR include:

- *Both deliver an auditory stimulus,*

- *Both stimulate the auditory system,*

- *Both record bioelectric responses from the auditory system via electrodes, and*

- *In each protocol, the patient does not have to respond volitionally.*

The differences between ABR and ASSR include:

- *ABR stimulus is usually a click or a tone burst (one tone and one ear at a time) presented at a slower rate, whereas ASSR uses amplitude or frequency modulated sounds presented rapidly to excite the auditory system while stimulating four frequencies and both ears simultaneously.*

- *ABR is highly dependent on a relatively subjective analysis of amplitude versus latency. ASSR is dependent on a statistical analysis of the probability of a response, usually at a 95% confidence level.*

- *The ABR response is measured in millionths of a volt (microvolts), and the ASSR is measured in billionths of a volt (nanovolts).*

Automated otoacoustic emissions (AOAE): An automated OAE is performed during a hearing screen and generally offers no further information other than a "pass" or "refer" result. An otoacoustic emission is a response or echo from the cochlea. The AOAE can consist of distortion product otoacoustic emissions (DPOAE) or transient evoked otoacoustic emissions (TEOAE). This is

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[159] Beck, D.L., Speidel, D.P., Petrak, M.S. (2009). Auditory steady-state response (ASSR): A beginner's guide Retrieved from:
http://www.interacoustics.de/eprise/main/de_de/Content/Articles/PDFs/ASSR_BeginnersGuide_HR-12-2007.pdf

different from an OAE, which provides detailed clinical data to be interpreted only by a diagnostic audiologist.

Bilateral hearing loss: A hearing impairment in both ears[160].

Chirp stimulus: Chirps are a more recent stimuli which provide several advantages, including an increase in waveform amplitude. Some ASSR systems use special chirp stimuli which have detection algorithms which decrease screen times and increase the speed of data collection. ASSR has research supporting its reliability and effectiveness in the prediction of hearing thresholds[161]. There are various types of chirp including CE, bandwidth and narrowband.

Conductive hearing impairment: Conductive hearing impairment occurs when sound is not conducted efficiently through the outer ear canal to the eardrum and the tiny bones (ossicles) of the middle ear[162].

Corrected age: Corrected age takes into account the time between premature birth and the actual due date of a full term pregnancy. Calculating corrected age provides a truer reflection of what the baby's developmental progress should be[163].

Decibel (dB): The unit of measurement for the loudness of a sound. The higher the decibel level, the louder the sound.

Degree of hearing impairment: Describes the impact of a measured hearing loss on an individual's communication ability. Hearing levels are measured in the better ear:

> **Mild: 26 – 40 dB.** Affected individuals are able to hear and repeat words spoken in a normal voice at a distance of one metre. Speech and language usually develop normally if a child is fitted with hearing aids early.

> **Moderate: 41 – 60 dB.** Affected individuals can hear and repeat words spoken in a raised voice at a distance of one metre. Speech and language

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[160] Australian Government. (2013). National framework for neonatal hearing screening.

[161] Beck, D.L., Speidel, D.P., Petrak, M.S. (2009). Auditory steady-state response (ASSR): A Beginner's Guide.

[162] American Speech Language and Hearing Association. (2014). Permanent childhood hearing loss. Retrieved from: http://www.asha.org/public/hearing/Conductive-Hearing-Loss/

[163] Australian Government. (2013). National framework for neonatal hearing screening.

development are generally affected if a hearing aid is not provided early to a child born with this degree of loss.

Severe: 61 – 80 dB. Affected individuals are able to hear some words when shouted into the better ear. Speech and language do not develop spontaneously. Hearing aids will greatly assist a child to develop speech, but speech quality is likely to be affected.

Profound: 81 dB or greater, including deafness. Individuals with this level of impairment are unable to hear and understand a shouted voice. Learning to speak is difficult for children born with a profound hearing loss. Many children with profound hearing loss are now fitted with a cochlear implant (Australian Hearing 2005).

Diagnostic audiology assessment: An assessment that occurs after a child has received a „refer" result in a second hearing screen. The assessment is performed by an audiologist, and includes diagnostic hearing tests to assess the type and degree of hearing impairment[164].

Distortion product otoacoustic emission (DPOAE): The DPOAE is also an echoed response which reflects the outer hair cell activity within the cochlea. The DPOAE measures sound waves generated in the inner ear (cochlea) in response to a continuous signal with two simultaneous but different frequencies. These are emitted and recorded via very sensitive miniature microphones. The microphones are situated in the external ear canals of the infant within a tiny flexible plug. In the analysis of the response, the integrity of the auditory pathway up to the cochlea or inner ear is assessed, including the middle and inner ear. This assessment does not assess the integrity of the entire auditory pathway[165].

Double refer: A double refer occurs when a child has not passed the screen on two separate occasions and further investigation is required by an audiologist[166].

Early intervention programs: Programs which aim to provide hearing impaired children in the first six months of life with immediate intervention.

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[164] Australian Government. (2013). National framework for neonatal hearing screening.

[165] OAE Forum. (2012). Basics of OAEs. Retrieved from:
http://www.otoemissions.org/old/definitions/DPOAE.html

[166] Australian Government. (2013). National framework for neonatal hearing screening.

Children who undergo early intervention have significantly better outcomes than later-identified children in both speech and social-emotional development[167].

Electrophysiological test: Electrophysiological tests measure the physical response of a specific part of the auditory system to sound. Results from electrophysiological tests can also be helpful in determining which part of the complex auditory (hearing) system is involved in a hearing loss[168].

False negative: False-negative results occur when a screen cannot, for a range of possible reasons, detect the presence of indicators of the condition or disease of interest (Pettigrew et al., 2000). In hearing screening this refers to the proportion of infants not identified as having a significant hearing impairment by screening who are subsequently found to have a significant hearing impairment.

False positive: False positive results occur when a screen identifies a condition, but for whatever reason, the child goes on to not test positive for the condition. In hearing screening this means the proportion of infants identified as having a significant hearing impairment by the screening process who are subsequently found to not have a significant hearing impairment.

First refer: A refer occurs when a child does not pass the initial newborn screen.

First refer rate: The percentage of children who do not pass the initial newborn screen. It is calculated by dividing the number of infants who fail the initial screen by the total number of infants to be screened.

Hearing aid: An electronic device that amplifies sound and conducts it to the ear.

Hearing screening: Hearing screening aims to identify children who are at risk for a hearing loss, so that they can be referred for further detailed assessment. A screening result can be a pass (hearing is at levels required for normal speech and language development at the time of screen) or refer (at risk for hearing loss and requiring further assessment). Infants in Australia have their hearing screened with either A-ABR or AOAE[169].

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[167] Australian Government. (2013). National framework for neonatal hearing screening.

[168] ibid.

[169] ibid.

Informed consent: In order to provide informed consent, a consumer needs to know what options are available, what the expected outcomes are for each option, and what the success rates and incidence of side-effects are for each option[170].

Initial screen: The first hearing screen that occurs after a baby is born, within 24 – 72 hours of birth[171].

Otoacoustic emissions (OAE) test: The OAE test measures the response of the outer hair cells in the inner ear (cochlea) to sound. A small probe is placed in the ear canal. A series of clicks or tones is presented to the child's ear and a small microphone records echoes (emissions) that come from the cochlear. There are two types of OAE – TEOAE and DPOAE. These are also explained in this glossary[172].

Pass (negative): No hearing loss is detected at the initial newborn hearing screen, or at the subsequent rescreen. A negative screen result[173].

Permanent childhood hearing impairment (PCHI): Permanent childhood hearing impairment can be congenital, delayed-onset, progressive, or acquired in nature. Congenital hearing impairment refers to hearing impairment that is present at birth and is often identified through a newborn hearing screening conducted shortly after birth[174].

Refer rate: The percentage of children who refer from any given screen for further assessment (e.g. for a second screen or for diagnostic audiology).

Refer rate to audiology: The percentage of children who refer from the final screen for further assessment (i.e. for diagnostic audiology).

Rescreen: A second screening for babies who do not pass the initial screen. The rescreen should occur after 24 hours but within two weeks of the initial screen[175].

Screening stage: This is also sometimes called a session. It can be conceived of as an appointment where one, or for some regimes, two screens/s are

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[170] Australian Government. (2013). National framework for neonatal hearing screening.

[171] ibid.

[172] ibid.

[173] ibid.

[174] American Speech Language and Hearing Association. (2014). Permanent childhood hearing loss. Retrieved from: http://www.asha.org/Practice-Portal/Clinical-Topics/Permanent-Childhood-Hearing-Loss/

[175] Australian Government. (2013). National framework for neonatal hearing screening.

undertaken. Most commonly, regimes are two stage regimes – with a period of time (ideally a minimum of 24 hours) between the stages. Regimes considered by the review included one/two and three stage regimes.

Second refer: A refer occurs when a child does not pass the second newborn screen.

Second refer rate: The percentage of children who do not pass the second newborn screen. It is calculated by dividing the number of infants who fail the second screen by the total number of infants to be screened. For two stage programmes this is the same as the 'refer rate to audiology'.

Sensitivity: The sensitivity of a clinical test refers to the ability of the test to correctly identify those patients with the disease.

Sensorineural hearing impairment: Sensorineural hearing impairment results from damage to or disorders of the inner ear, which includes the cochlea, eighth cranial nerve, and the cochlear nuclei[176].

Specificity: The specificity of a clinical test refers to the ability of the test to correctly identify those patients without the disease.

Target condition: Babies with congenital permanent bilateral, unilateral sensory or permanent conductive hearing loss, including neural hearing loss, of greater than 40dB[177].

Transient otoacoustic emissions (TEOAE): The TEOAE is an echoed response which reflects the outer hair cell activity within the cochlea. The TEOAE measures sound waves generated in the inner ear (cochlea) in response to clicks or tone bursts. These are emitted and recorded via sensitive miniature microphones. The microphones are situated in the external ear canals of the infant within a tiny flexible plug. In the analysis of the response, the integrity of the auditory pathway up to the cochlear or inner ear is assessed, including the middle and inner ear. This assessment does not assess the integrity of the entire auditory pathway[178].

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[176] National Acoustics Laboratory. (2013). Hearing problems. Retrieved from: http://www.nal.gov.au/hearing-loss_tab_auditory.shtml

[177] Australian Government. (2013). National framework for neonatal hearing screening.

[178] Kemp, D. (2011). Use of DPOAEs in assessment following screening. NHSP Clinical Group. Retrieved from: http://hearing.screening.nhs.uk/searchwebsite.php?searchstring=tag:audiology

Unilateral hearing loss: A hearing impairment in one ear.

Wide-band acoustic immittance (WAI): A new technique to measure middle ear function.

APPENDIX A – Literature review search terms

#AABR

#AOAE/AABR

#AOAE

#Benefits/risks/advantages/disadvantages of AABR

#Benefits/risks/advantages/disadvantages of AOAE

#Benefits/risks/advantages/disadvantages of AOAE/AABR

#Benefits/risks/advantages/disadvantages of CE chirp

#Evaluating AABR

#Evaluating newborn hearing screening regimes

#Evaluating newborn hearing screening protocols

#ABR and neonatal hearing screening

#AOAE/AABR and neonatal hearing screening

#CE chirp

#AABR CE chirp

#Costs of neonatal hearing screen

#Costs of newborn hearing screen

#Cost efficiency of newborn hearing screen

#Cost effectiveness of newborn hearing screen

#Cost effectiveness of neonatal hearing screen

#Cost of establishing a newborn hearing screening programme

#Equipment used for neonatal hearing screening

#Equipment used for newborn hearing screening

#Devices for newborn hearing screening

#BERAphone AABR

#BERAphone chirp AABR

#ALGO AABR

Audioscreener

#Intelligent Hearing Systems

#SmartScreener-Plus 2

#SmartScreener

#OtoRead

#AccuScreen

#MB 11

#ALGO 5

#ALGO 3i

#Echo-Screen®

#ABaer®

#AuDX® I, AuDX® Pro, AuDX® Pro II, AuDX® Pro Plus

#Scout Sport

#ILO288 Echoport USB 1 or 2

#Otoport Screener

#Otocheck

#Otoport DP & TE

#Otoport Advanced

#Aurix

#ASSR CE chirp

#Refer rates for AABR

#Refer rates for AOAE

#Refer rates for AOAE/AABR

#Practical issues newborn hearing screen

#Follow up rates newborn hearing screening

APPENDIX B – Literature review summary of bottom line statements

Technologies on the horizon

Evidence from current research does not suggest that genetic testing, multi-frequency ASSR, or WAI are ready for implementation in a universal newborn hearing screening programme. Further research is required to validate and provide further information for test development and validation prior to use within a screening regime.

Screening regimes using AOAE only

AOAE regimes have a high false positive rate and therefore a high rate of referral, in comparison to other regimes. AOAE regimes do not detect ANSD.

Screening regimes using a combination of AOAE and A-ABR

AOAE/A-ABR regimes have a much lower false positive rate and therefore a lower rate of referral, in comparison to AOAE regimes. AOAE/A-ABR regimes do not detect ANSD, and may not detect mild hearing impairments.

Screening regimes using A-ABR only

A-ABR/A-ABR regimes have the lowest false positive rate and the lowest rate of referral, in comparison to AOAE and AOAE/A-ABR regimes. A-ABR/A-ABR regimes also identify ANSD. A-ABR/A-ABR regimes typically do not identify mild impairments due to the target condition of moderate or greater hearing impairment, but this can be adjusted in most equipment in consultation with manufacturers, if desired.

CE chirp stimulus

CE chirp ASSR/A-ABR is a reliable and validated screening tool. Chirp regimes have similarities to A-ABR regimes.

APPENDIX C – International programme reviews: interview questions

Review of Newborn Hearing Screening Regimes and

Associated Screening Devices

International Programme Reviews – Interview Questions

1. What screening regime is used by your programme?

2. What equipment is used by your programme?

3. What was the background to using the current screening regime and screening devices?

4. What is the balance of community to inpatient screening? What influences this arrangement?

5. What are the workforce arrangements of the programme?

6. How is data collected when the baby is born?

7. How is data collected when the baby is screened?

8. Who has access to and manages the data locally?

9. What audit processes are in place to ensure integrity of data?

10. Who manages the data at other levels, e.g. district, state, national?

11. How is the information from newborn hearing screening data used?

12. What are the governance arrangements for your programme?

13. Has your programme made any changes to its regime or equipment in recent years? What prompted these changes? What outcomes have resulted?

14. If you had the opportunity, what changes would you like to make to the programme regime or devices?

15. Is anything stopping your programme from making these changes?

16. What are the most significant strengths of your programme?

APPENDIX D – International programme reviews: data collection

Note: Please include number of infants - not percentages - Thank you.																
UNHS Data Request - Review of NZ UNHSEIP																
Name of programme	12 month period - start date - eg. 1/1/2012	12 month period -end date - eg. 31/12/2012	Screening regime - (CHOOSE FROM DROP DOWN LIST)	Number of infants screened in hospital prior to discharge	Number of live births	Number of eligible births*	Number declined screening	Number completed screening	Number screened < 30 days	Number referred on first screen	Number referred to diagnostic audiology	Number identified for targeted follow-up**	Number diagnosed PCHI - bilateral, moderate or greater in better ear	Number diagnosed PCHI - bilateral or unilateral, moderate or greater in better ear	Comments, e.g. issues unique to your jurisdiction; anticipation of changes to screening regime, etc.
* i.e. Live births excluding infants who have moved, neonatial deaths, nedically excluded																
** If follow up of infants with risk factors is part of your program - please leave blank if you do not do this																

APPENDIX E – UNHSEIP documents reviewed

Programme governance

Terms of Reference, UNHSEIP Advisory and Working Groups, National Screening Unit, Ministry of Health, September 2011.

Policies and Processes

UNHSEIP Co-ordination Activities for DHBs, National Screening Unit, Ministry of Health, August 2013.

Newborn Screening – Free Health Checks for Your Baby: Newborn Hearing Screening, National Screening Unit, Wellington, Ministry of Health, September 2008.

Newborn Screening – Free Health Checks for Your Baby: Newborn Hearing Screen Results, National Screening Unit, Wellington, Ministry of Health, September 2008.

Newborn Screening – Free Health Checks for Your Baby: Repeat Newborn Hearing Screen, National Screening Unit, Wellington, Ministry of Health, September 2008.

Newborn Screening – Free Health Checks for Your Baby: Referral to Audiologist, National Screening Unit, Wellington, Ministry of Health, September 2008 reprinted April 2009.

Quality management

UNHSEIP National Policy and Quality Standards, National Screening Unit, Wellington, Ministry of Health, June 2013.

UNHSEIP National Policy and Quality Standards – Appendix F: Diagnostic and Amplification Protocols, National Screening Unit, Wellington, Ministry of Health, June 2013.

UNHSEIP monitoring framework, National Screening Unit, Wellington, Ministry of Health, September 2009.

UNHSEIP protocol for monitoring of individual screener data, National Screening Unit, Wellington, Ministry of Health, July 2013

Quality improvement review of a screening event in the UNHSEIP, National Screening Unit, Wellington, Ministry of Health, December 2013.

UNHSEIP screening incident – recommendations implementation plan, National Screening Unit, Wellington, Ministry of Health, August 2013.

UNHSEIP Electronic Interface Technical Specification, National Screening Unit, Wellington, Ministry of Health, April 2012.

UNHSEIP Web-Based Application for Screening and Audiology Data: User Guide, National Screening Unit, Wellington, Ministry of Health, July 2013.

UNHSEIP Summary of DHB Service Audit Programme to July 2013, National Screening Unit, Wellington, Ministry of Health, July 2013.

UNHSEIP Audit: Southern District Health Board, Ministry of Health, New Zealand Government & Deloitte, March 2013.

UNHSEIP Audit – Action Plan: Southern District Health Board, Ministry of Health, New Zealand Government & Deloitte, March 2013.

UNHSEIP Audit: Tairawhiti District Health Board, Ministry of Health, New Zealand Government & Deloitte, February 2013.

UNHSEIP Audit – Action Plan: Tairawhiti District Health Board, Ministry of Health, New Zealand Government & Deloitte, February 2013.

Workforce

UNHSEIP Newborn Hearing Screening and Audiology Workforce Strategy and Action Plan, National Screening Unit, Wellington, Ministry of Health, June 2008.

UNHSEIP Workforce Survey Responses – Question 14, National Screening Unit, Wellington, Ministry of Health, 2012.

Training

UNHSEIP Newborn Hearing Screener Training, National Screening Unit, Wellington, Ministry of Health, June 2012.

UNHSEIP Screener Manual, Appendix 2: Glossary, National Screening Unit, Wellington, Ministry of Health, June 2013.

UNHSEIP Screener Manual, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 1: Programme Information, National Screening Unit, Wellington, Ministry of Health, August 2013.

UNHSEIP Screener Manual, Module 2: Understanding Hearing Loss and How Newborn Hearing Screening Works, National Screening Unit, Wellington, Ministry of Health, August 2013.

UNHSEIP Screener Manual, Module 3: The Maternity Unit and Patient Management, National Screening Unit, Wellington, Ministry of Health, August 2013.

UNHSEIP Screener Manual, Module 4: Pre-Screening Communication, National Screening Unit, Wellington, Ministry of Health, August 2013.

UNHSEIP Screener Manual, Module 5: Screening Well Babies, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 6: Screening Babies in Neonatal Care, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 7: Screening in the Community, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 8: Communicating Screening Outcomes, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 9: Care of Equipment, National Screening Unit, Wellington, Ministry of Health, September 2013.

UNHSEIP Screener Manual, Module 10: Overview of Referral to Audiology and Support Services, Wellington, Ministry of Health, September 2013.

Data

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, October 2009 – March 2010, National Screening Unit, Wellington, Ministry of Health, August 2011.

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, April 2010 – September 2010, National Screening Unit, Wellington, Ministry of Health, November 2011.

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, October 2010 – March 2011, National Screening Unit, Wellington, Ministry of Health.

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, April 2011 – September 2011, National Screening Unit, Wellington, Ministry of Health.

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, October 2011 – March 2012, National Screening Unit, Wellington, Ministry of Health.

UNHSEIP Monitoring Report on Newborn Hearing Screening Service Provision, April 2012 – December 2012, National Screening Unit, Wellington, Ministry of Health.

New Zealand Deafness Notification Data: January – December 2003, Auckland District Health Board, June 2004.

New Zealand Deafness Notification Data: January – December 2004, Auckland District Health Board, June 2005.

New Zealand Deafness Notification Data: January – December 2005, Auckland District Health Board, March 2007.

Deafness Notification Report 2010, New Zealand Audiological Society, April 2011.

Deafness Notification Report 2011, New Zealand Audiological Society, August 2012.

Deafness Notification Report 2012, Accessable: Environmental Health Management Services, May 2013.

Deafness Notification Database: Information for Audiologists, December 2011.

UNHSEIP Stocktake of Screening Equipment, National Screening Unit, Wellington, Ministry of Health, 2013.

APPENDIX F – Purpose and parameters of the cost analysis

Programme costs are key in decision making regarding a newborn hearing screening regime. Principle 9 of the World Health Organization guidelines in relation to screening states that 'The total cost of finding a case should be economically balanced in relation to medical expenditure as a whole.'[179] As one element of this review a cost analysis was undertaken, with the following aims:

- to enable consideration of cost comparisons for varying regimes, and

- to examine where variations in some aspects of a regime, or of screening practice, will affect the overall costs.

The standard regimes[180] included in the literature review were considered, with particular emphasis on two regimes, namely, AOAE, A-ABR/A-ABR (the regime closest to the one currently used by the UNHSEIP) and A-ABR/A-ABR[181].

There are a few important points to note about this costing analysis:

- it provides relative costs only and is not a budgeting exercise,

- it does not comprehensively cover all screening costs (e.g. infrastructure costs),

- it should not be used as the sole basis for decision-making regarding a newborn hearing screening regime, and

- it should not be used to formulate a programme budget.

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[179] World Health Organization. (1968). Principles and practice of screening for disease. Retrieved from: http://whqlibdoc.who.int/php/WHO_PHP_34.pdf

[180] Screening regimes that only use a single screen are not examined due to the inherently high refer rates and consequent diagnostic costs associated with them.

[181] Analyses for all regimes and detailed assumptions and costs used to inform the analysis are included at Appendix F.

Cost categories and analysis parameters and variations

A standard set of notional costs were calculated for five key cost categories:

- **Initial setup costs:** office setup, initial training, and equipment purchase (conservatively assuming a five year equipment lifespan).

- **Equipment maintenance costs:** annual supplier maintenance, local screener and co-ordinator maintenance – daily, weekly, monthly and annual.

- **Family costs where screening is not completed in hospital:** mileage and attendance costs (e.g. parking, babysitting).

- **Direct screening costs:** consumables, labour for each screen, additional labour for refer rescreens, co-ordination for refer results, management, additional costs for community screening (assuming 10% community screening rate), tracking of lost to follow-up or referred infants, screener training and competency assessment, additional hearing assessment and therapy for infants not diagnosed with ANSD in the context of a regime using AOAE as the first screen.

- **Diagnostic costs:** assessment of false positives (two appointments), assessment of permanent bilateral moderate or greater hearing loss, and other hearing losses (four appointments), data entry and report writing.

Data management costs were not included in the screening estimates due the difficulty of establishing a realistic average cost given the wide variety of data management processes used

The parameters for the analysis of notional annual costs are set out below. Annual costs for each regime were based on:

- full capture of 1,000 eligible well-baby births[182],

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[182] The review team acknowledges that international best practice requires the use of A-ABR screening for infants who have spent five days or more in NICU, however for this exercise the cost impacts of this difference were not assessed to be of a sufficient scale to make this distinction.

- use of the same: screening workforce; A-ABR first, second and/or third refer rates; AOAE first, second and/or third refer rates; lost to follow-up rate; and outpatient screening rates,

- use of consumables,

- current costs expressed in (NZ$),

- additional time being allocated to A-ABR screening (30 minutes) compared to AOAE (20 minutes), and

- incorporation of some conservative estimates of: costs to families[183]; and additional costs associated with non-detection of auditory neuropathy for regimes which begin with AOAE.

Variations to these standard costs are examined, including:

- the cost of screening alone, in contrast to the cost of screening and audiology combined,

- the cost of A-ABR screening with consumables, in contrast to the cost of A-ABR screening without consumables[184],

- the additional cost of screening infants in community clinics, rather than as inpatients.

Limitations

It is important to note a number of important limitations of this cost analysis exercise:

- reported refer rates and costs for a range of variables vary widely in the literature, as do costs affected by exchange rates and pricing based on bulk purchasing,

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[183] An evaluation carried out in 2006 by the UK NHS considered family costs associated with non-inpatient screening and found that 'An average family cost for NHSP, when the screen had not been completed in the maternity unit, was £20.10, consisting of £9.58 in direct costs (travel, car parking, child minding arrangements, etc.) and £10.52 in lost parental wage costs.'

Uus, K., Bamford, J., Taylor, R. (2006). An analysis of the costs of implementing the National Newborn Hearing Screening Programme in England, *Journal of Medical Screening, 13*(1), 14-9.

[184] All AOAE screening requires the use of consumables.

- some factors and/or flow on effects are unknown and vary in different situations, and

- the analysis includes immediate and direct expenses only and does not attempt to deal with complex issues such as lost productivity, long term life impacts, costs associated with remoteness or cultural factors.

Standard programme costs

Total annual standard programme costs for 1,000 infants were calculated for each cost category, based on the parameters above.

Table F1 below summarises the totals for the range of costs included in the analysis for all regimes identified in the rapid literature review. Also included are costs for A-ABR regimes (two and three stage) where no consumables are required (aside from twice-yearly replacement of earphones).

Total screening costs for 1,000 infants were estimated and broken down to several key categories – as outlined above. The final two columns total the cost of screening (including setup, maintenance, family and direct screening costs) and the cost of screening and audiology combined.

Table F1. Summary costs for all screening regimes.

Programme	Total setup costs	Total maintenance costs	Family costs	Screening costs summary (excluding data management)	Audiology costs summary	Cost per 1,000 screens excluding audiology	Cost per 1,000 screens including audiology
AOAE/AOAE	$3,515	$1,603	$4,920	$44,994	$20,029	$50,111	$70,140
AOAE, A-ABR	$9,030	$3,305	$4,100	$45,209	$6,896	$57,544	$64,440
AOAE/A-ABR	$9,030	$3,305	$4,920	$46,653	$2,956	$58,988	$61,945
AOAE, A-ABR/A-ABR	$9,030	$3,305	$4,305	$47,240	$2,792	$59,575	$62,367
A-ABR/A-ABR	$5,915	$1,703	$4,387	$53,885	$1,971	$61,503	$63,474
AOAE/AOAE/ A-ABR	$9,030	$3,305	$5,412	$53,364	$2,792	$65,699	$68,491
A-ABR/A-ABR/ A-ABR	$5,915	$1,703	$4,426	$55,372	$1,151	$62,990	$64,141
A-ABR/A-ABR (no consumables)	$5,915	$1,703	$4,387	$38,178	$1,971	$45,795	$47,767
A-ABR/A-ABR/A-ABR (no consumables)	$5,915	$1,703	$4,426	$39,526	$1,151	$47,143	$48,294

AOAE/AOAE programs are often cited as being the most cost effective and, for screening costs alone, this was the case. When diagnostic costs are included, however, the cost advantage disappears and AOAE/AOAE programs were the most expensive – due to the higher refer rates.

Standard programme costs AOAE, A-ABR/A-ABR and A-ABR/A-ABR

Figure F1 presents a comparison of annual costs of three screening regime options, including AOAE, A-ABR/A-ABR (the regime closest to the current UNHSEIP regime), and the recommended regime, A-ABR/A-ABR. This is costed with consumables and without. The first five sets of columns present a comparison of each of the five cost categories across the three regimes. The final two sets of columns combine these five categories, with the first of these two sets representing screening costs alone and the second representing screening costs combined with audiology costs.

Figure F1. Comparison of screening and audiology costs (NZ$) per 1,000 infants screened with AOAE, A-ABR/A-ABR and A-ABR/A-ABR with and without consumables.
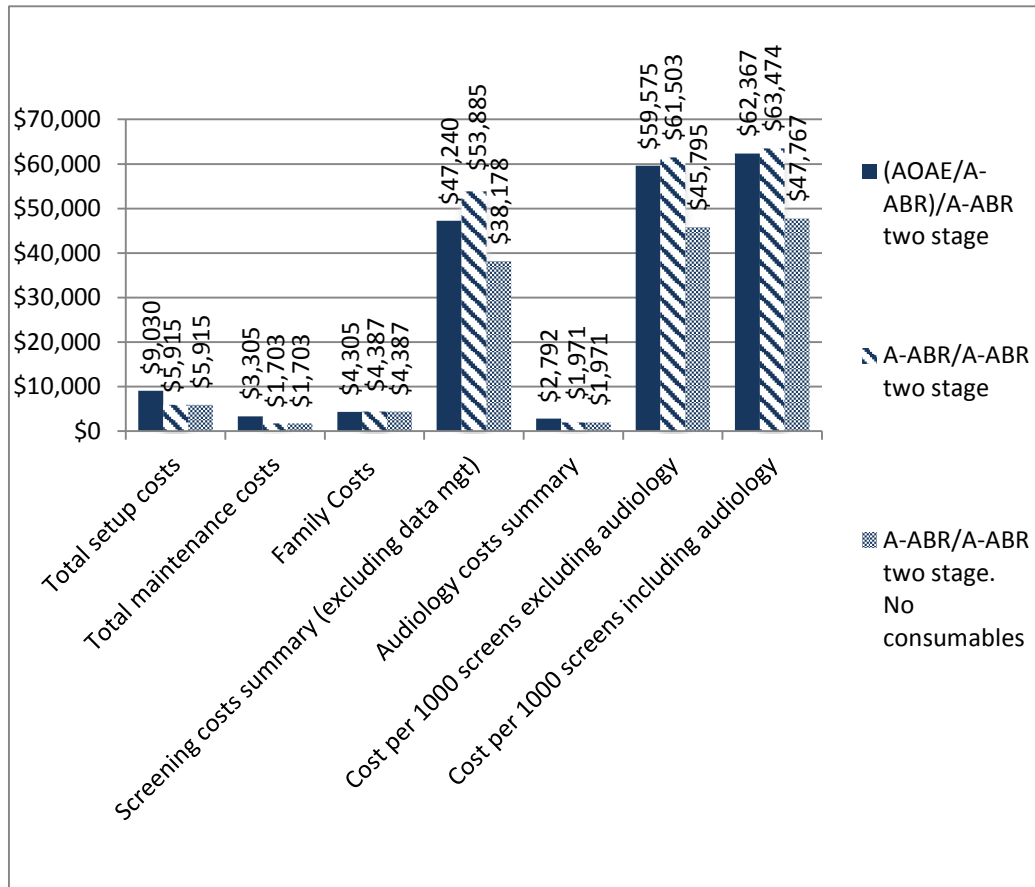
When comparing the three regimes, costs are lowest for the A-ABR/A-ABR regime using no consumables. This is the case for both screening alone ($38,178) as well as when screening and audiology costs are combined ($47,467). There are minimal differences between the overall costs for the AOAE, A-ABR/A-ABR regime ($62,367) and the A-ABR/A-ABR regime using consumables ($63,474).

Costs for an AOAE, A-ABR/A-ABR regime could not be significantly reduced through a change to an A-ABR device which did not require consumables, as a relatively small number of infants are screened using A-ABR under this regime.

Key observations that can be made from these comparisons include:

* Setup and maintenance costs are higher for the AOAE, A-ABR/A-ABR regime because this regime requires purchase of either two devices or two modules within the one device, compared to the A-ABR/A-ABR regime which only requires a single device.

- Although the AOAE, A-ABR/A-ABR regime has a high first refer rate, AOAE, A-ABR/A-ABR screening costs are slightly lower than screenings costs for the A-ABR/A-ABR regime using consumables. This is largely due to the direct impact of the cost of A-ABR consumables[185].

- The slightly higher audiology costs for the AOAE, A-ABR/A-ABR regime are due to the higher refer rate to audiology[186].

Refer rates for A-ABR screening

The A-ABR refer rates used in the cost analysis are estimates based on a combination of refer rates reported in the literature and refer rates from the programmes interviewed. As noted in the limitations section of the cost analysis, refer rates vary widely. An increase in a first or second refer rate will incur additional screening, co-ordination and family expenses, and for second refers, additional diagnostic expenses will be incurred. Some studies have found that A-ABR screening using a Chirp stimulus, has elevated second refer rates[187, 188]. One research team found a second refer rate of 2.7% for the MAICO MB 11 BERAphone compared to the 1.6% for the ALGO Portable[189].

Given that this analysis has identified a cost advantage for screening equipment which does not use consumables, a further analysis was done to examine the effects of higher refer rates, and whether this cost advantage remains in the presence of higher refer rates. For an A-ABR programme which uses consumables, for every 1% increase in first refer rate, an additional cost of $369

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[185] This analysis has used averaged standard costs for A-ABR consumables – without the benefit of bulk pricing. Competitive tendering and purchasing arrangements, however, could significantly lower these costs.

[186] A refer rate of 1.52% from the AOAE, A-ABR/A-ABR regime to audiology was used. This figure is based on the average refer rate from three screen programmes interviewed. The refer rate for the UNHSEIP is 1.7%.

(Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012.)

A refer rate of 0.95% from an A-ABR/A-ABR programmes was used. This figure is based on an average of the two screen A-ABR/A-ABR programmes interviewed.

[187] Cebulla, M., Shehata-Dieler, W. (2012). ABR-based newborn hearing screening with MB 11 BERAphone® using an optimized chirp for acoustical stimulation. *International Journal of Otorhinolayngology.* 76(4), 536-43. doi: 10.1016/j.ijporl.2012.01.012

[188] van den Berg et al. (2010). MB 11 BERAphone hearing screening compared to ALGO portable in a Dutch NICU: a pilot study.

[189] Cebulla, Shehata-Dieler. (2012). ABR-based newborn hearing screening with MB 11 BERAphone® using an optimized chirp for acoustical stimulation.

per 1,000 infants would be incurred. In contrast, for an A-ABR programme which does not use consumables, for every 1% increase in first refer rate, an additional cost of $223 per 1,000 infants would be incurred. For either type of A-ABR programme, where the second refer rate is increased by 1%, an additional cost of $1,915 per 1,000 infants would be incurred.

As a theoretical example, where both first and second refer rates are elevated – for example, 10% for the first refer rate and 4% for the second refer rate[190] – a cost advantage remains for A-ABR devices which do not use consumables. A likely or actual cost advantage for equipment which does not use disposable ear couplers has also been suggested by others[191, 192, 193].

Inpatient screening contrasted to community screening

All programmes require some outpatient screening and the standard costs in this analysis assume 10% of screening occurs post-discharge. However, UNHSEIP community screening rates in some DHBs are much higher than this. In some DHBs the majority of infants are screened in the community.

The additional cost to screen an infant in a dedicated community screening clinic, rather than as an inpatient is approximately $20 per infant. This additional cost includes screener travel and setup time, screener mileage, time associated with client non-attendance and rescheduling based on a 20% non-attendance rate.

The Australian Government, Medical Services Advisory Committee report on universal neonatal hearing screening concluded that community screening is effective when 'piggy-backed' with other health checks and immunisation. This

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[190] Australian Institute for Health and Welfare. (2013). National performance indicators for neonatal hearing screening in Australia.

[191] White et al. (2008). A comparative study of the MB 11 BERAphone and ABAER automated auditory brainstem response newborn hearing screening equipment.

[192] van den Berg et al. (2010) MB 11 BERAphone hearing screening compared to ALGO portable in a Dutch NICU: a pilot study.

[193] Cebulla, Shehata-Dieler. (2012). ABR-based newborn hearing screening with MB 11 BERAphone® using an optimized chirp for acoustical stimulation.

report also noted that losses to follow-up are more likely when long delays occur between rescreening or screening in the community after early discharge[194].

It is important to note that reducing community screening and increasing inpatient screening would not result in programme savings. For such a change to be achieved successfully, the additional cost required to undertake community screening would need to be redirected to the inpatient context to establish mechanisms for achieving optimal inpatient coverage and meeting programme benchmarks and outcomes.

Capture rates

While, for this exercise, a 100% capture rate has been assumed, all programmes report some infants as lost to follow-up.

The average capture rate from the international programmes reviewed was 98.0% (with a range of 96.5% to 99.5%). The reported capture rate for the UNHSEIP is 83.0%[195].

If an annual birth rate of 60,000 is assumed, a capture rate of 83.0% would mean that 10,200 infants would not be screened. Further, if it is assumed that one child per thousand would normally be diagnosed with the target condition (permanent bilateral hearing loss of moderate or greater degree) through the screening programme, then approximately 10 infants with the target condition might not be detected early, and a further 20 infants' mild or unilateral hearing losses could also remain undetected at birth.

Although some direct costs associated with the non-detection of auditory neuropathy have been estimated for comparisons between AOAE and A-ABR regimes, the cost of low capture rates and subsequent late detection of permanent hearing loss have not been estimated in this exercise. Unlike auditory neuropathy which is not detected by AOAE programmes, low capture rates are not necessarily linked with any particular type of regime. Also as noted above, the costing of late detection involves complex issues such as lost productivity, and long term life impacts which are beyond the scope of this exercise.

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[194] Australian Government, Department of Health and Ageing. (2008). Universal neonatal hearing screening: November 2007, Medical Services Advisory Committee

[195] Ministry of Health. 2013. UNHSEIP monitoring report on newborn hearing screening service provision, April 2012 – December 2012.

Case detection

A 2006 evaluation of universal newborn hearing screening in the UK included a 2003 cost effectiveness analysis of universal newborn hearing screening, compared to the infant distraction test screening. This analysis included audiology costs. A mean cost per 1,000 infants for a number of universal newborn hearing screening programmes in the UK was reported as being £37,383, with a range of £27,992 to £61,087. It also reported a mean cost per case identified of £34,826, with a range of £15,835 to £88,680[196].

For the purposes of this cost analysis, one case of the target condition (permanent bilateral moderate or greater hearing loss in the better ear) per 1,000 infants is estimated. Costs of detection for the three regimes are: AOAE, A-ABR/A-ABR, $62,367; A-ABR/A-ABR, $63,474; A-ABR/A-ABR – no consumables, $47,767.

Assumptions relating to each of the cost items are detailed in the following tables.

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[196] Uus, K., Bamford, J., Taylor, R. (2006). An analysis of the costs of implementing the National Newborn Hearing Screening Programme in England, *Journal of Medical Screening, 13*(1), 14-9.

Setup and maintenance cost assumptions

Table F2. Setup equipment and maintenance costs.

Cost category	Detail	Measure	Comments
Setup costs	Setup office	NZ$4,000	Standard items, e.g. desk, computer etc.
	Initial training	NZ$11,150	per 1,000 infants
Equipment costs	AOAE equipment	NZ$10,000	Estimate based on costs of several pieces of equipment
	A-ABR equipment	NZ$22,000	Estimate based on costs of several pieces of equipment
	Equipment lifespan	5 years	Advice from UNHSEIP – conservative estimate – other programme report longer lifespans.
Equipment maintenance costs	Supplier maintenance – AOAE	NZ$500	Estimate
	Supplier maintenance – A-ABR	NZ$600	Estimate
	Weekly maintenance	0.25 hours	Estimate based on programme experience
	Monthly maintenance	0.5 hours	Estimate based on programme experience
	Annual maintenance	0.5 hours	Estimate based on programme experience
	Troubleshooting	1 hour/month	Estimate based on programme experience

Family cost assumptions

The scope of this review did not allow a comprehensive assessment of costs to families of community screening, therefore the two cost assumptions included are likely to be a very conservative of the cost impact on families. Lost productivity costs, for example, are not included. An evaluation by the NHS in the UK considered family costs associated with non-inpatient screening and reached the following conclusion, 'An average family cost for NHSP, when the screen had not been completed in the maternity unit, was £20.10, consisting of £9.58 in direct costs (travel, car parking, child minding arrangements, etc.) and £10.52 in lost parental wage costs.'[197]

Assumptions from other sections are also used to calculate family costs. Refer rates for various regimes are used due to the variation in first and second refer rates across regimes. Also the percentage/s of infants screened in a dedicated community clinic are used to estimate family costs.

Table F3. Family costs.

Cost category	Measure	Comments
Family travel costs	20kms	Estimate
Family ancillary costs, e.g. babysitting	$25	Estimate

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[197] Uus, K., Bamford, J., Taylor, R. (2006). An analysis of the costs of implementing the National Newborn Hearing Screening Programme in England, *Journal of Medical Screening, 13*(1), 14-9.

Screening cost assumptions

Table F4. Screening costs.

Cost category	Measure	Comments
Screener cost – lower range	$32,000/annum	Advice from NSU
Screener cost – upper range	$53,000/annum	Advice from NSU
Screener cost – average	$42,500/annum	
Screener cost – average/hour including on costs	$30/hour	
Screener on costs	30%	Estimate
Time/screen – AOAE	0.33 hour	Estimate
Time additional AOAE first refer one step	0.10 hour	Estimate
Time additional A-ABR first refer one step	0.25 hour	Estimate
Time/screen – A-ABR click	0.50 hour	Estimate based on experience with newborn hearing screening
Time first refer reschedule	0.10 hour	Estimate based on experience with newborn hearing screening
First refer rate – AOAE	20%	Estimate based on the range of published first refer rates and data reported through the review of international programmes.
First refer rate – A-ABR	7%	As above
Second refer rate – AOAE	12%	As above
Second refer rate – A-ABR	0.95%	As above

Cost category	Measure	Comments
Second refer rate A-ABR – one stage	5%	Based on advice from two DHBs in NZ
Second refer follow up	0.33 hour	Estimate based on programme experience
Final refer rate (AOAE/AOAE/A-ABR)	1.52%	Estimate based on average referral rates reported by programmes meeting this criteria (international Programme 6 and Programme 7 and the UNHSEIP)
Third refer rate (A-ABR/A-ABR/A-ABR)	0.5%	Estimate based on diagnostic outcomes from ABR testing following referral from two stage A-ABR where approximately half of the babies referred to audiology have a hearing loss (including conductive losses)
Local coordinator/screen	0.02 hour	Estimate based on experience with newborn hearing screening
Local coordinator/first refer	0.08 hour	Estimate based on experience with newborn hearing screening
Local coordinator/second refer	0.50 hour	Estimate based on experience with newborn hearing screening
Local coordinator including on costs	$35/hour	Estimate
Inpatient screening rate	90%	Estimate based on reports from international hospital-based programmes interviewed

Cost category	Measure	Comments
Dedicated community clinic screening rate – low	10%	Estimate based on reports from international hospital-based programs interviewed
Travel – mileage	$0.74	Based on 2014 Australian Tax Office mileage rate
Travel – kilometres/clinic	200km	Based on advice from two DHBs
Travel – transport/clinic	$148	Based on above two assumptions
Travel + setup time/clinic	1.50 hours	Assuming 30 minutes travel each way and 15 minutes setup and pack up
Community clinic – babies/clinic	10 babies	Estimate based on advice from two DHBs
Community clinic – lost to follow-up rate, reschedule	20%	Estimate based on advice from two DHBs
Did Not Attend rate/1,000 births	15%	Based on UNHSEIP Monitoring Report
Did Not Attend follow-up time tracking/baby	0.20 hours	Estimate based on experience with newborn hearing screening
Labour – DHBs time	5 hours/month	Estimate based on experience with newborn hearing screening
Estimate DHB costs	$35	Estimate
Labour – corporate	2.0 FTE	Estimate
Labour – corporate average hourly rate including on costs	$50	Estimate
Labour – corporate costs/1,000 infants	$3,310	Estimate

Cost category	Measure	Comments
Labour – estimated national data input, 1.0 FTE including on costs	$54,925	Estimate
Labour – estimated data analysis, 0.4 FTE including on costs	$43,062	Estimate
Training – facilitator costs	16 hours/year	Estimate based on experience with newborn hearing screening
Training – screener update	8 hours/year	Estimate based on experience with newborn hearing screening
Training – screener competency	2 hours/year	Estimate based on experience with newborn hearing screening
Training – coordinator competency	2 hours/year	Estimate based on experience with newborn hearing screening
Auditory neuropathy detection rate	0.06%	Published evidence[198]
Additional costs incurred due to non-detection of ANSD in the previous year.	$7,900/child	Conservative estimate based on costs of additional health care visits and early intervention required per year for a child with a PCHI detected after 12 months of age

[198] Kirkim, G., Serbetcioglu, B., Erdag, T.K. Ceryan, K. (2008). The frequency of auditory neuropathy detected by universal newborn hearing screening program. *International Journal of Pediatric Otorhinolaryngology, 72*(10), 1461-9. doi: 10.1016/j.ijporl.2008.06.010

Audiology cost assumptions

Table F5. Audiology costs.

Cost category	Measures	Comments
Audiology – number of appointments/false positive	2	Estimate based on experience with newborn hearing screening
Audiology – time/appointment	2 hours	Advice from NZ
Audiology – rate of false positives: AOAE	11.8%	Estimate based on 1) detection of permanent HL at diagnosis and 2) a range of reported referral to audiology rates from AOAE programs
Audiology – rate of false positives: A-ABR	0.8%	Estimate based on 1) detection of permanent HL at diagnosis and 2) a range of reported referral to audiology rates from A-ABR programs
Audiology – rate of false positives: A-ABR one step	3.8%	Estimate based on 1) detection of permanent HL at diagnosis and 2) advice re referral rates from the AOAE/A-ABR stage of the AOAE, A-ABR/A-ABR regime
Audiology – rate of false positives: A-ABR three step	0.3%	Estimate based on 1) detection of permanent hearing loss at diagnosis and 2) estimate of referral rate to audiology from three stage A-ABR regime
Audiology – rate false positives AOAE/A-ABR	1.4%	Estimate based on 1) detection of permanent hearing loss at diagnosis and 2) estimate of referral rate to audiology from two stage AOAE/A-ABR
Audiology rate false positives – where A-ABR is the last of three screens	1.3%	Estimate based on 1) detection of permanent HL at diagnosis and 2) average referral rates where A-ABR is the last of three screens.
Audiology – cost including on costs	$76/hour	Estimate

Cost category	Measures	Comments
Audiology – number of appointments/diagnosis of target condition	4	Estimate based on experience with newborn hearing screening
Audiology – rate of target condition	0.1%	Estimates based on reported rates in literature and review of international programmes
Audiology – rate of other hearing loss	0.1%	Estimate based on reported rates in literature and review of international programmes
Audiology – data entry: false positive	16 minutes	Estimate based on experience with newborn hearing screening
Audiology – data entry: diagnosis of target condition	33 minutes	Estimate based on experience with newborn hearing screening

Data management assumptions

Table F6. Data management costs.

Cost category	Measures	Comments
Data manager: cost	$30/hour	Estimate based on experience with newborn hearing screening
Data – demographics: manual upload partial	0.02 hours/baby	Estimate based on experience with newborn hearing screening
Data – demographics: manual upload full	0.03 hours/baby	Estimate based on experience with newborn hearing screening
Data – demographics: auto upload	0.08 hours/upload	Estimate based on experience with newborn hearing screening
Data – screen: manual upload: child	0.03 hours/baby	Estimate based on experience with newborn hearing screening
Data – screen: auto upload	0.1 hours/day	Estimate based on experience with newborn hearing screening
Data management – first refer: database	0.05 hours/1st refer	Estimate based on experience with newborn hearing screening
Data management – second refer: database	0.03 hours/2nd refer	Estimate based on experience with newborn hearing screening

Cost category	Measures	Comments
Data management – first refer: manual	0.08 hours/1st refer	Estimate based on experience with newborn hearing screening
Data management – second refer: manual	0.16 hours/2nd refer	Estimate based on experience with newborn hearing screening
Assumed number of births/upload	30/upload	Estimate