Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.

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ABSTRACT

Children with Down syndrome are at an increased risk of developing conductive hearing loss as a result of chronic/recurring otitis media with effusion (Barr, Dungworth, Hunter, McFarlane, & Kubba, 2011; Bernardi, Pires, Oliveira, & Nisihara, 2017) which may exacerbate their difficulties in speech and language development (Tharpe, 2016). Additionally, children with Down syndrome who experienced temporary or fluctuating hearing loss between 2-4 years of age have been found to demonstrate delayed speech and language development beyond the time of resolution of the hearing loss (Laws & Hall, 2014). The aim of this study was to evaluate the utility of using the Ling 7-Sound Test with four parents to monitor the hearing of their young children with Down syndrome on a daily basis over the period of one school term. Children's responses to the Ling 7-Sound Test were compared to objective results obtained from tympanometry, performed weekly by the researcher at the early intervention centre the children attend. If the child was found to have otitis media with effusion, as evidenced by a type B low tympanogram, parents were encouraged to employ environmental modifications and communication strategies to mitigate the negative impacts of otitis media with effusion. Pure tone audiometry was used to confirm the hearing status when a child's tympanometry results changed. The Parents' Evaluation of Aural/Oral Performance of Children (PEACH) and Teachers' Evaluation of Aural/Oral Performance of Children (TEACH) questionnaires (Ching & Hill, 2005) were completed by parents and early intervention educators to evaluate the child's listening in the home and preschool environments. Following the study, parents completed a social validity questionnaire and participated in a semi-structured interview. All four families successfully incorporated the Ling 7-Sound Test into their daily routines and reported value in doing so. One child presented with binaural type B low tympanograms and a mild to moderate hearing loss, consistent with middle ear fluid during two weeks of the study. Three of the children

completed the Ling 7-Sound Test with 45 dB A stimuli, while the fourth completed it with 40 dB A stimuli due to a different sensitivity in hearing. The qualitative nature of the comments provided by parents through the PEACH, Ling 7-Sound Test, questionnaires and interviews increased the holistic understanding of each child’s health, attention and listening behaviour in relation to their ear health and hearing during the 10 week study period.

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CHAPTER 1 LITERATURE REVIEW

1.1 Introduction

1.1.1 Down syndrome

Down syndrome was first described by John Langdon Down in 1867 (Down, 1867). It is a genetic disorder affecting approximately 1 in 1000 births (Cocchi et al., 2010; de Graaf, Buckley, & Skotko, 2015; Morris, 2009), making it the most common genetic syndrome (Hassold & Jacobs, 1984). Down syndrome is usually caused by a third copy of all or part of chromosome 21, a discovery originally identified by Lejeune, Gautier, and Turpin (1959) . Down syndrome is characterised by distinct facial features along with physical growth, cognitive, and language differences (Fidler, 2005). There is wide variability in phenotypical presentation among people with Down syndrome; as such the severity of the challenges experienced by each person with Down syndrome falls anywhere on a spectrum from mild to profound. Despite this variability, children with Down syndrome share a distinctive profile of strengths and challenges, which are important considerations when working with this population. Children with Down syndrome experience challenges related to cognition, auditory processing, motor planning and execution, and language (Abbeduto, Warren, & Conners, 2007; Cardoso, de Campos, dos Santos, Santos, & Rocha, 2015; Nadel, 2003; E. Smith, 2015). Relative strengths are evident in aspects of visuospatial processing and social interaction (Feeley, Jones, Blackburn, & Bauer, 2011; Fidler, Hepburn, & Rogers, 2006).

1.1.2 The speech and language profile of children with Down syndrome

Children with Down syndrome present with speech and language abilities and challenges that vary greatly between individuals (Roberts, Price, & Malkin, 2007). The phenotype includes delays in language acquisition, poor speech intelligibility, and challenges with motor planning and execution, with relative strengths in affect sharing and their ability to form relationships (Fidler, 2005; Fidler et al., 2006). Also impacting on their ability to develop speech and language, children with Down syndrome experience challenges in sensorimotor (Fidler, Hepburn, Mankin, & Rogers, 2005) and cognitive development (Bull, 2011). Children with Down syndrome are more likely than their typically developing peers to have hearing and vision loss (Shott, Joseph, & Heithaus, 2001; Stephen, Dickson, Kindley, Scott, & Charleton, 2007) and delays in their development of both gross and fine motor skills (Fidler et al., 2005). Hypotonia, oral structural differences and reduced speed and range of movement may contribute to dysarthric or apraxic speech imprecision in people with Down syndrome (Kent & Vorperian, 2013; Rupela, Velleman, & Andrianopoulos, 2016). As well as motor speech differences affecting articulation, children with Down syndrome have presented with differences in their morphosyntactic abilities (i.e. structure within words and sentences) (Andreou & Katsarou, 2013), that further impact their ability to engage in conversation effectively with others. Despite these challenges, children with Down syndrome are described as having relative strengths in visual processing (Feeley et al., 2011) and fine motor movements of their hands, making the use of gesture and sign an effective tool to increase the communication abilities of the child (Abbeduto et al., 2007). People with Down syndrome also present with cognitive impairment ranging from mild to severe (Bull, 2011). A delay in cognitive skills such as ideation, reasoning, memory and understanding the world may result in language delay, which may result in further cognitive delay because of the important role language plays in knowledge acquisition (Perlovsky & Sakai, 2014). The above challenges that face those with Down syndrome to a greater or lesser extent may all impact on pragmatic language development. Pragmatic competence refers to the ability of a person to “use language effectively and appropriately in interaction with other people” (O'Neill, 2014, p. 363). Pragmatic language skills include requesting, commenting, asking questions, using humour, adapting communication to different people and producing narratives. This profile creates specific considerations for the professionals who support those with Down syndrome to reach their full potential. Hearing loss may further delay the speech and language development of children with Down syndrome.

1.1.3 Paediatric hearing loss

Infants and children may experience hearing loss as a result of factors occurring before, during or after birth. The nature of such losses may be conductive, sensory or neural and may range in severity from mild to profound. A conductive hearing loss results from attenuation of sound between the outer ear and inner ear or cochlea. Conductive hearing loss can be a result of differences in anatomy such as aural atresia, or middle ear anomalies acquired prenatally (Declau, Cremers, & Heyning, 1999). Other postnatally acquired disorders may also result in a temporary or fluctuating conductive hearing loss such as impacted cerumen (Roeser & Ballachanda, 1997) and otitis media (Shekelle et al., 2002). Otitis media has been reported to be the most common cause of hearing loss in children worldwide (Graham, Delap, & Goldsmith, 2002). Otitis media is a general term used to describe inflammation of the tympanic membrane and middle ear. It is further categorised according to the presence or absence of effusion in the middle ear cavity, effusion type and duration of the inflammation (Stach & Ramachandran, 2014). Most frequently, otitis media is caused by the accumulation of fluid behind the tympanic membrane; a result of the Eustachian tube being blocked or not effectively draining fluid (Castillo & Rolland, 2007). This is more common in children than adults as their Eustachian tubes are more horizontal and flaccid, becoming increasingly vertical with age. It is also common in those with craniofacial differences such as Down syndrome (Northern & Downs, 2002).

There are three types of otitis media: acute otitis media, otitis media with effusion and chronic otitis media. Acute otitis media is an infection of the middle ear, usually with a rapid onset and accompanied by symptoms including middle ear fluid, bulging tympanic membrane, and otalgia (Gribben, Salkeld, Hoare, & Jones, 2012). Children may experience earache and/or a raised temperature and the infection is commonly treated with antibiotics (Marriage, Brown, & Austin, 2017).

Otitis media with effusion is characterised by inflammation, the presence of fluid behind the eardrum and an absence of infection (Gribben et al., 2012). In most cases, Otitis media with effusion will resolve without intervention within a number of weeks. It is often symptomless, and as such, parents and children may not be aware that a conductive hearing loss is present (Marriage et al., 2017). Cole and Flexer (2015) report that approximately 50% of episodes of otitis media with effusion are not detected by parents as the child does not appear unwell and the child is not developmentally able to communicate to their parents that they are not hearing well. Although the presence of otitis media with effusion will not necessarily result in a hearing loss, if hearing loss is present, it may range in severity from slight to moderate (P. Thompson, 2008). The severity of hearing loss is related to the volume of fluid in the middle ear space, which causes a reduction in admittance of sound through the tympanic membrane and an increase in mass of the middle ear system (Gravel, 2003; Ravicz, 2004). Experiencing otitis media with effusion can be unpleasant for children as the feeling of aural fullness may be uncomfortable and the associated hearing loss increases the effort involved in everyday listening. If persistent and untreated, otitis media with effusion may result in tympanic membrane perforation, scarring (tympanosclerosis) or abnormal skin growth in the middle ear space (cholesteotoma) (Slattery & House, 1998; J. Smith & Danner, 2006).

Chronic otitis media indicates infection lasting longer than three months with or without perforation of the eardrum (Cole & Flexer, 2015).

Sensorineural hearing loss is usually permanent, resulting from the cochlear mechanism failing to varying degrees to convert and deliver the mechanical signals arriving from the middle ear to neural impulses in the vestibulocochlear nerve (Stach & Ramachandran, 2014). As with conductive hearing loss, sensorineural hearing loss may be caused by factors occurring prenatally or postnatally, although over half of childhood sensorineural hearing loss is attributable to genetic conditions (Brown, Chau, Atashband, Westerberg, & Kozak, 2009). There may be both structural and teratogenic causes of sensorineural hearing loss as a result of maternal infection during pregnancy. The resultant hearing loss may be due to differences in inner ear anatomy (Reilly, 1998), cytomegalovirus (Fowler & Boppana, 2006), congenital syphilis (Pletcher & Cheung, 2003), maternal rubella (Niedzielska, Kątska, & Szymula, 2000) toxoplasmosis (Brown et al., 2009), measles (Morzaria, Westerberg, & Kozak, 2004), mumps and TORCH infections (toxoplasmosis, others, rubella, cytomegalovirus, and herpes simplex viruses) (R. Smith, Bale Jr, & White, 2005). Postnatal causes of sensorineural hearing loss include persistent pulmonary hypertension as a newborn (Lasky, Wiorek, & Becker, 1998), meningitis (Kutz, Simon, Chennupati, Giannoni, & Manolidis, 2006), autoimmune inner ear disease (Bovo, Aimoni, & Martini, 2006), viral infections (Kawashima et al., 2005; Lasisi, Ayodele, & Ijaduola, 2006), exposure to ototoxic drugs (Montaguti et al., 2002) and prolonged neonatal jaundice (R. Smith et al., 2005). Neural hearing disorders can be categorised into one of three types depending on the affected structure or function within the auditory system. Firstly, retrocochlear hearing loss results from a structural lesion in the nervous system beyond the cochlea and may be characterised by sensorineural, unilateral hearing loss, poor speech perception inconsistent with hearing level and vertigo (Stach & Ramachandran, 2014). Retrocochlear hearing loss can be difficult to discriminate from cochlear hearing loss with audiology results alone as the above characterisations could be that of retrocochlear or sensorineural hearing loss. Therefore, the site of lesion probably responsible for retrocochlear hearing loss is usually confirmed using magnetic resonance imaging (Gal, Rottenberg, Pazourkova, Vanicek, & Vogazianos, 2018). Secondly, lesions more medial to those resulting in retrocochlear hearing loss may cause auditory processing disorders (APD). APD is defined as “a deficit in the way the neural representation of sounds are processed by the brain, resulting in a distorted neural representation of the auditory signal within the auditory nervous system. APD creates difficulty in listening (i.e. hearing with intent to extract information)" (Dillon & Cameron, 2015). Lastly, auditory neuropathy spectrum disorder (ANSD) is reported to be caused by a presynaptic lesion to the inner hair cells, a postsynaptic lesion to the auditory nerve or auditory ganglion cells, or a brainstem lesion (Rance & Starr, 2015). ANSD is characterised by typical cochlear amplification by the outer hair cells, but disordered neural conduction. Regardless of the type of hearing loss a child experiences, a proactive approach in diagnosis and management is advised because undetected hearing loss affects language development (Moeller & Tomblin, 2015; Nicholas & Geers, 2006). All children require precise linguistic input, made possible by audibility of the entire speech spectrum, to accurately perceive spoken language (Gravel, 2003). Unlike adults, for whom hearing loss may present against a backdrop of a mature language system, children are in the process of developing language. Furthermore, for children with hearing loss who also have a developmental disability such as Down syndrome, the consequential delay in language development may be exacerbated.

1.1.4 Hearing thresholds

Normal hearing thresholds are classified as 15 decibels hearing level (dB HL) or better (Goodman, 1965). The following table (Table 1) displays the classification of degree of hearing loss generally accepted internationally and used in the New Zealand professional practice standards and best practice guidelines (New Zealand Audiological Society, 2016).

Table 1. Degree of hearing loss and its associated pure tone audiometry air conduction thresholds at 500, 1000 and 2000 Hz.

Average Hearing Thresholds 500-2000 Hz (Goodman, 1965)	Degree of Hearing Loss
-10 to 15 dB HL	Normal hearing
16 to 25 dB HL	Slight hearing loss
26 to 40 dB HL	Mild hearing loss
41 to 55 dB HL	Moderate hearing loss
56 to 70 dB HL	Moderately-severe hearing loss
71 to 90 dB HL	Severe hearing loss
>91 dB HL	Profound hearing loss

1.1.5 Hearing loss in children with Down syndrome

The reported prevalence of hearing loss among preschool age children with Down syndrome ranges from 38%-78% (Tharpe & Seewald, 2016) and is conductive in nature for up to 86% of that population (Raut et al., 2011). Kreicher, Weir, Nguyen, and Meyer (2018) evaluated the hearing of 1088 children up to the age of 21 with Down syndrome retrospectively and found 75.4% of the children had experienced two or more episodes of acute otitis media or chronic otitis media with effusion. Otitis media is also highly prevalent in typically developing children, with 50-80% of all children reported to experience an episode before the age of four (Williamson, 2015). There are a number of contributing factors that lead to the increased incidence of conductive hearing loss in children with Down syndrome when compared to their typically developing peers. For example, children with Down syndrome have smaller craniofacial anatomy, making them more susceptible to mucous being trapped in the middle ear space (Ramia, Musharrafieh, Khaddage, & Sabri, 2014). Additionally, 40-50% of children with Down syndrome have stenotic ear canals, in which the narrowness of the canal walls predisposes them to impacted cerumen and/or foreign objects, which can contribute to a conductive loss, as well as causing difficulties observing the tympanic membrane otoscopically (Ramia et al., 2014; Shott, 2006). Children with Down syndrome also experience muscle hypotonia, which can extend to the tensor veli palatini muscle responsible for the opening of the Eustachian tubes (Shott, 2006). Eustachian tubes connect the middle ear to the back of the throat. Normally, they remain closed at rest, opening under neural control during certain activities such as swallowing and yawning. Failure of the Eustachian tubes to open, as a sequela of hypomobility, can result in fluid becoming trapped in the middle ear space, and can result in a conductive hearing loss (Shott, 2006). Furthermore, children with Down syndrome experience an increased incidence of respiratory tract infections, which are a frequent precursor to otitis media with effusion (Nightengale, Yoon, Wolter-Warmerdam, Daniels, & Hickey, 2017; Ramia et al., 2014). Conductive hearing loss in children with Down syndrome is often recurrent and persistent due to the aforementioned predispositions. Additionally, children with Down syndrome are likely to experience their first episode of otitis media at an earlier age than their typically developing peers; similarly, their last episode often occurs at a later age than typically developing children (Tharpe & Seewald, 2016).

Children with Down syndrome may also present with sensorineural or neural hearing loss or a combination of the two (mixed), though these types are not as common as conductive hearing loss and usually occur later in life (Porter & Tharpe, 2010). Park, Wilson, Stevens, Harward, and Hohler (2012) identified 332 children with Down syndrome using the Utah Department of Health's Newborn Hearing Screening database and found 46% had hearing loss. Within this group of children with Down syndrome who had hearing loss 3.9% had a sensorineural type, 2.0% had a mixed type, 5.9% had an indeterminate type, and 88.2% had a conductive type.

1.1.6 Impact of conductive hearing loss for children with Down syndrome

Like their typically developing peers, children with Down syndrome who experience hearing loss are at increased risk of poor language outcomes (Austeng et al., 2013). Even a slight or mild hearing loss may impact speech development in children because audibility of even quiet sounds is important for learning the subtleties of language used in interaction with other people (Bess, 1985; Dobie & Berlin, 1979). For a child with Down syndrome, who is likely to already be experiencing a delay in their speech and language development, such losses can be especially detrimental. A child's hearing sensitivity must be 15 dB HL or better in both ears in order to hear the entire speech spectrum at an appropriate sensation level above threshold (Tharpe, 2016). A 30 dB HL hearing loss can result in a child missing 25-40% of a speech signal, if it is not managed appropriately, due to the speech signal arriving at the ear at a level below threshold (Cole & Flexer, 2015). Additionally, the effort required to hear will be greater, and can result in fatigue (Hicks & Tharpe, 2002; Rosenfeld, Goldsmith, & Tetlus, 1997).

Children with Down syndrome who experience temporary or fluctuating hearing loss between 2-4 years of age have been found to demonstrate delayed speech and language development beyond the time of resolution of the hearing loss (Laws & Hall, 2014). Delays in the perception and production of speech/phonology, morphosyntax and pragmatics may be exacerbated by hearing loss.

A fluctuating incoming acoustic signal results in a child receiving an inconsistent language model, which can create difficulties in organising the auditory input in a meaningful and useful way and can affect the development of auditory processing and short-term verbal memory skills (Partington & Galloway, 2005). Children with Down syndrome are reported to have difficulties in their short term memory for speech sound information (phonological memory) beyond what would be expected from their cognitive abilities (Faught, Conners, Barber, & Price, 2016; Silverman, 2007; Yoder & Warren, 2004). Hearing loss can exacerbate poor phonological memory which may further contribute to difficulty in language learning (Jarrold, Thorn, & Stephens, 2009).

Morphosyntactic language development can also be affected by childhood hearing loss. For children with Down syndrome, morphosyntax is often more impaired than other areas of language (Zampini & D'Odorico, 2011). Delays in morphosyntax are characterised by infrequent use of function words, short utterance length, limited use of subordinate clauses and difficulty in combining words (Caselli, Monaco, Trasciani, & Vicari, 2008; Draghi & Zampini, 2018). Even minimal hearing loss may create difficulty in the comprehension of soft speech, speech spoken from a distance, speech in the presence of background noise, detection of subtle conversational cues and detecting phonetic markers such as the bound morpheme *–s* (Cole & Flexer, 2015). All of these effects may exacerbate delays in morphosyntactic language development.

Delays in the pragmatic use of language for communication may also be delayed by otitis media with effusion due to reduced audibility, limited exposure to different communication strategies and behaviours, and later development of theory of mind (Most, Shina-August, & Meilijson, 2010). Firstly, children with hearing loss experience decreased audibility (Laws & Hall, 2014). It may be that children with hearing loss have the pragmatic skills needed to respond appropriately, but do not use these skills suitably due to missing the acoustic cue from their conversation partner or they do not respond with language that is articulated precisely enough to be understood by others (Laws & Hall, 2014). Secondly, untreated hearing loss results in limited exposure to communication strategies and behaviors (Most et al., 2010). Children learn how to use language through observing others and learning from their modelling, then attempting to use these techniques in their own interactions. They get feedback both through hearing themselves and observing and listening to the interlocutor's response, a process referred to as the phonological loop (Laws & Hall, 2014). Such communication may not always be audible to children with hearing loss which may account for a delay in their pragmatic development (Most et al., 2010). Lastly, childhood hearing loss has been associated with delayed theory of mind development (Most et al., 2010). Theory of mind refers to a person's ability to see another person's perspective and is a skill that develops throughout life (Miller, 2012). It is suggested that language ability plays a significant role in theory of mind development in young children (Woolfe, Want, & Siegal, 2002). During infancy and early childhood, typically developing children learn to imitate others, recognize emotion, understand that different people like, want and think different things and engage in pretend play (de Villiers & de Villiers, 2014; Westby & Robinson, 2014). Development of theory of mind is indicated by the child's language, being able to adapt conversation to different people based on what they know, providing adequate background information and recognizing and repairing communication breakdowns. Reduced audibility, limited exposure to communication strategies and behaviours, and delayed theory of mind are all sequelae of hearing loss that may contribute to delayed communication development. Because a significant correlation between poor pragmatic language abilities and poor social outcomes has been documented (Goberis et al., 2012), pragmatics is not an aspect of language that should be overlooked, including amongst children with Down syndrome.

Although the possible impacts of conductive hearing loss on a child with Down syndrome are considerable, environmental factors that influence a child's participation may be protective such as being in a setting with low levels of background noise and the use of specific communication strategies (see section 1.3.2) (DeConde Johnson, 2000).

1.2 Diagnosis

Accurate and timely diagnosis of otitis media with effusion and conductive hearing loss is essential to mitigate the negative effects of hearing loss. In New Zealand, infants and young children receive hearing screening at birth and 4 years old. The purpose of the hearing screenings is to differentiate members of the population who will not experience communication difficulties from those who would benefit from diagnostic testing as there is a high probability they have hearing loss. Those who do not pass the hearing screening or present with concerns regarding their hearing are referred for a diagnostic hearing assessment. Diagnostic hearing assessments are performed by an audiologist and consist of an assessment battery that provides a comprehensive depiction of a person’s hearing. This information is then used to guide management decisions and refer to other professionals such as general practitioners and Ear, Nose and Throat specialists when indicated. Current methods of screening for and diagnosing hearing loss are discussed below.

1.2.1 Protocols

The purpose of hearing screening is to identify members of a population who may need additional services to mitigate any negative impacts that may result from hearing loss (Ministry of Health, 2016). Screening may be universal or targeted. Universal screening is screening applied to an entire population, such as newborn hearing screening in New Zealand, whereas targeted screening involves screening all members of a target population (Ministry of Health, 2016). Because of the increased risk of developing otitis media with effusion, young children with Down syndrome receive targeted screening to detect the presence or absence of hearing loss (see Figure 1).

In New Zealand, all parents of newborn babies are invited to participate in the Universal Newborn Hearing Screening and Early Intervention Programme (Ministry of Health, 2016). As part of this programme hearing is screened using automated auditory brainstem response (AABR) before the baby is one month old. For babies with Down syndrome, regardless of the result of this screening, their hearing is assessed using a diagnostic ABR by the time they are three months old. The result of this assessment then determines the course of action that will be followed. The following two paragraphs describe the screening pathway specifically for infants with Down syndrome (see Figure 1).

If a child is found to have normal hearing in the ABR, they will be reviewed by an audiologist and an Ear, Nose and Throat specialist to obtain distortion product otoacoustic emissions (DPOAE) and check for presence of otitis media with effusion. If the child is found to have present DPOAEs (indicative of typical outer hair cell function) or no otitis media with effusion is present, the child will be reviewed annually by an audiologist, and be referred to an Ear, Nose and Throat specialist as required. If DPOAEs are absent or otitis media with effusion is present, the child will be managed by an Ear, Nose and Throat specialist, and reviewed by an audiologist as appropriate.

If a child is found to have a conductive hearing loss greater than or equal to 45 dB estimated hearing level (eHL) in the ABR, absent DPOAEs or otitis media with effusion, they will be managed by an Ear, Nose and Throat specialist, and reviewed by an audiologist as appropriate. If a child has a significant, persistent conductive hearing loss, a discussion of the suitability of bone conduction hearing aids is completed with the family. Some infants with Down syndrome are not candidates for ventilation tubes or grommets due to the small size of their ear canals particularly when they are very young. If a child is diagnosed with a sensorineural hearing loss in the ABR, they are referred to an audiologist and an Ear, Nose and Throat specialist for management such as hearing aids or cochlear implants. This approach for children with Down syndrome is more intensive than that for typically developing children, who will not be seen by an audiologist unless they do not pass the newborn hearing screening, or are referred by their general practitioner if any concerns arise during early childhood. All children have their hearing screened by a nurse at approximately age 4, before they start school (Ministry of Health, 2016).

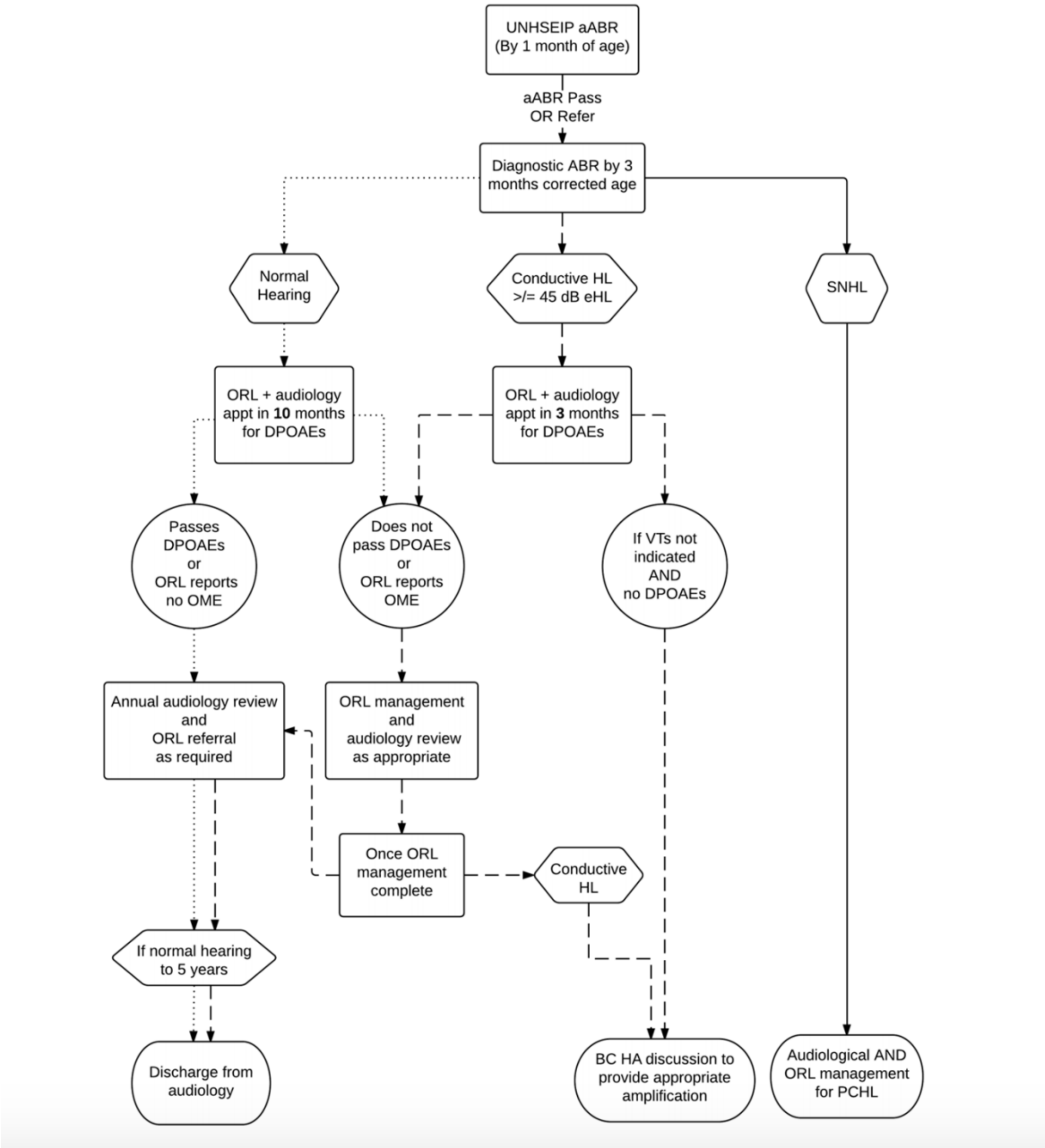

Figure 1. Audiological management of babies with Down syndrome (Ministry of Health, 2016). Copyright 2016 by the Ministry of Health. Reprinted with permission.

1.2.2 Screening

There are several screening methods available including objective and subjective methods. Some are more suitable than others for the detection of the mild, fluctuating, conductive hearing loss that children with Down syndrome are likely to experience as a sequela to otitis media with effusion. An example of a commonly used screening test is the use of tuning forks. Tuning forks of various frequencies are commonly used in medical settings to identify conductive and unilateral hearing loss. One example is the Rinne test (Rinne, 1855), in which a health professional strikes a tuning fork on a hard surface and holds the tines of fork near the ear canal, then on the patient's mastoid and asks the patient which placement was louder. If the patient reports the sound from the tuning fork is louder on the mastoid than when near the ear canal, a conductive hearing loss is indicated (Burkey, Lippy, Schuring, & Rizer, 1998). This is a quick and effective tool found to have high sensitivity (96.6%) in detecting conductive hearing loss. However, the patient must be able to express which sound is louder, which involves cognitive, receptive and expressive language skills that are likely above that of a young child with Down syndrome.

More modern screening methods include the use of tablets, smartphones and telehealth (Govender & Mars, 2017). Children are generally motivated to participate in activities involving technology, and several programmes have reportedly been accurate in their use in screening for hearing loss (Govender & Mars, 2017). However, most studies have examined technology use with typically developing children over 5 years of age, and such technology may not be appropriate for use with younger children with Down syndrome because of the developmental stage required to complete the tasks. Alternatively, parents may be able to provide audiologists with adequate information about their child's hearing based on their observations.

1.2.3 Parent report measures

Subjective assessments such as parent report measures have been shown to provide useful information about children's functional hearing in non-clinical environments (Allum, Greisiger, Straubhaar, & Carpenter, 2000; Ben-Itzhak, Greenstein, & Kishon-Rabin, 2014; Harrison & Seewald, 2000). Parent report measures are inexpensive and can pair with objective measures to provide a multidimensional depiction of a child's listening abilities. If children do not achieve satisfactory results in parent report measures, referral to an audiologist may be warranted for administration of objective measures. While measures such as play audiometry, VRA, DPOAEs and tympanometry provide valuable objective information regarding hearing, they give little insight into the child's listening abilities in everyday communication (Bagatto, Moodie, Seewald, Bartlett, & Scollie, 2011). Parents, on the other hand have the unique opportunity to observe their child's behaviour in a range of natural settings, across time and with different interlocutors.

It has been suggested that questionnaires may be an accurate and cost effective tool in screening children for hearing loss (Samelli, Rabelo, & Vespasiano, 2011).

The LittlEARS Auditory Questionnaire (Tsiakpini, Weichbold, & Kuehn-Inacker, 2004) is a parent report measure developed for young infants with cochlear implants, to measure auditory development. More recently it was validated for children who wear hearing aids (Bagatto, Moodie, Malandrino, et al., 2011). The LittlEARS consists of 35 yes/no questions and is designed to be administered by parents of children under the age of 24 months. This questionnaire is widely distributed by audiologists as it can typically be administered easily by parents. Normative data from 3309 children with normal hearing is available for comparison (Coninx et al., 2009). Although not designed to diagnose hearing loss, it may be sensitive to changes in hearing over time when administered repeatedly. For example, parents of children who use hearing aids or cochlear implants may fill the questionnaire out periodically and scores may decline if the child experiences otitis media with effusion, if the child's hearing loss is progressive in nature, or if the child is not progressing as expected in spite of parent input and early intervention.

Once a child has reached the ceiling score on the LittleEARS or the age of 24 months, the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) may be administered (Ching & Hill, 2005). The PEACH and Teachers' Evaluation of Aural/Oral Performance of

Children (TEACH) diaries were designed to be used by parents and teachers respectively to evaluate the effectiveness of a child's amplification (Ching & Hill, 2005). More recently, the PEACH and TEACH rating scales were developed due to the significant amount of time required to completed the diaries (Bagatto & Scollie, 2013). The rating scales differ from the diaries in both length and construct, because the diaries are much longer than the rating scales and require detailed written reflection from the person completing the diary. In comparison, the rating scale version consists of 13 questions that parents can complete in just a few minutes, using the five-point rating scale, and has been found to yield results consistent with those obtained from the longer PEACH and TEACH diaries.

Another option for parents observing their child's behaviour is to complete tasks such as the Early Listening Function and the Ling Sound Test. These tasks guide parent observations through simple listening activities completed with their child on a regular basis. The Early Listening Function (ELF) for infants and toddlers (Anderson, 2002a) consists of 12 listening activities to be performed by the parent and child in their home. The activities consist of stimuli that are quiet, normal and loud in volume. Parents complete the activities in a quiet environment as well as an environment with background noise and record whether their child responded when they were 6 inches, 3 feet, 6 feet, 10 feet and >15 feet away. The ELF is not used to detect or confirm hearing loss, rather, it is a tool used to aid parents in understanding the real life implications of the hearing loss on the child's speech perception. The activities aim to demonstrate the child's ability to detect sound at differing volumes, at differing distance, both in the presence of background noise and in quiet settings. Parents use the information gained from these activities to determine the support needed for their child to understand speech in different settings. For example, a child may need the environment to be quiet, and have the person speaking within one meter of them in order to hear well. The information gained by the parents in this assessment is known as the child's "listening bubble”, within which the child has the best access to communication (Anderson, 2002b). Observations by parents using the ELF have reportedly identified both progressive and fluctuating hearing loss as a result of otitis media with effusion (Anderson, 2002b). The ELFLING (Anderson, 2016) is an adaptation of the Early Listening Function which uses the Ling 6 Sounds as the stimuli.

The Ling Sound Test (Ling, 1976) is a tool recommended for routine use by parents to assess and monitor their child’s ability to hear across the speech spectrum when wearing hearing aids or cochlear implants.

Parents are encouraged to complete the Ling Sound Test with their child in a quiet place at home each day. Originally the Ling Sound Test consisted of five sounds called speech phonemes (/a/, /u/, /i/, /s/, and / ʃ/) that the parent would present verbally to the child. More recently, the sound /m/ was added to encompass the very low frequency components of speech (Ling, 1989). The Ling sounds, spoken by an American English speaker, encompass the frequency range of American speech sounds. However, when spoken in New Zealand or Australian English, the /u/ sound does not have low first and second formant frequencies (F1, F2) as it does in American and English English. Instead, F1 is low and F2 has a mid-frequency formant peak. Therefore, it has been suggested that, in Australia and New Zealand, the sound /ɔ/, which has a lower F2, is added or used in the place of the /u/ sound to ensure that the entire speech spectrum is assessed due to the difference in pronunciation (Agung, Purdy, & Kitamura, 2005).

Various methods of completing the Ling Sound Test have been reported (Agung et al., 2005; Baudhuin, Cadieux, Firszt, Reeder, & Maxson, 2012; Beltrame et al., 2013; Burgdorf, 2014; Glista, Scollie, Moodie, & Easwar, 2014; Kilcullen, 2014; Tolan et al., 2017). However, there is seldom documentation of the exact methodology of administering the test as an assessment or monitoring tool. There are many variables which would affect the administration and results of the assessment including distance of the presenter from the child, intensity of speech or recorded Ling sounds at the child's ear, the duration of each Ling sound, and the assessment environment. This variability results in difficulty both in replicating previous methodology accurately and comparing results across studies.

Despite the lack of clarity in the literature, there are several components that can be made consistent when completing the Ling Sound Test in the home. To begin with, a quiet place is selected. The parent stands 1 metre behind their child to ensure the child does not receive any visual cues. The parent then says the speech sounds at a normal conversational voice level; for most adults this volume is between 50-65 dB SPL. The child is taught by their parent to respond when they hear a sound. The child will respond by completing a behavioural task appropriate to their developmental level. For example, young infants may turn their head in response to a sound and be rewarded with social reinforcement from their parent behind. Once a child is older, they may post items in a box in response to the sounds (detection), and when a child is developmentally able, they may repeat back the sound they heard, or select a card from a range that represents the sound they heard (identification). For example, children may be taught that the sound /m/ is associated with the picture of ice-cream, thus when the child hears the /m/ sound, they would select the card that has the ice-cream. The parent scores the child based on whether they responded or not, and, if they are at the identification stage, the parent would note if the child could hear the sound as well as correctly identify which sound it was. For example, a child may detect the /s/ sound, indicating they have heard something, but may select the wrong card, indicating they do not have enough audibility to obtain accurate information from the sound.

Information from the Ling Sound Test is used by parents to verify the effectiveness of their child's hearing aid/s or cochlear implant/s. Because parents perform this short check each day, a change in performance, especially if over several days, may indicate that they need to consult with their audiologist. A change in performance in the Ling sound test may indicate that the child's device is damaged, or hearing thresholds may have changed, such as that experienced with otitis media with effusion or a progressive hearing loss. In comparison, if their child responds correctly to all of the sounds, it is an indication that their child has access to the entire speech spectrum, at normal conversational level, in quiet conditions within the distance the sounds were spoken at. If a child does not respond correctly, parents may move closer or further away from their child to establish the child's 'listening bubble'; the area within which the child has access to all of the speech sounds.

Although the Ling Sound Test is most commonly used as a daily tool for parents to verify the effectiveness of their child's hearing aids, the test has also been used for other purposes. As an example, in New Zealand, audiologists may use pre-recorded, narrow bandwidth Ling sound stimuli to obtain an aided audiogram in clinic. Additionally, a recent study evaluated the usefulness of the Ling 6-Sound Test as a bedside screening tool in adults. Burgdorf (2014) reported adults with hearing loss were significantly less able to repeat back the sounds of the Ling 6-Sound Test than were their peers with hearing thresholds within normal limits, concluding it could be a useful bedside tool in detecting hearing loss in this population. In conclusion, the Ling Sound Test is a widely used clinical tool that has the flexibility that allows it to be appropriate to use with children across a range of developmental stages.

The Ling 7-Sound Test can be used clinically and in the home as soon as an infant can sit up and turn their head, and subsequently using a developmentally appropriate response task. Similarly, the PEACH/ TEACH rating scales, are designed to be used with children who have reached the ceiling score on the LittlEARS as opposed to children of a certain chronological age, which facilitates its use with children with a range of developmental trajectories. There is a lack of research of alternate methods of detecting hearing loss in young children with developmental disabilities. Given that children with Down syndrome typically experience a delay in their acquisition of skills (Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015) it is usually appropriate to apply the same measures for children with Down syndrome that would be used with slightly younger, typically developing children.

1.2.4 Methods of audiological diagnostic evaluation

Hearing loss in preschool children can be diagnosed using a variety of behavioural and objective measures such as ABR, visual reinforcement audiometry, play audiometry, speech perception tests such as the Kendall Toy Test, the otoacoustic emission test and tympanometry/acoustic reflex tests. ABR, visual reinforcement audiometry and the Kendall Toy Test and otoacoustic emission testing have been used to evaluate the hearing of the participants in the past and have helped to build a picture of what is known of the children's hearing. In the current study, play audiometry and tympanometry were appropriate to detect middle ear effusion.

During ABR, the ear is stimulated with tone bursts (short tones containing energy at a single pure tone frequency). Response to the tone bursts is measured through electrodes placed on the scalp and mastoid bone located behind the ear. These electrodes measure electrical activity in the auditory nerve and brain stem that coincides with the timing of the tone bursts. The measured electrical activity is then amplified, averaged and displayed as a waveform by computer software. The audiologist then finds the lowest intensity of tone burst at which a response waveform can be repeated (threshold).

Visual reinforcement audiometry (Lidén & Kankkunen, 1969) is a method of assessment usually used with babies with a developmental age of 6-30 months, however it may also be used with older children who cannot be conditioned to perform play audiometry. The infant must be able to execute a conditioned head turn in response to sound. The head turn is rewarded with some form of visual reinforcement such as a lighted, animated toy adjacent to the speaker producing the stimuli (Porter & Tharpe, 2010). During visual reinforcement auditory, stimuli are usually presented through speakers in the sound field, which results in frequency specific thresholds for the better ear (Porter & Tharpe, 2010). Headphones and bone conduction headbands, whereby the transducer is held in place on the child's mastoid by a metal headband, should used to obtain frequency specific and ear specific information, and determine the type of hearing loss (Ministry of Health, 2016). An infant with Down syndrome may not be able to sit supported and produce a head turn at 6 months due to their delay in motor control, so the VRA procedure may not be suitable with this population until an older age (Porter & Tharpe, 2010). Greenberg, Wilson, Moore, and Thompson (1978) found that infants with Down syndrome required a mental age of 10-12 months, measured by the Bayley Scales of Infant Development, before visual reinforcement audiometry could be completed successfully. It would also be necessary to confirm the child's ability to hold their head up for a period of time and be able to make repeated head turns in the direction of a visual reinforcer.

Play audiometry (Lowell, Rushford, Hoversten, & Stoner, 1956) is a more complex task for the child than visual reinforcement audiometry and is therefore used with older children. In play audiometry the child becomes a more active listener and is conditioned to withhold their response, wait for the presentation of auditory stimuli, and perform a motor task to indicate they have heard the stimuli (Lowell et al., 1956). Stimuli may be presented via a speaker in the sound field, via air conduction headphones or bone conduction transducers, and may be presented to each ear separately to give frequency specific information, ear specific information, and determine the type of hearing loss. Typically developing children can usually learn to complete the task once they are about 2 ½ years old. However, as with visual reinforcement audiometry, children with developmental disabilities may need to be older before they can reliably complete the task (Cole & Flexer, 2015).

Detection of the test stimulus requires both sensation (integrity of the peripheral auditory system) and perception (cognitive auditory processing abilities) (Norrix, 2015). As such, children vary in their ability to respond at their true sensory threshold. A consideration of the use of such behavioural techniques is that the child may respond at an elevated sensation level. Children are reported to respond within 5 to 10 dB of their true threshold (Karzon, 2007). The level at which a child responds varies with the stimulus type, developmental age and state of the child (Sabo, 1999). Children have been reported to respond closer to true threshold when speech stimuli are utilised, compared to pure tones or frequency modulated pure tones (warble) (Karzon, 2007; Sabo, 1999). As a child gets older, a less intense stimuli will be required to elicit a behavioural response (Sabo, 1999). A child must be alert and motivated in order to respond close to their sensory threshold (Sabo, 1999; Shott et al., 2001). For children with developmental disabilities, the age at which they respond at their true threshold may be older than their typically developing peers (Karzon, 2007). Obtaining results using behavioural testing methods such as visual reinforcement audiometry (VRA) and play audiometry can be challenging with young children with Down syndrome due to cognitive and behaviour difficulties along with the demands of the assessment environment often exceeding the child's capabilities (Porter & Tharpe, 2010) (see section 1.2.6).

The Kendall Toy Test is a speech perception assessment commonly used in paediatric audiology in New Zealand and has many similarities to the Ling 7-Sound Test. This assessment consists of 5 sets of minimal pairs (for example, "car" and "star"). The child is shown the toys or pictures that represent the 10 words and is instructed to point to the corresponding item/picture when the audiologist speaks the stimulus word. The presenting audiologist obstructs their mouth to ensure no visual cues are provided and uses a sound level meter to monitor the intensity of their voice. Pre-recorded words can also be presented via a loudspeaker. A passing level of at least 90% of the test items correctly identified at 40-50 dB

A is generally accepted as being consistent with normal hearing thresholds (Ministry of Health, 2016). The above method of completing the Kendall Toy Test in New Zealand is understood to be adapted from the Australian version (Antognelli, 1986).

Objective measures such as tympanometry/ acoustic reflex testing and distortion product otoacoustic emission testing (DPOAE) can provide useful information about the health of the middle ear, the function of the inner ear and its neural connections, and whether or not a hearing loss is present. However, these measures cannot tell the degree of the hearing loss, if one is in fact present.

Tympanometry is an objective measure used to detect middle ear pathology such as otitis media with effusion, through measuring mobility of the tympanic membrane (Harris, Hutchinson, & Moravec, 2005). Tympanometry involves sealing the ear canal with a probe tip, which contains three parts: a probe unit which produces a tone, an air pump which changes the air pressure in the ear canal, and a microphone which measures the sound pressure level of the tone in the ear canal. Once the ear canal is sealed, the tympanometer generates a tone into the ear canal. Some of the sound energy from the tone is reflected off the tympanic membrane and measured with the microphone while the air pressure in the ear canal is swept from positive to negative. The data is then converted to graphic form, known as a tympanogram. Tymapnograms are interpreted with regard to the shape of the graph, from which measures such as the equivalent middle ear pressure at the point of peak compliance and the equivalent volume of the ear canal can be read. A type A tympanogram displays a peak and is consistent with normal middle ear function. Poor mobility/ low compliance of the tympanic membrane along with a low volume (known as a type B, or flat tympanogram) may be associated with middle ear effusion in young children (Onusko, 2004). A type C tympanogram displays a negative peak pressure and can be associated with Eustachian tube dysfunction. Tympanometry is commonly used to detect middle ear effusion in New Zealand, though it is not always accurate. In a review of 16 studies including 3784 children, 81% of children with otitis media with effusion presented with Type B tympanograms (sensitivity), while 75% of those without otitis media with effusion presented with Type A or C tympanograms (specificity) (Takata et al., 2003). Similarly to typically developing infants under 6 months of age, children with Down syndrome present with anatomical factors that may impact the results obtained through tympanometry, such as ear canal stenosis, joint laxity and softer tissue constitution (Lewis, Bradford Bell, & Evans, 2011). Ear canal stenosis may result in difficulty inserting the tympanometer's probe tip and a lower ear canal volume measurement, while joint laxity and softer tissue constitution may result in increased mobility of the middle ear system.

Typically, a probe tone of 226 Hz is used in tympanometry for all people over the age of 6 months. However, alternative methods of tympanometry have been suggested when assessing young children with Down syndrome. This included the use of a 1000 Hz probe tone (Lewis et al., 2011) . The use of a 1000Hz probe tone, when compared to a 226Hz probe tone may be more specific in detecting fluid in the middle ear space in a small sample of children ages 6 months to 18 years. Assessment on a larger population of children with Down syndrome is needed to verify the use of a 1000 Hz probe tone, and to determine at which age within this population it is more specific for detecting middle ear fluid.

Distortion product otoacoustic emission testing (DPOAE) should be completed as a cross check when performing ABR, visual reinforcement audiometry or play audiometry to corroborate findings and provide an accurate diagnosis. The DPOAE test can be completed on anyone and does not require a behavioural response from the person. DPOAEs use a sensitive microphone to detect small sounds produced by the cochlea in response to an external sound of a range of frequencies. For an otoacoustic emission to be recorded, the sound must pass through the middle ear to the cochlea, and then the emission must also transmit from the cochlea through the middle ear to be recorded by the microphone sitting in the probe tip in the ear canal. The presence of these emissions from the cochlea is indicative of normal functioning of the outer hair cells in the cochlea (Prieve & Lamson, 2014). As such, otoacoustic emissions are not a test of hearing, however as they are present when hearing thresholds are 40 dB HL or better, they are used to corroborate audiometry results. Otoacoustic emissions are a useful clinical tool as they can usually be obtained very quickly and provide separate ear and frequency specific information. Additionally, otoacoustic emissions do not require the child to perform a behavioural task which can be challenging for children who experience difficulties maintaining attention and understanding test instructions (Lyons, Kei, & Driscoll, 2004). However, the presence of otitis media with effusion or other middle ear pathologies will prevent emissions being present, even if there is no cochlear hearing loss. This is an important consideration when working with children with Down syndrome who may have frequently occurring otitis media with effusion because the clinician then must rely on behavioural testing alone to assess and confirm hearing thresholds.

1.2.5 Monitoring

Regardless of the measures used to assess hearing, children need to be monitored over time to detect changes in hearing. Intensive monitoring results in a high degree of active management. An intensive approach to monitoring has been shown to be effective in enabling high levels of normal hearing in young children with Down syndrome. Shott et al. (2001) used a combination of an initial diagnostic ABR followed by regular appointments with an audiologist and regular phone calls from a professional to monitor the hearing of young children with Down syndrome. A 98% normal hearing rate in 48 children with Down syndrome was achieved when an intensive management approach was adopted. Children were seen for audiological evaluation every 6 months. If the children had stenotic ear canals, they were seen every 3 months. Additionally, parents received a phone call every two weeks to monitor any care received from general practitioners and Ear, Nose and Throat doctors. This intensive approach was also associated with a substantially higher retention rate than has been previously reported. Only one child was lost to follow up, a marked difference to the rate reported by Park et al. (2012) who found 25.3% of parents of children with Down syndrome in Utah who failed their newborn hearing screening were lost to follow up.

1.2.6 Evaluating hearing in children with developmental disabilities

There are several challenges associated with assessing the hearing of children with developmental disabilities. For children with developmental disabilities, including those with Down syndrome, a hearing test can be frightening and unpredictable (N. Thompson & Yoshinaga-Itano, 2014). This may be because it is outside of their normal routine, and it involves people, places and objects that are unfamiliar. The result of this is that the child may exhibit behaviour that makes it difficult for the audiologist to successfully complete the evaluation completely (N. Thompson & Yoshinaga-Itano, 2014). Various strategies can be employed by parents and audiologists to prepare children for the tasks involved in an audiology appointment such as using a visual schedule or performing role play with headphones at home. Visual schedules display a sequence of events and can result in the child feeling more calm and willing to participate because they know what to expect and the transitions that will occur (N. Thompson & Yoshinaga-Itano, 2014). Parents use role play at home, such as wearing headphones and playing listening games, or show their child videos of children having their hearing tested to prepare them for the appointment (N. Thompson & Yoshinaga-Itano, 2014). Such an approach may also capitalise on the relative strength children with Down syndrome often have in visual processing (Feeley et al., 2011). Working collaboratively with parents can further enhance audiologists' understanding of children's hearing, which is of particular importance if a comprehensive diagnostic test battery is not practical.

1.2.7 The role of parents

Parents of children with Down syndrome can play an important role in the monitoring and management of their child's hearing loss. In order for a parent to best support their child's hearing, they must have basic information about hearing, hearing loss, signs of hearing loss and management of hearing loss (Valle, 2018). The clinician must also support parents to understand the impact hearing loss may have on language development, and provide emotional support and guidance when necessary (Valle, 2018). The Institute of Medicine (2004) defines health literacy as the "interaction between the skills and knowledge of individuals and the demands of the health system". To bridge the gap between current knowledge and skills and the requirements of the health system, the audiologist can find out the current knowledge of the parent, and support them in building their health literacy. Parents/caregivers are more likely to adhere to health care advice for their child when they understand the advice and believe the advice is beneficial (DiMatteo, 2004). In order for information to be accessible to the public, it is recommended that it be written at a fifth or sixth reading grade level (Doak, Doak, & Root, 1996; Weiss & Coyne, 1997). In the situation of a young child with Down syndrome, a parent may be more likely to adhere to advice given about their child's hearing if they understand it, believe their child is at risk of developing otitis media with effusion, believe this would be severe or have negative impacts on the child, such as poor hearing which may impact language acquisition, and believe that the proposed course of action is manageable and beneficial. Additionally, the self-efficacy and locus of control a person has can impact their likelihood of adhering to health advice (Lawrence & Rittner, 2009). A person with high self-efficacy is more likely to adhere to advice as they feel more confident in themselves to carry out the recommended course of action. An audiologist can increase a parent's feeling of self-efficacy through educating them about how hearing loss affects the communication development of their child, how to monitor their child's hearing, and through providing feedback to parents on their child's development (Blaiser, 2012). Similarly, a person with an internal locus of control is more likely to adhere to advice as they believe they are in control of their child's health, as opposed to believing external forces outside of their control are responsible for their child's health.

Parents of children with Down syndrome have been reported to be more frequently lost to follow up in audiology when compared to parents of typically developing children (Park et al., 2012). Children with Down syndrome are at an increased risk for cardiac, respiratory and digestive problems, and will likely be receiving early intervention such as physiotherapy, speech language therapy, and occupational therapy (Bull, 2011). In New Zealand, children with Down syndrome are typically enrolled in some form of multi-disciplinary early intervention service during infancy and early childhood (Alliston, 2007). As such, the number of commitments and appointments may be great, and may be of higher priority than hearing and ear health (DiMatteo, 2004). Ensuring children enrolled in such services have access to sound is especially important for those therapies reliant on language such as speech therapy and early intervention teaching. Additionally, because a delay in speech and language development is often considered typical for those with Down syndrome, there is potential for diagnostic overshadowing, in which symptoms are attributed to a person's underlying disability (McClimens, Brennan, & Hargreaves, 2015). As a result, hearing impairment may not be regarded as a contributor to speech and language delays.

1.3 Habilitation

Children with Down syndrome are an at risk population for hearing loss. The negative implications of not actively managing hearing loss in children with Down syndrome can extend to their speech and language development and auditory processing abilities (Laws & Hall, 2014; Nightengale et al., 2017). Therefore, every hearing loss, no matter how severe, warrants management. New Zealand audiology protocols encourage audiologists to provide parents of children with Down syndrome with information about mild hearing loss and speech and hearing milestones (Ministry of Health, 2016). Medical treatment and audiological management for conductive hearing loss in children resulting from otitis media with effusion includes “watchful waiting”, antibiotics, grommets, air conduction hearing aids, or bone conduction hearing aids (Rosenfeld et al., 2016). There are limitations associated with each of these management options such as prolonged hearing loss (Rosenfeld & Kay, 2003), resistance to antibiotics (Rosenfeld et al., 2016), premature extrusion of grommets (Robb & Williamson, 2016), and poor retention of hearing aids (Munoz, Preston, & Hicken, 2014). Communication strategies such as reducing background noise; slowing rate of speech; reducing distance between the child and interlocutor, supporting speech with sign and child and getting the child’s attention before speaking to them, are considered to be effective in mitigating the negative effects of otitis media with effusion (Rosenfeld et al., 2016).

1.3.1 Watchful waiting

Otitis media with effusion commonly resolves spontaneously. Due to this, general practitioners and audiologists manage children with otitis media with effusion with watchful waiting for a period before more active management options are considered. This period is usually approximately three months from the first clinical evidence of effusion (Rosenfeld et al., 2016). Children with risk factors, such as those with Down syndrome, may be referred to an Ear, Nose and Throat specialist sooner for consideration of grommet surgery (see section 1.3.4). There is a risk that watchful waiting will result in prolonged hearing loss for those with persistent otitis media with effusion. However, in the interest of not intervening in a usually self-limiting condition and unnecessary use of resources, a three month time frame is generally recommended (Rosenfeld et al., 2016). During the three month period, it has been reported that 75-90% of cases of otitis media with effusion occurring after an episode of acute otitis media will spontaneously resolve and 56% with newly diagnosed otitis media with effusion indicated by a type B tympanogram will resolve to a non-type B tympanogram in typically developing children (Rosenfeld & Kay, 2003). To minimise the impact on the child's hearing and language development during the period of hearing loss due to otitis media with effusion, professionals are advised to educate families about the natural course of otitis media with effusion, the importance of timely follow up appointments, the sequelae the child may experience and communication strategies to employ, assuming a child's hearing will be affected (see section 1.3.2) (Rosenfeld et al., 2016). Furthermore, as children with Down syndrome are at an increased risk of experiencing chronic and recurrent otitis media with effusion as well as delayed language development, this education and demonstration of communication strategies to employ is of the upmost importance.

1.3.2 Communication strategies

Communication strategies are the first habilitation method recommended to families following a diagnosis of otitis media with effusion, prior to other management being recommended. In particular, communication strategies and environmental modifications such as getting the child's attention before speaking to them, reducing background noise, reducing the distance between the speaker and the child, and speaking slowly and clearly can be employed to mitigate the negative impact of otitis media with effusion (Rosenfeld et al., 2016). Ensuring the child can see the speaker's face will give them access to visual cues that support receptive understanding (Marriage et al., 2017). Similarly, supporting speech with simple signs, gestures and pictures can also support comprehension (Rosenfeld et al., 2016).These strategies are all free of cost, can be implemented immediately, and may enhance communication regardless of whether the child has a hearing loss or not, making them ideal for children with fluctuating losses. If a child presents with persistent otitis media with effusion following the 3 month watchful waiting period when communication strategies are utilised, management strategies such as antibiotics, grommets and amplification will be considered.

1.3.3 Antibiotics

The use of antibiotics to treat otitis media with effusion is a controversial and complex topic. Some children respond well to antibiotics and they are an effective treatment; others experience negative side effects and persistent otitis media with effusion. Children with otitis media with effusion who are treated with antibiotics are more likely to experience full recovery in three months following treatment (Burton et al., 2016). However, children may experience negative side effects of the antibiotics such as vomiting, diarrhoea and skin irritation (Burton et al., 2016). The scientific evidence as to whether treatment with antibiotics results in improved hearing thresholds and decreased need for grommets is of low quality and conflicting. Additionally, caution must be exercised when considering antibiotics as a treatment option due to the risk of bacterial resistance (Rosenfeld et al., 2016). It has been concluded that the risks of treating with antibiotics may outweigh the benefits, and they are therefore not recommended.

1.3.4 Grommets

In cases of chronic middle ear effusion (lasting 3 months or more), children are considered for medical intervention such as grommet surgery. Grommets are small hollow plastic tubes that are inserted into the tympanic membrane to equalise pressure between the outer and middle ear and drain fluid that may not be effectively drained through the Eustachian tube (Simon et al., 2018). Grommets are typically inserted under general anaesthesia by an Ear, Nose and Throat specialist and may be effective for approximately 6 months before they extrude themselves into the ear canal (Simon et al., 2018). One study found that children with Down syndrome who had grommets *in situ* were 3.6 times more likely to have normal hearing thresholds compared to children with Down syndrome without grommets (Shott, 2006). However, low frequency thresholds are typically poorer with a patent grommet, likely caused by the low frequency soundwaves passing through the grommet into the middle ear cavity (Martin, Munro, & Langer, 1997). Additionally, children with Down syndrome have been reported to have premature extrusion of grommets when compared to their typically developing peers (Robb & Williamson, 2016). Therefore, grommets have been found to be effective in improving hearing thresholds in children with Down syndrome while they are *in situ*, but may not remain *in situ* for as long as they do in typically developing children. Due to the incision in the tympanic membrane during grommet insertion, there is also a risk of permanent perforation, granulation or scarring to the tympanic membrane. This risk is increased for those who have had multiple sets of grommets (Simon et al., 2018). The improved hearing thresholds resulting from patent grommets in situ typically outweigh the risks of damage to the tympanic membrane, particularly for children who have not had multiple previous grommet insertions.

1.3.5 Amplification

Hearing aids can provide a child who has hearing loss with much improved access to the sounds necessary for typical speech and language development through shaping and amplifying the incoming signal so that it is audible to the child (Gan, Overton, Benton, & Daniel, 2017). A positive result with hearing aids relies on accurate diagnosis, the hearing aids being fitted correctly, exposure to frequent high quality language at the optimal levels at the hearing aid microphones, and a child wearing the aids consistently (American Academy of Audiology, 2013; Cole & Flexer, 2015). Fitting air conduction hearing aids to a fluctuating loss is challenging because one cannot predict the duration of the hearing loss and young children are often unable to communicate this to their caregivers. This requires frequent appointments to check middle ear status, either with a general practitioner or audiologist, to determine when the aids need to be adjusted.

There is also a risk of over amplification with air conduction aids when the otitis media with effusion resolves and the child's hearing thresholds improve, therefore a bone conduction hearing aid is typically more suitable for children with fluctuating conductive hearing loss. There is less risk of overamplification with bone conduction hearing aids because they are programmed for a fixed range of inputs for bone conduction stimulation direct to the cochlea. In the fluctuation of air conduction hearing, such as with conductive hearing loss, cochlear sensitivity is assumed to remain constant. Therefore, a more stable level of input is heard by the wearer. In contrast, when wearing air conduction hearing aids, the levels of amplification heard may fluctuate due to fluctuating middle ear function, to a degree where the amplification may at times be unsuitable. A young child is unlikely to be able to operate a volume control to regulate the amplification or inform an adult that the volume has become unsuitable. Hence, bone conduction aids are generally considered more suitable for fluctuating conductive hearing losses.

Some children (both typically developing and those with developmental disabilities) will not tolerate objects in or on their ears, and as such, retention of hearing aids can be a challenge for some families (Munoz et al., 2014). A recent study found that most typically developing children with otitis media with effusion who received hearing aids did wear them, however, there was large variability in the duration of use (Gan et al., 2017). The children's usage ranged from occasional to several hours per day.

Children require consistent access to sound to mitigate delays in their speech and language development and therefore need to wear hearing aids for much more than a few hours each day to have adequate access to environmental and speech sounds (Tomblin et al., 2015). Parents can be motivated to adhere to consistent hearing aid use for their child if they receive counselling from their audiologist about the benefits of consistent amplification (Blaiser, 2012). Audiologists may demonstrate what a hearing loss is like to parents by asking them to block their ear canals with their fingers, wear earmuffs, or by using an online hearing loss simulator. The audiologist may explain that an adult with established language may be able to sufficiently predict the message even without a clear acoustic signal. In contrast, a child is still developing language and relies on having a clear acoustic signal in order to communicate effectively with others (Blaiser, 2012).

In summary, watchful waiting, antibiotics, grommets and amplification may be effective in some children. However, not all children will have access to these management options in a timely manner due to wait times for services, due to the cost that may be associated with treatment and the feasibility for the family to attend appointments.

Regardless of the type of management recommended for a child with otitis media with effusion, it is imperative that such management occurs in a timely manner to maximise the benefits of early intervention, especially for children with developmental disabilities.

1.4 Study Rationale

- The present study is investigating the use of the Ling 7-Sound Test to detect conductive hearing loss due to its high incidence in children with Down syndrome (Park et al., 2012).
- Parents of children with Down syndrome are generally supportive of the need for research in the area of audiology (Fortnum et al., 2014).
- Current management of fluctuating conductive hearing loss as a sequelae to otitis media generally requires complete audiological information, which can be more difficult to obtain in children with Down syndrome (Tharpe & Seewald, 2016).
- Children with Down syndrome are already at an increased risk for language difficulties and delays (Abbeduto et al., 2007), thus the potential to respond to reduced hearing status in a timely fashion is important.
- Medical management, such as the use of antibiotics and grommets, and audiological management is not always optimal, so there is a need for research investigating alternative methods of diagnosis and management of hearing change in young children with Down syndrome (Robb & Williamson, 2016).

1.5 Research questions

The current study is interested in exploring the following research questions:

- What is the extent to which the children with Down syndrome experience fluctuating conductive hearing loss as a result of otitis media with effusion?
- What are the experiences of parents when administering the Ling 7-Sound Test?
- What is the relationship between tympanometry, play audiometry, the Ling 7-Sound Test, the PEACH, TEACH and social validity measures?
- What is the extent to which the Ling 7-Sound Test, PEACH and TEACH are sensitive to conductive hearing loss?

CHAPTER 2 METHODS

2.1 Introduction

This descriptive study employed a case study design to investigate the utility of an approach to monitoring hearing over a ten week period in a cohort of preschool children with Down syndrome attending the same early intervention service. The five measures used in the current study to monitor hearing were tympanometry, the Ling 7-Sound Test, Parents' Evaluation of Aural/ Oral performance in Children (PEACH), Teachers' Evaluation of Aural/ Oral performance in Children (TEACH), and a social validity questionnaire and semi-structured interview.

2.2 Participants

This study involved four children with Down syndrome, their parents/caregivers, and three of the four children's early intervention educators. Early intervention educators are employed by the early intervention centre and support children at the mainstream early childhood centre that the child attends, from the time they are three years old. Early intervention educators typically spend three hours a week with each child and use this time to integrate the early intervention programme into the child's routines at the early childhood centre and liaise with the staff employed in each setting. All children were four years of age at the beginning of the study. The children attend the Champion Centre in Christchurch with their parent/s for centre based, family-centred, multi-disciplinary early intervention one day a week. Three of the children did not have any sensorineural hearing loss prior to the study, evidenced by diagnostic audiology results completed within the 6 months prior to their recruitment. One child did not have sensorineural hearing loss prior to the study but had not had an audiology evaluation in the 6 months prior to the study commencing. Therefore, at the beginning of the present study a diagnostic assessment was carried out for this child, including otoscopy, tympanometry and separate ear air conduction thresholds, all of which indicated normal hearing.

2.3 Measures

The measures used in the current study were the Ling 7-Sound Test, audiological measures (otoscopy, tympanometry and play audiometry), PEACH, TEACH, and a social validity questionnaire.

2.3.1 Ling 7-Sound Test stimuli

The present study will use the Ling 7-Sound Test. See section 1.2.3 for discussion of the Ling Sound Test. The Ling 7-Sound Test stimuli used in this study were recorded according to the following procedure. The Ling 7-Sound Test stimuli were recorded in a sound treated room, using a Bruel and Kjaer 2250 4192 0.5 inch microphone and the researcher’s voice. The noise floor in the sound treated room was 20dB A. The Ling sound stimuli were produced by a native New Zealand English speaker at typical conversation level using normal vocal effort, positioned 60cm from the microphone; with an absolute peak recording level of 60 dB A. A female voice was utilised because this was similar to mothers’ voices who were most likely to be completing the Ling 7-Sound Test and also to that of therapists, majority of whom are women.

The duration and intensity of the speech sound stimuli are two adjustable variables of the Ling Sound Test. For the purposes of this study, the 7 stimuli were recorded to be two seconds in duration and were attenuated to 45 dB A. The rationale for this duration was based on the recommended duration of stimuli for play audiometry and visual reinforcement audiometry for (1-2 seconds) (Ministry of Health, 2016). Children with Down syndrome have been reported to have lower alertness and endogenous attention when responding to stimuli when compared to their typically developing peers (Moore, Oates, Hobson, & Goodwin, 2002). Therefore, it may take children with Down syndrome longer to process information that they may have more difficulty paying attention to, due to poorer attention regulation, than typically developing children. Importance was also placed on the naturalness of the sounds. In running speech, the sounds exist for less than one second. It became increasingly difficult to record the sounds for greater than two seconds, while maintaining the natural quality of the sounds with consistent pitch and intensity. For these reasons, the sounds are approximately two seconds in duration (see Table 2).

The rationale for the intensity of the sounds was to utilise a sensation level loud enough to obtain a response from the children when they had normal hearing thresholds, but quiet enough that the children would not respond when they experienced a flat or a slightly rising configuration conductive hearing loss as a result of otitis media with effusion. There also needed to be a positive signal to noise ratio between the background noise level in the children's homes, which when sampled ranged between 25 and 35 dB A, and the stimuli. It was deemed appropriate that the intensity of the Ling stimuli be in keeping with the intensity used clinically to assess hearing, such as the Kendall Toy Test (see section 1.2.4). The criteria to pass this test is for the child to correctly identify at least 90% of the test items at 40-45 dB A (Ministry of Health, 2016). Additionally, an intensity close to that of a normal voice effort was utilised to improve the face validity of the task to the parents. In natural speech fricative sounds are less intense than voiced vowels. However, for the purposes of the current study the phonemes were all attenuated to a fixed sound pressure level of 45 dB A at the ear. The Ling sounds were played by the parents using an iPhone mobile phone model 5 or 6 through a Bluetooth speaker (Clip2, JBL). Android mobile phones were not used in the current study due to the variability in microphones and speakers between models.

To obtain the frequency response of the Bluetooth speaker (Clip2, JBL), white noise was played through the speaker connected to a laptop running a software-based digital spectrum analyser in the same sound treated room. The output spectrum was recorded with a dynamic microphone (FX-508; Yoga) at a 44.1 kHz sample rate at three intensities (10 dB attenuation, 20 dB attenuation and 30 dB attenuation) to ensure a flat frequency response as shown in Figure 2.

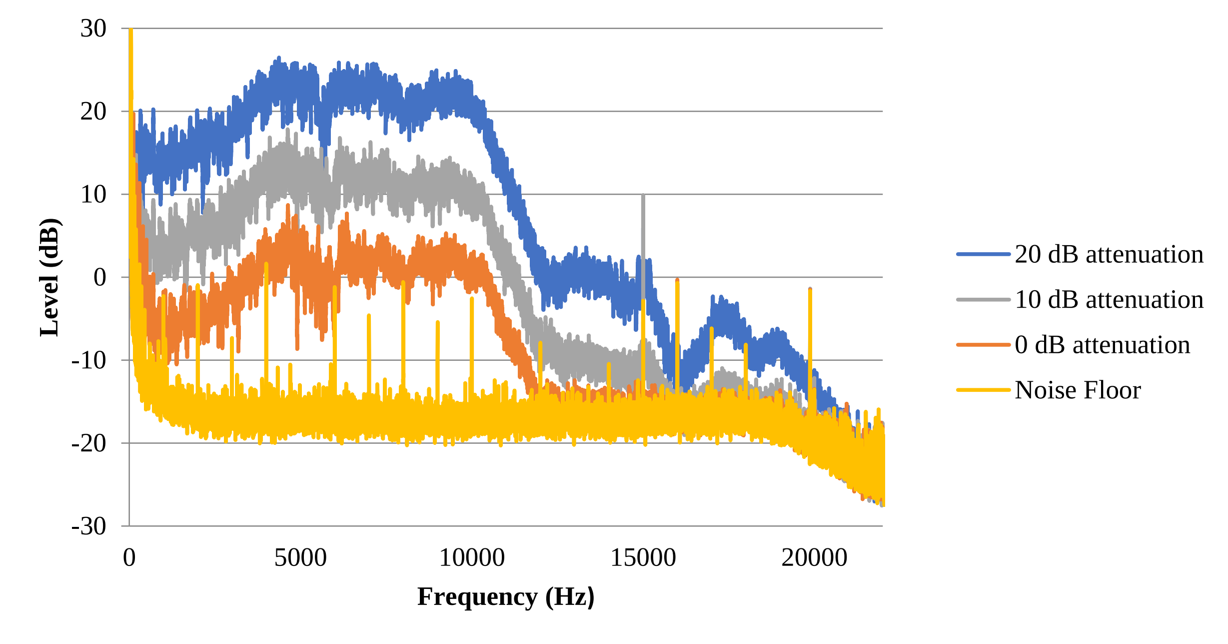

Figure 2. Spectra displaying frequency response of Bluetooth speaker at 0, 10 and 20 dB attenuation

This output was recorded at a 60cm distance, 0 degrees azimuth from the Bluetooth speaker (Clip2; JBL). The sound file, in .wav format, was split so that each sound was a separate .wav file with silence either side of the 2 second audio. They were then converted to stereo files (one channel silence, one channel stimulus because there were two speakers inside the Bluetooth speaker, which could not be used together due to interference) using Audacity software version 2.2.2. The individual sound files were further converted to .mp3 format using iTunes, transferred to an iPhone 5, and played through the “Notes” application which was simple to use, and did not alter the sound level by applying an onset or offset ramp, which could be perceived in other applications. Audacity software version 2.2.2 was used to attenuate the sound files, so that they were within 1 dB of 45dB A when played through the iPhone and speaker both on the highest volume setting. This was verified using a second sound level meter (Solo, 01dB) in a sound proof room with an ambient noise level of 20 dB A. The sound level meter was positioned at the same height as the Bluetooth speaker, at a 1 metre distance, 0 degrees azimuth.

The vowel and nasal sound files were analysed in Praat version 5.3.55 for the frequencies of formants 1, 2 and 3 displayed in Table 2.

Table 2. Duration and formant frequencies of nasal and vowel Ling 7-Sound Test stimuli

	Speech sound	**Duration**	**F1 (Hz)**	**F2 (Hz)**	**F3 (Hz)**
/m/	"mm"	2.36s	288	1819	2772
/a/	"ah"	2.38s	883	1639	2975
/u/	"oo"	2.23s	443	1779	2657
/i/	"ee"	2.36s	428	2262	3430
/ɔ/	"or"	2.33s	566	1088	2648

Audacity was utilised to analyse the frequency content of the 7 Ling stimuli used in the current study (see figure 3).

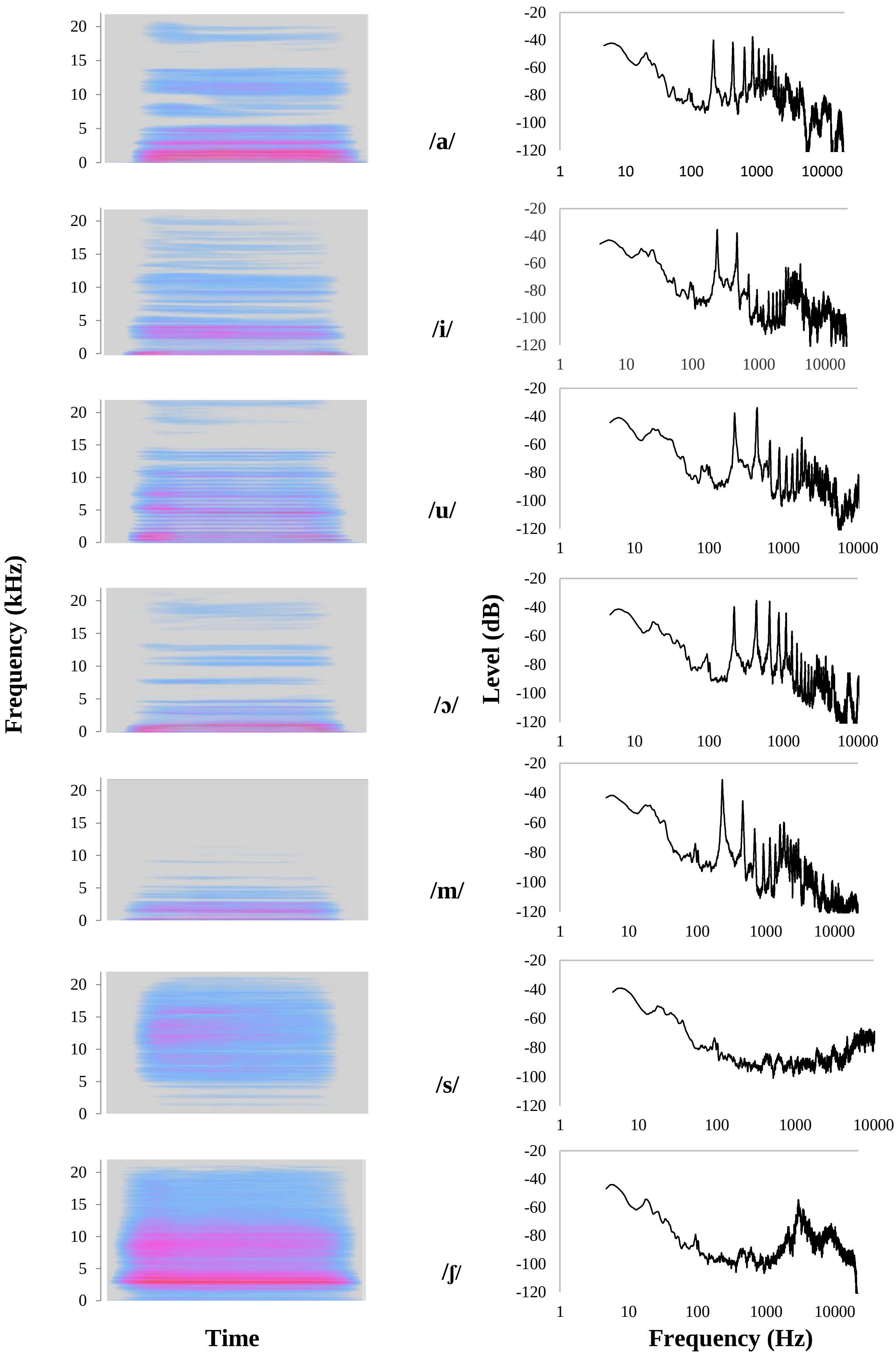

Figure 3. Spectrograms (left) and corresponding spectra (right) of the Ling stimuli.

2.3.2 Audiological measures

The three audiological measures utilised in the current study were otoscopy, tympanometry and play audiometry. The first measure otoscopy, is a method of viewing the tympanic membrane, typically with a hand held otoscope. Otoscopy was completed prior to tympanometry each week to ensure no contraindications for performing tympanometry were present such as a discharging ear. The second measure, tympanometry, is an objective measure which measures mobility of the tympanic membrane, equivalent middle ear pressure and equivalent ear canal volume. Tympanometry was completed each week at the early intervention centre to detect the presence of middle ear fluid. The third measure is play audiometry, in which the child is conditioned to withhold their response, wait for the presentation of pure tone auditory stimuli, and perform a motor task to indicate they have heard the stimuli (Lowell et al., 1956). Play audiometry was performed any time there was a binaural change in tympanometry results. While tympanometry can be used to detect fluid behind the tympanic membrane, pure tone audiometry can be used to determine if such fluid has resulted in a conductive hearing loss.

2.3.3 Parents' Evaluation of Aural/ Oral performance in Children (PEACH) questionnaire

Two questionnaires were utilised to examine children's aural/oral performance. The first was the Parents' Evaluation of Aural/ Oral performance in Children (PEACH) questionnaire (Ching & Hill, 2005). The PEACH questionnaire consists of 13 questions in which parents are asked to reflect on their child's listening over the past week and answer the questions using the 5 point Likert scale (never-rarely-sometimes-often-always). The questions consist of scenarios in both quiet and noisy environments. For example: "when you call, does your child respond to his/her name in a quiet situation?" and "How often does your child understand what you say in the car/bus/train?". A "never" response is scored 0; "rarely" is scored 1; "sometimes" is scored 2; "often" is scored 3 and "always" is scored 4, for all questions except “how often has your child complained or been upset by loud sounds?” for which the opposite direction of scores is applied. Scores are added and used to calculate a “quiet”, “noise” and “overall” score that can be converted into a percentage. A higher score is indicative of better perceived listening ability.

2.3.4 Teachers’ Evaluation of Aural/Oral performance in Children (TEACH) questionnaire

The second questionnaire utilised to examine children’s aural/oral performance was the Teachers’ Evaluation of Aural/Oral performance in Children (TEACH) questionnaire (Ching & Hill, 2005). The TEACH questionnaire consists of 11 questions in which teachers are asked to reflect on the child’s listening over the past week and answer the questions using the same 5 point Likert scale as the PEACH. A not applicable option is also available. The questions are similar to those in the PEACH questionnaire and consist of scenarios in both quiet and noisy environments. For example: “How often does the child initiate/participate in conversation in a quiet situation?” and “When asked, does the child follow simple instructions or do a simple task in a noisy situation?”. As with the PEACH, scores are added and used to calculate a “quiet”, “noise” and “overall” score that can be converted into a percentage. The majority of the questions included in the PEACH and TEACH are not specific to hearing aid users, so it was proposed in the present study that the recorded observations may provide useful insight into the fluctuating hearing of children with Down syndrome.

2.3.5 Social validity questionnaires and semi-structured interviews

Two novel social validity questionnaires were developed for the present study and were completed in the context of semi-structured interviews. The aim of these measures was to ascertain the perspectives of parents and the early intervention team leader regarding the children’s hearing and their experiences during the study. The first questionnaire was to be completed by parents, the second by the early intervention team leader. The social validity questionnaires posed questions regarding the training, how easy it was to incorporate the Ling 7-Sound Test into the family routine, the usefulness of the information gained from the Ling 7-Sound Test, and how confident parents/teachers were in the results obtained from each of the measures. Parents answered the 22 questions using a 5 point Likert scale of never-rarely-sometimes-often-always. One question asked for a short written answer. Similarly the early intervention team leader answered 12 questions with the same 5 point Likert scale. Following completion of the questionnaires, the researcher engaged the parents and early intervention team leader in individual semi-structured interviews to explore the reasoning behind their responses given in the social validity questionnaire. The interviews were audio recorded and transcribed by the researcher for qualitative analysis.

2.4 Procedure

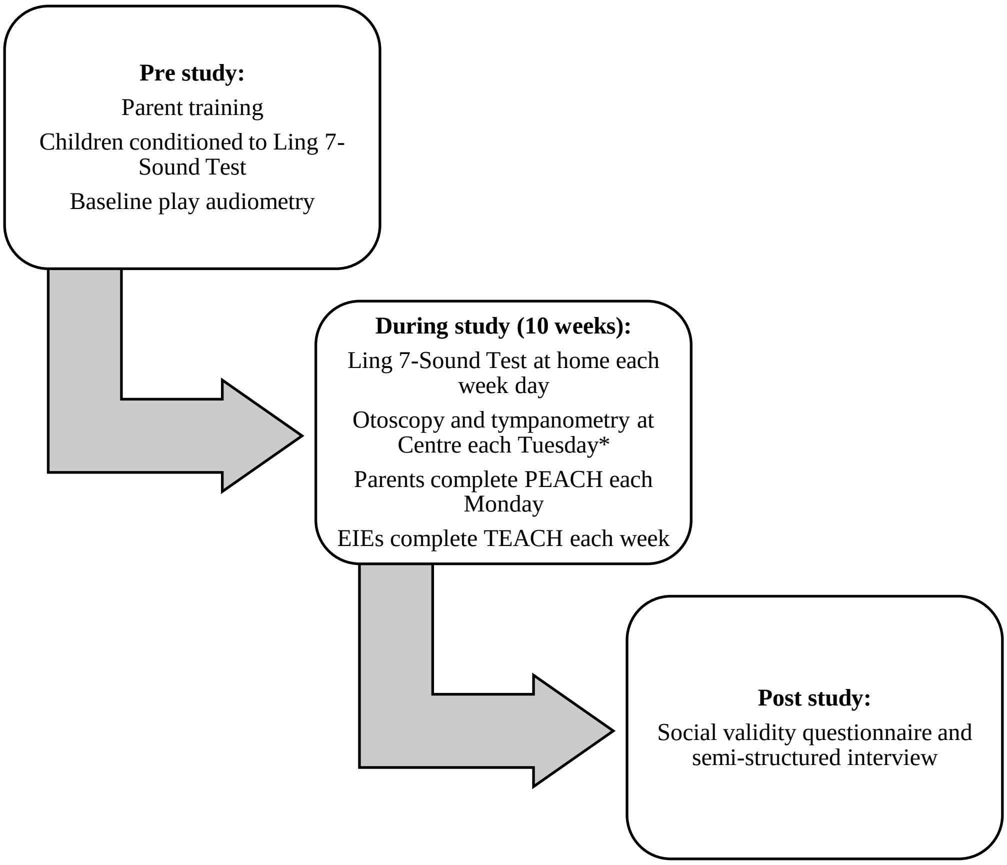

Figure 4. Overview of the procedure followed in the current study.
Note EIEs is an abbreviation for early intervention educators.
*Play audiometry completed at centre following binaural change in tympanometry results.

2.4.1 Subject recruitment

The following recruitment method was approved by the University of Canterbury Educational Research Human Ethics Committee (see appendix A). All children ages 3-6 years with Down syndrome and their parents who attend the Champion Centre were invited to participate in the present study. There were no exclusion criteria; however, tympanometry would not be performed on any children who had had grommets inserted fewer than six weeks prior, as this may compromise ear health. Children and parents attend the Champion Centre for therapy on a weekly basis. During one such weekly session at the Champion Centre, parents and their children with Down syndrome were invited to participate in the study by way of a 20 minute group presentation outlining the aims and methodology, the author's involvement and information about middle ear health and hearing loss. The researcher used a detailed poster of the ear to describe otitis media with effusion and conductive hearing loss. It was explained that children with Down syndrome experience otitis media with effusion and conductive hearing loss at a higher rate than their typically developing peers, and that the resultant hearing loss may have an impact on their speech and language development. The researcher explained that parents would complete the Ling 7-Sound Test on a daily basis, the researcher would assess the children's middle ear status with a tympanometer at their weekly sessions, parents and early intervention educators would complete the PEACH and TEACH questionnaires respectively each week, and at the completion of the study parents and the early intervention team leader would complete a social validity questionnaire and semi-structured interview.

The researcher played a demonstration version of the Ling 7-Sound Test on her mobile phone to the parents to demonstrate the task that they would be completing at home. It was discussed that the researcher and parents would work together to decide on motor tasks that would be motivating for the children to use as a response method, and that parents would use a score sheet (see Appendix C) to record their results from the Ling 7-Sound Test. The researcher then demonstrated the use of a tympanometer, and explained that it would be used in this circumstance to detect results consistent with the presence or absence of fluid in the middle ear space.

The researcher described the PEACH and TEACH questionnaires and showed copies of each to the parents. Finally, the researcher explained that parent observations and reflections of their experiences during the study would be recorded in a short questionnaire and semi-structured interview upon completion of the study. Following the description and demonstration of all measures, parents were given the opportunity to ask questions about the study, and were given reassurance that the researcher would be present at each of their Tuesday early intervention mornings and able to provide support as needed. Parents were provided with an information sheet about the study, and consent and assent forms to complete if they wished to participate (see Appendix B). Parents were asked to return the consent forms to the researcher within two weeks. This time frame was necessary to ensure parents had adequate time to consider their participation and ask any questions they had before training commenced. The children's early intervention educators were invited to participate via phone or email following the return of the parent consent forms.

Four parents and their children agreed to participate in the study. These children's four early intervention educators (EIE) were also invited to participate, three of whom agreed. The researcher explained that the role of the EIE would be to reflect on the child's listening and communication in the early childhood centre setting by completing the TEACH questionnaire each week. Early intervention educators were provided with information and consent forms and were asked to return these within a two week period if they were willing and able to participate (see Appendix B).

2.4.2 Parent training

Following recruitment, the researcher held an education and training session for parents during their morning session at the Champion Centre. The researcher taught the parents how to administer the Ling 7-Sound Test to their child, and to record their results on a provided A4 form (see Appendix C) that they could keep on the fridge to remind them to do the test each day. This session also involved transferring the sound files to the parents' phones (and providing an iPhone to one family), playing these through the provided speaker, and demonstration of the use of the speaker provided.

To ensure that the results were reliable and that the entire speech spectrum was assessed, the importance of playing all of the sounds twice each in the random order determined by the researcher was explained. Parents were instructed to avoid providing any non-stimulus cues such as movement and to vary the interval between stimulus presentations. It was encouraged that parents provided social reinforcement to their child following response to the Ling stimuli.

The researcher discussed with parents the possible response options for their children. Many of the children had not completed play audiometry previously, but instead had completed visual reinforcement audiometry, in which the child is required to make a conditioned head turn to a visual reinforcer in response to sound. Therefore, it was discussed that a head turn is acceptable for the Ling 7-Sound Test although there may be more motivating tasks that they may like to use such as posting games - dropping a block in a bucket or racing a car down a track. All information that the therapist delivered verbally was reinforced with written information, at no higher than 5th reading class level (approximately 10 years of age). This written information was particularly important as one family did not speak English as their first language. This family was included in the study because they had a high level of understanding English and could speak the language fluently.

Once informed consent was received, the researcher went to each family's home to teach them how to complete the Ling 7-Sound Test as per the study protocol. To ensure as many variables as possible were consistent throughout the study, the researcher and the parents first worked together to determine the setting in which the Ling 7-Sound Test would be completed. Taking into account the configuration in the family's home, and once the location had been decided, a 1 metre marker was also set, at which point the parent would hold the Bluetooth speaker behind their child.

Following the set up, the researcher and parent began the conditioning phase whereby the Ling 7-Sound Test was practiced by the parent to ensure adherence to the protocol and ensure that their child was able to respond correctly. Practice was continued until the parent reported that they felt confident to complete the Ling 7-Sound Test as per the study protocols, including measuring and recording the background noise on the phone application, presenting the stimuli in the randomised order provided, presenting the list of stimuli twice, not providing any additional cues to their child, and recording their child's health, attentiveness and Ling 7-Sound Test scores. Parents were also provided with PEACH questionnaires and given instructions on how to complete these.

The three early intervention educators who agreed to participate in the study were provided with a sufficient supply of TEACH questionnaires for the entirety of the study as well as written instructions for completing the questionnaires, with relevant sections highlighted. The early intervention educators were also given the opportunity to ask questions.

To further aid with conditioning the children to perform the Ling 7-Sound Test, the researcher led a short group song in the group music session at the Champion Centre that the children take part in each week. The song had a familiar tune (Old McDonald) and intuitive lyrics so parents and other staff could easily join in. The lyrics used were "When we play our listening game we stop, wait, listen. And when we hear a sound behind (pause for parents to say Ling sound) we put our block inside". The researcher instructed parents to say "ah" when prompted and guide their child to post a wooden block into a container when they heard the "ah" in the song. All children participated well with help from their mothers to post the block. The song was completed twice each Tuesday in group music until all children were conditioned to respond to all of the Ling 7-Sound Test stimuli during the musical activity three weeks later.

2.4.3 Study overview

For 10 weeks parents administered the Ling 7-Sound Test to their child each week day and recorded the results for each sound on the A4 form provided (see Appendix C). Parents returned this form each week to the researcher, who provided them with a new one for the following week. The researcher performed tympanometry on all children participating each week during their weekly session at their early intervention centre, unless the child presented with any contraindications for performing tympanometry. The researcher was available to provide any support when needed and answer any questions the parents had. To avoid confirmatory bias, the researcher did not view the weekly results from the Ling 7-Sound Test until the weekly tympanometry had been completed. The parents were informed of any results from tympanometry that were clinically significant (i.e. tympanometry that yields a Type B result consistent with fluid behind the eardrum) and advised of the recommended management such as consultation with a general practitioner and/or to employ modified communication strategies. If a child was found to have a binaural change in their tympanometry results, play audiometry was completed to confirm if a hearing loss was present. Parents were instructed that the results from tympanometry would not necessarily correlate with the results from the Ling 7-Sound Test, and that it was reasonable for the child's hearing to fluctuate over the duration of the study. Additionally to the Ling 7-Sound Test and tympanometry, parents and teachers completed the PEACH/TEACH questionnaire each week. Parents were reminded via text message to complete the Ling 7-Sound Test and PEACH and return the forms to the centre each week. Parents and the early intervention team leader also completed the novel social validity measure and semi-structured interview following the 10 week data gathering period, in which they reported on their experience of taking part in the study.

2.4.4 Ling 7-Sound Test

The Ling 7-Sound Test, as described above, was completed by parents and their children each week day for the duration of the study. Parents used a mobile smartphone (iPhone 5 or 6, Apple corp.) connected to a Bluetooth speaker (Clip2; JBL) to play the seven sounds. Parents were all provided with the same model speaker, and also with an appropriate model iPhone for the duration of the study if they did not already own one. Parents were instructed to complete the Ling 7-Sound Test at a time they felt that their child was at their best and would be likely to pay sufficient attention to the stimuli. The Ling 7-Sound Test was completed in the exact location each day, where practical. To ensure the environment was quiet enough, the score sheet (see Appendix C) reminded parents to ensure the television and radio were off and the room was quiet. Additionally, parents recorded the background noise level with a smartphone sound level meter application (National Institute for Occupational Safety and Health (NIOSH)) over a period of 30 seconds. The purpose of this was for parents to gauge whether the environment was quiet enough for the child to be able to hear the Ling stimuli with normal hearing and make modifications to reduce the level of background noise when required. The smartphone sound level meter application performance has been evaluated recently, and when used with a MicW i436 external microphone, was found to comply with the International Electrotechnical Commission (IEC) 61672-3 standard (Celestina, Hrovat, & Kardous, 2018). In the current study, the internal microphone of the phone was utilised. As such, the app provided parents with an approximate level of background noise. As well as reminding parents to ensure the environment was quiet, the score sheet also asked parents to report on their child's health and attention using a 3-point Likert scale each day the Ling 7-Sound Test was completed. Once parents had completed the background noise check, and documented their child's health and attention they set their child up with a motivating task such as posting toys into a container, usually at the table, and instructed the child to "stop, wait and listen". Following this, the parent stood 1 metre behind their child and played the list of sounds twice, in the provided random order. Parents were instructed to vary the time between each sound to avoid a presentation rhythm the child may respond to. Parents then recorded whether the child responded to either of the two presentations of each sound. Each score sheet allowed for five days of recording and had space for parents to record any comments they had about the experience. Parents brought the score sheet to the early intervention centre each week, where the researcher could clarify any comments and ensure completeness of the documentation.

2.4.5 Otoscopy and tympanometry

Otoscopy was completed before tympanometry to examine the ear canal and tympanic membrane, and ensure there were no contraindications to performing tympanometry such as recent grommet insertion, discharging ear or otitis externa. Each week tympanometry was completed at the early intervention centre by the researcher using a hand held portable Interacoustics MT10 tympanometer. Otoscopy and tympanometry were completed within a relaxed and familiar environment during a quiet play task. Children were presented with a visual schedule which displayed pictures and a basic description of the steps involved in tympanometry so they understood the process each time (see Appendix E). Tympanograms were classified according to the guidelines of the American Speech-Language-Hearing Association (1990), and consistent with the University of Canterbury clinical protocols.

2.4.6 Play audiometry

Play audiometry was performed using a portable screening audiometer (Interacoustics AS208) when a child had a change in tympanometry result binaurally or parents presented with concerns about the child's hearing. It was deemed unnecessary to perform pure tone audiometry more frequently as parents had received education about communication strategies, and there was a very low likelihood of a child developing a progressive hearing loss during the 10 week study period. A conditioned play audiometry technique was used to obtain the threshold at 2000 Hz and 500 Hz in one ear and 4000 Hz and 1000 Hz in the other ear, consistent with the University of Canterbury clinical protocols. The play audiometry was completed in a quiet non-sound treated room at the early intervention centre that the child was familiar with. Because the room was not sound-treated, only air conduction and not bone conduction thresholds were obtained because of the background noise level at the early intervention centre. As with tympanometry, children were provided with a visual schedule which displayed pictures and a basic description of the steps involved in having their hearing tested so the children understood what was expected (see Appendix E). Children were able to be referred for a comprehensive diagnostic hearing assessment if the play audiometry results yielded any concerns, such as results not consistent with tympanometry results or air conduction thresholds outside the normal range.

2.4.7 PEACH

The purpose of the PEACH questionnaire in the context of this study was to measure parents' perception of their child's listening in the environments they are typically in and to determine if the questionnaire was sensitive to changes in hearing such as those from otitis media with effusion. This questionnaire was completed by parents each week for the duration of the study. Questions not applicable to this cohort (such as hearing aid and cochlear implant use) were removed. Parents were also asked to reflect on the 3 days of the week before tympanometry was performed on the child rather than the entire week. This was to reduce the averaging that parents would be doing, which in turn could blur the reflection, if a child was demonstrating they were listening well some days of the week but less so on others. This method assumed that that it was unlikely a child's hearing would change significantly in three days. However, parents were asked to comment in the comments section on the last page of the questionnaire if their child had been unwell or if the child's listening behaviour had changed significantly within the 3 day period.

2.4.8 TEACH

The purpose of the TEACH questionnaire in the context of this study was to measure early intervention educators' perception of the child's listening in the early childhood centre environment and to determine if the questionnaire was sensitive enough to detect changes in hearing such as those from otitis media with effusion. This questionnaire was completed by 3 early intervention educators each week for the duration of the study. Again, questions not applicable to this cohort (such as hearing aid and cochlear implant use) were removed. Early intervention educators were also asked to complete the TEACH on the one day of the week they were with the child.

2.4.9 Social validity questionnaires

Upon completion of the study, parents and the early intervention team leader completed novel social validity questionnaires (see Appendix D) within a semi-structured interview to gain an in depth understanding of their beliefs and experiences during the study. The interviews were conducted by the researcher and audio recorded. Participants could request any section be deleted or request not to have the interview recorded. Participants could also request a copy of the audio recording for their records. Written notes were taken, and interviews were transcribed by the researcher. Participants were supplied with a copy of the transcription, and could request any items be struck from the transcription.

CHAPTER 3 RESULTS

Four case studies will be presented describing children's ear health and hearing history, the training phase of the study, and results obtained during the study. Results will be reported from tympanometry, play audiometry, Ling 7-Sound Test, Parents' Evaluation of Aural/ Oral performance in Children questionnaire, Teachers' Evaluation of Aural/ Oral performance in Children questionnaire as well as data collected during the social validity questionnaire and semi-structured interview. Social validity data from the children's early intervention team leader will also be presented. Pseudonyms have been used to protect the privacy of the participants.

3.1 Case Study One: Isobel

Isobel was 4 years and 11 months old at the beginning of the study. Isobel lives with both parents and is one of four children. Isobel uses both aural/oral language and New Zealand sign language to communicate. As well as attending the Champion Centre one morning a week with her mother and younger brother, Isobel attends an early childhood centre two days a week where she is supported by a Champion Centre early intervention educator.

3.1.1 Ear health and hearing history

Isobel's hearing was first assessed using automatic brainstem response under the Universal Newborn Hearing Screening programme which she passed. Isobel's hearing was subsequently tested annually as part of the follow up pathway for children with Down syndrome. Isobel had ventilation tubes inserted in 2015 as a result of persistent otitis media with effusion. Isobel's early intervention educator reported that generally Isobel is more responsive to family members speaking and less so to other people unless they are loud, and that she is visually active and can lose focus on auditory information if the environment is visually busy.

Isobel attended a hearing review appointment at the hospital 6 months before the present study began. At this appointment, bilateral type B high tympanograms were found, but it was not clear if this result was due to patent grommet/s or extruded grommets with tympanic membrane perforations. This uncertainty was present because although a grommet was visible in the left ear during otoscopy, the audiologist was not able to determine whether the grommet was patent or extruded and no grommet was visualised in the right ear. The stenotic nature of her ear canals and the challenges of persuading her to have things in her ears meant this distinction could not be made. Play audiometry revealed a mild conductive hearing loss in her right ear. The left ear was not assessed as it proved too difficult to retain Isobel's interest over the period of time required to complete the task. Isobel's mother reported no concerns about Isobel's hearing because her speech sounds were developing well and the articulation of some of her words had become clearer recently. Isobel had a history of ear infections with the most recent ear infection occurring 3-4 weeks prior to this hearing review appointment, for which she was treated with antibiotics. Isobel did not present with any symptoms of an ear infection at the time of the study commencing.

3.1.2 Training

Isobel was compliant throughout the practice and enjoyed posting cards in a box as the response method. Isobel's mother provided social reinforcement following Isobel's correct responses. Isobel's older sister and grandmother were also present and provided social reinforcement during this conditioning phase. Isobel was able to complete the Ling 7-Sound Test with little instruction during this first practice.

3.1.3 Study

Table 3. Isobel's Results Week 1

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		67.5	
Tuesday	B high				
Wednesday		100			50
Thursday					
Friday		86	/u/		

Week 1: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Isobel and her mother completed the Ling 7-Sound Test on the Monday, Wednesday and Friday. Isobel scored 100%, 100% and 86% respectively. On the Friday Isobel did not respond to the /u/ sound; no comments were provided regarding Isobel's attention.

Isobel scored an overall PEACH score of 67.5% (75% in quiet, 60% in noise) and an overall TEACH score of 50% (55% in quiet, 44% in noise). Isobel's early intervention educator reported that Isobel looked well after having been unwell in the holidays (the previous two weeks) and that the preschool environment was particularly quiet at the time of evaluation.

Table 4. Isobel's Results Week 2

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		86	/ɔ/	75	
Tuesday	B high				
Wednesday		100			55.5
Thursday					
Friday		100			

Week 2: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Tympanometry was completed at Isobel's home as her sister was

unwell so they could not attend the Champion Centre. Isobel's mother reported that completing the Ling 7-Sound Test 3 times a week is a manageable frequency for their family. As such, Isobel and her mother completed the Ling 7-Sound Test on the Monday, Wednesday and Friday. Isobel scored 86%, 100% and 100% respectively. On the Monday Isobel did not respond to the /ɔ/ sound; no comments were provided regarding Isobel's attention.

Isobel scored an overall PEACH score of 75% (87.5% in quiet, 60% in noise) and an overall TEACH score of 55.5% (65% in quiet, 44% in noise).

Table 5. Isobel's Results Week 3

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		68	
Tuesday	B high				
Wednesday		100			58
Thursday					
Friday		100			

Week 3: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Isobel and her mother completed the Ling 7-Sound Test on the Monday, Wednesday and Friday. Isobel scored 100% each day. It was reported that Isobel was "very well" and paid attention "always" on these days.

Isobel scored an overall PEACH score of 68% (79% in quiet, 55% in noise) and an overall TEACH score of 58% (65% in quiet, 56% in noise).

Table 6. Isobel's Results Week 4

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		68	
Tuesday	B high				
Wednesday					28
Thursday					
Friday					

Week 4: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Isobel and her mother completed the Ling 7-Sound Test on the Monday and Isobel scored 100%.

Isobel's mother reported that she was well but she had noticed a change in Isobel's behaviour because she was less compliant and seemed clumsy and off balance. Isobel scored an overall PEACH score of 68% (79% in quiet, 55% in noise) and an overall TEACH score of 28% (30% in quiet, 25% in noise). Isobel's TEACH score was lower this week than it has been in the past. Isobel's early intervention educator reported some concerns about Isobel's hearing including the fact that Isobel did not attend to a car alarm going off outside which other children were asking questions about. It was also reported that Isobel started a new early childhood centre and was quieter than usual.

Table 7. Isobel's Results Week 5

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday					
Tuesday	B high				
Wednesday		100			
Thursday					
Friday		100			

Week 5: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Isobel and her mother completed the Ling 7-Sound Test on the Wednesday and Friday. Isobel scored 100% both days.

Isobel's mother was not able to complete a PEACH this week and because Isobel did not attend Playcentre this week, a TEACH could not be completed either.

Table 8. Isobel's Results Week 6

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		73	
Tuesday	B high	86	/u/		
Wednesday		86	/u/		50
Thursday					
Friday		100			

Week 6: Isobel's general health was good. Isobel presented with type B high tympanograms binaurally on the Tuesday. Isobel and her mother completed the Ling 7-Sound Test on the Monday, Tuesday, Wednesday and Friday. Isobel scored 100%, 86%, 86% and 100% respectively. Isobel did not respond to the /u/ sounds on the Tuesday and Wednesday; no comments were provided regarding Isobel's attention.

Isobel scored an overall PEACH score of 73% (83% in quiet, 60% in noise) and an overall TEACH score of 50% (50% in both quiet and noise). Isobel's early intervention educator reported that Isobel is now attending a second early childhood centre, which is a change in Isobel's routine. However, the TEACH was completed based on her observations of Isobel in the Playcentre as this was the more familiar environment although it was an inside day due to poor weather.

Table 9. Isobel's Results Week 7

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		70	
Tuesday	B high (Left)	71	/a/ twice /ɔ/ and /u/ once		
Wednesday					58
Thursday		86	/ɔ/ and /u/ once each		
Friday					

Week 7: Isobel was reported to be slightly unwell this week. Isobel presented with a type B high tympanogram in her left ear on the Tuesday. Her right ear was discharging so tympanometry was not completed. Isobel's mother reported that she noticed a clear discharge coming from Isobel's right ear, for which her general practitioner prescribed antibiotics. The general practitioner could not determine if the discharge was indicative of a burst eardrum or whether her grommets were still patent. Isobel and her mother completed the Ling 7-Sound Test on the Monday, Tuesday and Thursday. Isobel scored 100%, 71% and 86% respectively. On the Tuesday Isobel did not respond to /a/ on both presentations and /ɔ/ and /u/ on one of the two presentations. On the Thursday Isobel did not respond to /ɔ/ and /u/ on one of the two presentations. It was reported that Isobel was "somewhat well" and paid attention "sometimes" on these days.

Isobel scored an overall PEACH score of 70% (79% in quiet, 60% in noise) and an overall TEACH score of 58% (70% in quiet, 44% in noise). Isobel's early intervention educator reported that Isobel responded to sudden loud sounds behind her despite the ear infection.

Table 10. Isobel's Results Week 8

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		93	/ɔ/		
Tuesday	B high (left)				
Wednesday					41
Thursday					
Friday					

Week 8: Isobel's general health was good, except for the ear infection. Isobel presented with a type B high tympanogram in her left ear on the Tuesday. Her right ear was still discharging so tympanometry was not completed. Isobel and her mother completed the Ling 7-Sound Test on the Monday this week, during which Isobel scored 93% as she did not respond to the /ɔ/ sound once. It was reported that Isobel was "somewhat well" and paid attention "sometimes" on the Monday.

A PEACH was not completed this week. Isobel scored an overall TEACH score of 41% (50% in quiet, 37.5% in noise). Isobel's early intervention educator reported that she had to tap Isobel's shoulder to ensure she understood verbal instructions.

Week 9: Isobel was reported to be unwell this week. As a result of her poor health she did not attend the Champion Centre or her early childhood centre. Due to her absence and poor health, tympanometry, the Ling 7-Sound Test, PEACH and TEACH questionnaires were not completed this week.

Table 11. Isobel's Results Week 10

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday					
Tuesday	Did not test	100			
Wednesday		100			
Thursday					
Friday		100			

Week 10: Isobel was reported to be unwell this week also. As a result of her poor health she did not attend the Champion Centre or early childhood centre. Consequently, neither tympanometry nor the PEACH and TEACH questionnaires were completed this week. However, Isobel and her mother completed the Ling 7-Sound Test on the Tuesday, Wednesday and Friday this week, during which Isobel scored 100% each day.

3.1.4 Social validity

In the social validity interview, Isobel's mother reported that she was always concerned about Isobel's hearing at low levels. This concern was due to the reoccurring hearing tests Isobel had had in the past that showed a low level of hearing loss, as well as the difficulty she had getting Isobel's attention in noisy places. Following the training, Isobel's mother reported that she was confident to administer the Ling 7-Sound Test and that because Isobel enjoyed the posting aspect it was often easy to engage her in the task. A challenge of administering the Ling 7-Sound Test that Isobel's mother reported was ensuring that Isobel's older sister did not give Isobel any cues. Isobel's mother reported that receiving a text message reminder each week was very useful in reminding her to complete the Ling 7-Sound Test and PEACH questionnaire. Isobel's mother reported that the Ling 7-Sound Test results always matched her informal observations of Isobel's hearing but the Ling 7-Sound Test results were always informative as the results provided specific information about which sounds she was and was not hearing. Similarly, Isobel's mother reported that the PEACH was also always informative because it provided her with an opportunity to reflect on Isobel's hearing, health and behaviour over the past three days. Isobel's mother reported that the Ling 7-Sound Test and PEACH were equally useful as a hearing monitoring device as the measures work well together to provide holistic information. Finally, Isobel's mother reported that she was equally confident in all of the measures utilised in the current study.

3.1.5 Results summary – Case 1: Isobel

Overall across the ten weeks Isobel presented consistently with type B high tympanograms bilaterally. This result is likely due to ongoing perforations in both eardrums or possibly a patent grommet in the left eardrum. Isobel's Ling 7-Sound Test scores were generally high and ranged from 71% to 100% (see Figure 5). Her lowest score (71%) was obtained during the time she had an ear infection, although she also scored 100% in the Ling 7-Sound Test during this period. Similarly, Isobel's PEACH scores were largely constant throughout the study with overall scores ranging from 67-75% (see Figure 6). Isobel's PEACH scores in quiet were higher than her PEACH scores in noise. Similarly, Isobel's TEACH scores in quiet were also higher than her TEACH scores in noise (see Figure 7). More variability was present in her TEACH scores which ranged from 28-58% overall. Isobel's TEACH scores were consistently lower than her PEACH scores (see Figure 8). Isobel's Ling 7-Sound Test scores were on average higher than the scores obtained through the questionnaires (see Figure 8). Figure 9 can be referred to for overall percentage scores for all measures.

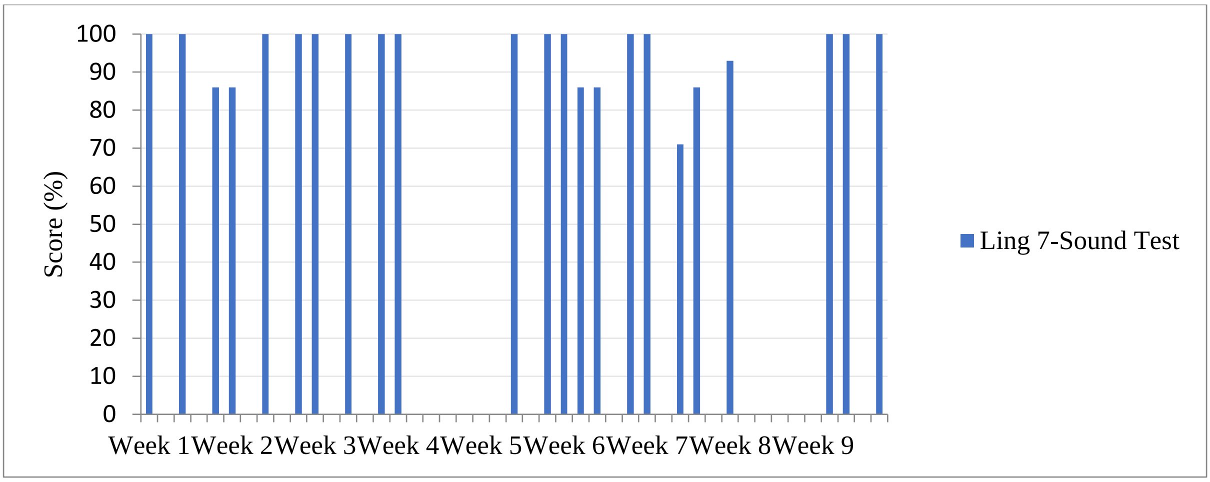

Figure 5. Isobel's Ling 7-Sound Test results during the 10 week study

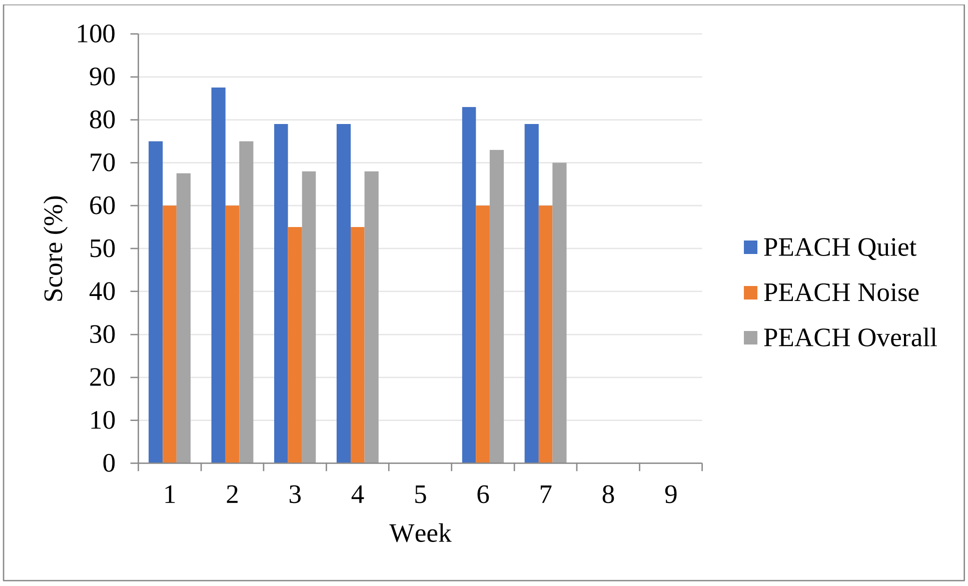

Figure 6. Isobel's PEACH scores during the 10 week study

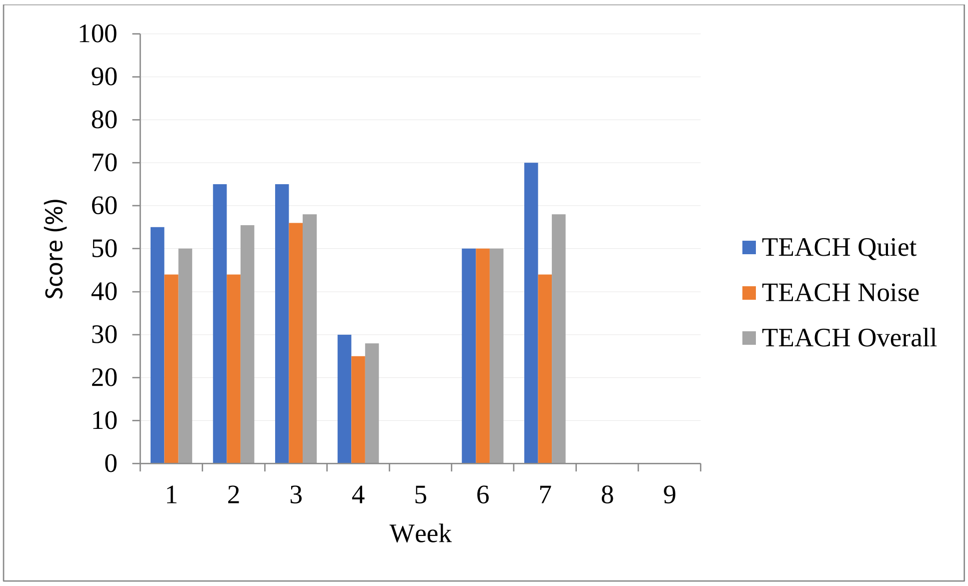

Figure 7. Isobel's TEACH scores during the 10 week study

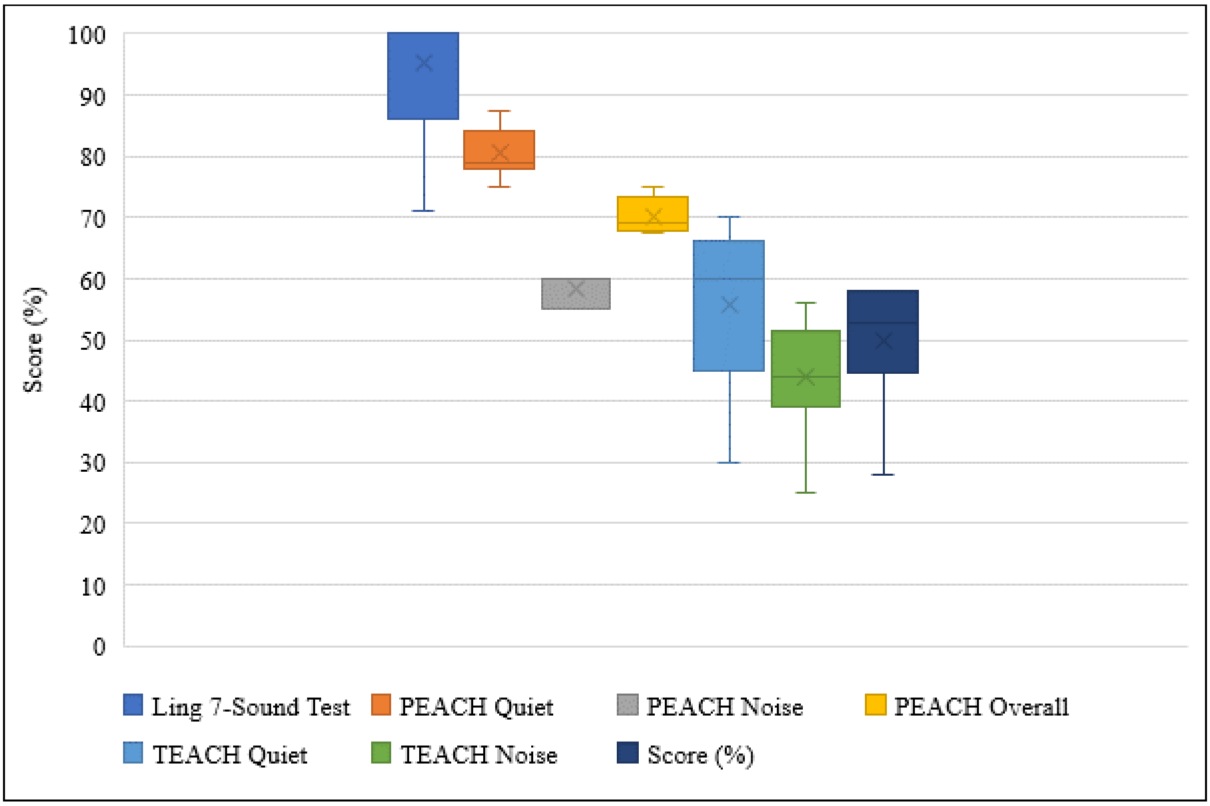

Figure 8. Isobel's Ling 7-Sound Test, PEACH and TEACH scores during the 10 week study

3.2 Case study two: Sam

Sam was 4 years and 4 months old at the beginning of the present study. Sam lives with both parents and his older brother. Sam uses predominantly non-verbal language and New Zealand sign language to communicate. As well as attending the Champion Centre one morning a week with his mother, Sam is supported by a Champion Centre early intervention educator at his grandmother's house once a week. Sam's early intervention educator reports that Sam may not respond to his name or instructions if he is very focused on the activity he is engaged in and that he enjoys toys that have interactive music or animal sound buttons. Additionally, she reports Sam will recognise and talk to his grandmother on the phone, but not typically with other people.

3.2.1 Ear health and hearing history

Sam's hearing was first assessed using automated ABR under the Universal Newborn Hearing Screening programme which he passed. Sam's hearing was subsequently tested annually as part of the follow up pathway for children with Down syndrome. Sam had grommets inserted in 2017 as a result of persistent otitis media with effusion.

Sam attended a hearing review appointment at the hospital two months before the study began. At this appointment Sam presented with a type B high tympanogram in his right ear and a type C tympanogram in his left ear. Prior to this appointment, Sam had had an upper respiratory tract infection which was the likely cause of the negative pressure in his left ear. Sound field visual reinforcement audiometry revealed mildly elevated air conduction thresholds at 500Hz and results within normal limits for 1000-4000Hz in the better ear. Bone conduction thresholds were within normal limits in the better ear. At this appointment Sam's mother reported having no concerns about his hearing or ear infections, although she noted that when he is congested he appears to have difficulty hearing.

3.2.2 Training

Sam was compliant throughout the practice and enjoyed dropping dinosaurs in a bucket as the response method. Sam's mother also provided social reinforcement following Sam's correct responses. Sam was able to complete the Ling 7-Sound Test with little instruction during this first practice.

3.2.3 Study

Table 12. Sam's Results Week 1

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		57	/m/, /s/, /ʃ/	75	
Tuesday	B high (right) B low (left)	43	/u/, /s/, /ʃ/, /m/		
Wednesday		71	/s/, /ʃ/		44
Thursday		71	/s/, /m/		
Friday		71	/s/, /ʃ/		

Week 1: Sam's general health was good. In his right ear, Sam presented with a type B high tympanogram consistent with a patent ventilation tube and a type B low tympanogram consistent with the presence of middle ear fluid in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each week day this week. Sam scored 57% on the Monday, 43% on the Tuesday, and 71% on the Wednesday, Thursday and Friday. On the Monday Sam did not respond to the /m/, /s/ or /ʃ/ sounds, on the Tuesday Sam did not respond to the /u/, /s/, /ʃ/ or /m/ sounds, on the Wednesday Sam did not respond to the /s/ or /ʃ/ sounds, on the Thursday Sam did not respond to the /s/ or /m/ sounds and on the Friday Sam did not respond to the /s/ or /ʃ/ sounds. Sam's mother reported that he paid attention "sometimes" during the Ling 7-Sound Test from the Monday to Thursday and "always" on the Friday. In summary, this week Sam did not respond to the /s/ sound at all or the /m/, /u/ or /ʃ/ variously. Sam's mother reported that he was very tired on the Tuesday which made it difficult to settle him into the task. On the Thursday they completed the Ling 7-Sound Test

later than usual. This was due to Sam participating in a hydrotherapy session during which his mother reported he may have got water in his ears as he had been putting his fingers in them after the session. Sam's mother made the observation that he seems to not respond to the /s/ and /ʃ/ sounds.

Sam scored an overall PEACH score of 75% (70% in quiet, 80% in noise) and an overall TEACH score of 44% (60% in quiet, 25% in noise).

Table 13. Sam's Results Week 2

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		71	/ɔ/, /u/	75	
Tuesday	B high (right) A (left)	86	/s/		
Wednesday		86	/u/		55 (quiet)
Thursday		100			
Friday		100			

Week 2: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each week day this week. Sam scored 71% on the Monday, 86% on the Tuesday and Wednesday, and 100% on both the Thursday and Friday. On the Monday Sam did not respond to the /ɔ/ or /u/ sounds, on the Tuesday Sam was not engaged with the task during the /s/ sounds so did not respond to this sound, and on the Wednesday Sam did not respond to the /u/ sound.

Sam's mother reported that he has begun repeating the sounds back now to indicate he has heard a sound, rather than dropping dinosaurs in a bucket. Sam scored an overall PEACH score of 75% (70% in quiet, 80% in noise) and a TEACH score of 55% in quiet. As Sam is not at an early childhood centre, rather at his grandmother's house, there is not typically sufficient background noise to make an observation of his listening performance in noise.

Table 14. Sam's Results Week 3

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		93	/ɔ/ once	67.5	
Tuesday	B high (right) A (left)	100			
Wednesday		93	/ʃ/ once		65 (quiet)
Thursday		100			
Friday		64	/ʃ/, /s/ twice /ɔ/ once		

Week 3: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each week day this week. Sam scored 93% on the Monday, 100% on the Tuesday, 93% on the Wednesday, 100% on the Thursday and 64% on the Friday. On the Monday Sam did not respond to the /ɔ/ sound once, on the Wednesday Sam did not respond to the /ʃ/ sound once, on the Friday Sam did not respond to the /ʃ/ or /s/ sounds either time or the /ɔ/ sound once. It was reported that on the Friday the Ling 7-Sound Test was completed in a novel environment.

Sam scored an overall PEACH score of 67.5% (80% in quiet, 55% in noise) and a TEACH score of 65% in quiet.

Table 15. Sam's Results Week 4

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		64	/s/ or /ʃ/ twice /m/ once	77.5	
Tuesday	Did not test	100			
Wednesday		100			90 (quiet)
Thursday		93	/s/ once		
Friday		93	/ʃ/ once		

Week 4: Sam's general health was good. Tympanometry was not completed this week as Sam was not in Christchurch. Sam and his mother completed the Ling 7-Sound Test each week day this week. Sam scored 64% on the Monday, 100% on the Tuesday and Wednesday, and 93% on the Thursday and Friday. On the Monday Sam did not respond to the /s/ or /ʃ/ sounds either time and the /m/ sound once, on the Thursday Sam did not respond to the /s/ sound once, on the Friday Sam did not respond to the /ʃ/ sound once. As with the Friday of last week, the Ling 7-Sound Test was completed in a novel environment on the Monday. Sam scored an overall PEACH score of 77.5% (80% in quiet, 65% in noise) and a TEACH score of 90% in quiet.

Table 16. Sam's Results Week 5

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100			
Tuesday		100			
Wednesday		100			90 (quiet)
Thursday	B high (right) A (left)	100			
Friday		100			

Week 5: Sam's general health was good. Tympanometry was completed on the Thursday this week as he was not able to attend the Champion Centre on the Tuesday. At this time Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear. Sam and his mother completed the Ling 7-Sound Test each week day this week during which Sam scored 100% each day. Sam's mother was not able to complete a PEACH this week. Sam scored a TEACH score of 90% in quiet. Sam's early intervention educator reported that Sam did not give any response when the phone rang. However, when a plane flew overhead, he stopped what he was doing and looked at her, then out the window, and

back at her. Additionally when they were playing with coloured blocks, Sam imitated the verbal name of the colours in response to her verbal models.

Table 17. Sam's Results Week 6

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		70	
Tuesday	B high (right) C (left)	100			
Wednesday		100			80 (quiet)
Thursday		86	/s/, /ʃ/ once each		
Friday		50	/ʃ/, /s/ twice each /u/, /a/, /ɔ/ once each		

Week 6: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type C tympanogram consistent with negative middle ear pressure in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each day this week. Sam scored 100% on the Monday, Tuesday and Wednesday, 86% on the Thursday and 50% on the Friday. On the Thursday Sam did not respond to the /s/ or /ʃ/ sound once each, and on the Friday Sam did not respond to the /ʃ/ or /s/ sound either time and the /u/, /a/ and /ɔ/ sound once each. On the Friday Sam was unwell and his mother reported he had significant difficulty maintaining his attention.

Sam scored an overall PEACH score of 70% (75% in quiet, 65% in noise) and a TEACH score of 80% in quiet. Sam's early intervention educator reported instances of Sam attending to environmental sounds and following verbal instructions. For example, Sam stopped what he was doing and looked to her when an oven timer rang and also attempted to point to the correct animal when she asked him to "find the animal that says…".

Table 18. Sam's Results Week 7

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		54.5	
Tuesday	B high (right) C (left)	93	/s/ once		
Wednesday		100			86
Thursday		100			
Friday		78	/ʃ/ twice /s/ once		

Week 7: Sam was reported to have had a temperature and unsettled nights this week. Sam presented with a type B high tympanogram in his right ear and a type C tympanogram in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each day this week. Sam scored 100% on the Monday, 93% on the Tuesday, 100% on the Wednesday and Thursday and 78% on the Friday. On the Tuesday Sam did not respond to the /s/ sound once, and on the Friday Sam did not respond to the /ʃ/ sound and the /s/ sound on one of the two presentations. It was reported on the score sheet that Sam found it more difficult to pay attention on the Thursday and Friday.

Sam scored an overall PEACH score of 54.5% (62.5% in quiet, 45% in noise) and an overall TEACH score of 86% (75% in quiet, 94% in noise).

Table 19. Sam's Results Week 8

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		93	/m/ once	73	
Tuesday	B high (right) A (left)	100			
Wednesday		93	/a/ once		80 (quiet)
Thursday		93	/s/ once		
Friday		100			

Week 8: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each day this week. Sam scored 93% on the Monday, 100% on the Tuesday, 93% on the Wednesday and Thursday and 100% on the Friday. On the Monday Sam did not respond to the /m/ sound on one of the two presentations, on the Wednesday Sam did not respond to the /a/ sound on one of the two presentations and on the Thursday Sam did not respond to the /s/ sound on one of the two presentations.
Sam scored an overall PEACH score of 73% (83% in quiet, 60% in noise) and a TEACH score of 80% in quiet.

Table 20. Sam's Results Week 9

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		86	/ʃ/, /s/ once each	66	
Tuesday	B high (right) A (left)	100			
Wednesday		100			100 (quiet)
Thursday		100			
Friday		86	/ʃ/, /s/ once each		

Week 9: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear on the Tuesday. Sam and his mother completed the Ling 7-Sound Test each day this week. Sam scored 86% on the Monday, 100% on the Tuesday, Wednesday and Thursday and 86% on the Friday. On the Monday and Friday Sam did not respond to the /ʃ/ and /s/ sounds on one of the two presentations each.
Sam scored an overall PEACH score of 66% (71% in quiet, 60% in noise) and a TEACH score of 100% in quiet.

Table 21. Sam's Results Week 10

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		86	/ʃ/, /m/ once each	73	
Tuesday	B high (right) A (left)				
Wednesday					80 (quiet)
Thursday					
Friday					

Week 10: Sam's general health was good. Sam presented with a type B high tympanogram in his right ear and a type A tympanogram in his left ear on the Tuesday, which was the last day of the study. Sam and his mother completed the Ling 7-Sound Test on the Monday during which Sam scored 86% as he did not respond to the /ʃ/ and /m/ sound on one of the two presentations each.

Sam scored an overall PEACH score of 73% (79% in quiet, 65% in noise) and a TEACH score of 80% in quiet. In the TEACH, Sam's early intervention educator reported instances of Sam attending to environmental sounds again this week such as planes and helicopters flying overhead and noticing a fence creaking in the wind.

3.2.4 Social validity

In the social validity interview, Sam's mother reported that prior to the study she was sometimes concerned about Sam's hearing, particularly when he was unwell, and that during the study she noticed the fluctuations in Sam's Ling 7-Sound Test results along with increased difficulty maintaining his attention in the test when he wasn't feeling well. However, Sam's mother reported there were instances when her informal observations of Sam's hearing did not match the results from the Ling 7-Sound Test. Following several weeks of being in the study, Sam's mother reported that completing the Ling 7-Sound Test at home became part of their daily routine with the score sheet and text reminders being helpful sometimes in reminding her to complete the Ling 7-Sound Test and questionnaires. These reminders became less necessary to her once the tasks were part of their routine. A positive aspect of the Ling 7-Sound Test that Sam's mother reported was that when Sam was able to repeat back the sounds, it was an opportunity for Sam to practice his speech sounds. With regard to the usefulness of the measures used, Sam's mother reported that the Ling 7-Sound Test was more useful to her than the PEACH questionnaire because the Ling 7-Sound Test provided real time feedback on Sam's hearing, whereas the PEACH was guiding reflective practice on past listening behaviour, by which time she may have missed out on time to implement communication strategies if he did not appear to be hearing well. Sam's mother reported that she was equally confident in the results obtained from all of the measures used in the study (Ling 7-Sound Test, PEACH, TEACH and tympanometry) as they all gave slightly different information.

3.2.5 Results summary – Case 2: Sam

Overall across the ten weeks Sam presented consistently with type B high tympanograms in his right ear. In his left ear, Sam had a type A tympanogram on 6 weeks, a type C tympanogram on two weeks, and a type B low tympanogram on the first week of the study only. Sam's Ling 7-Sound Test scores ranged between 43% and 100% (see Figure 9). There were 5 instances when he scored below 70%. Sam's overall PEACH scores ranged from 62.5% to 83% throughout the study (see Figure 10). As with Isobel, more variability was present in his TEACH scores which ranged from 55-100% in the quiet category (see Figure 11). Overall, Sam's TEACH scores in quiet were similar, albeit slightly higher than his PEACH scores in quiet (see Figure 11). Figure 12 displays percentage scores for all three measures.

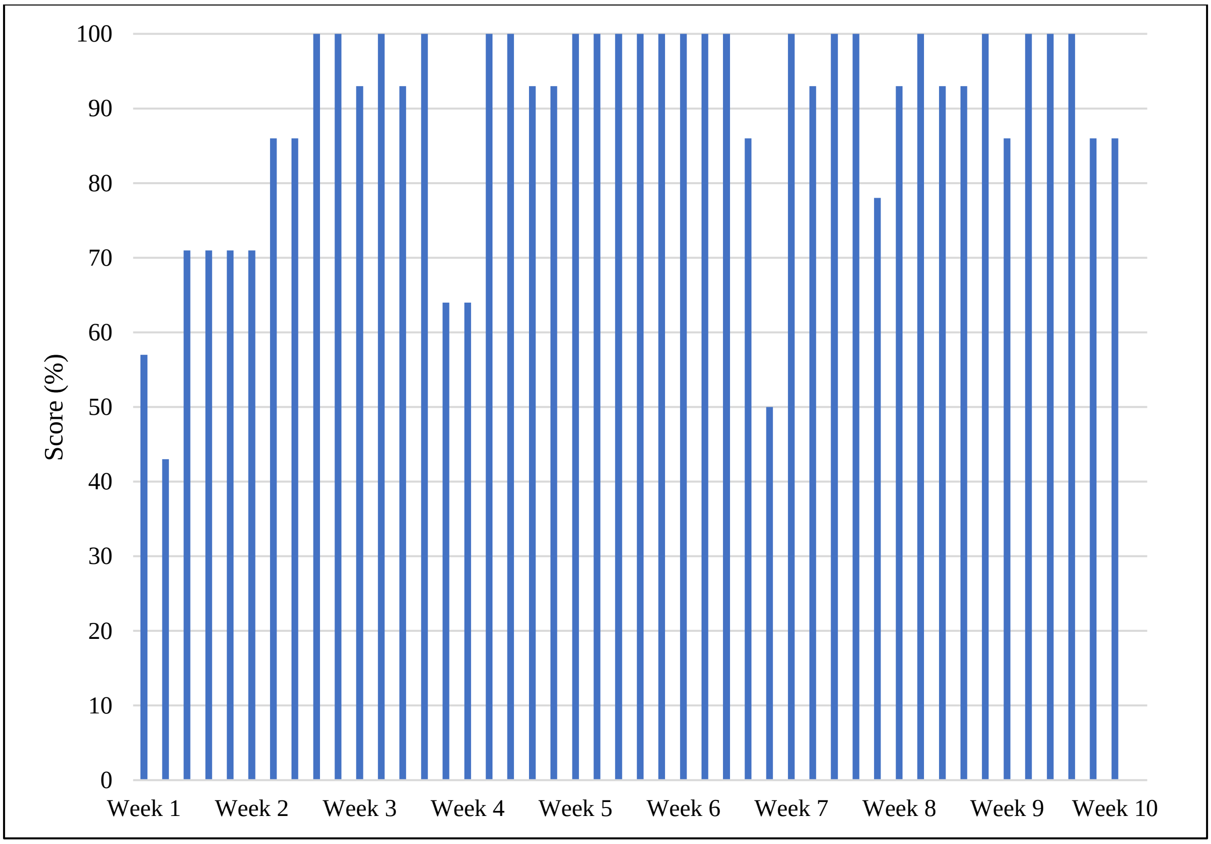

Figure 9. Sam's Ling 7-Sound Test results during the 10 week study

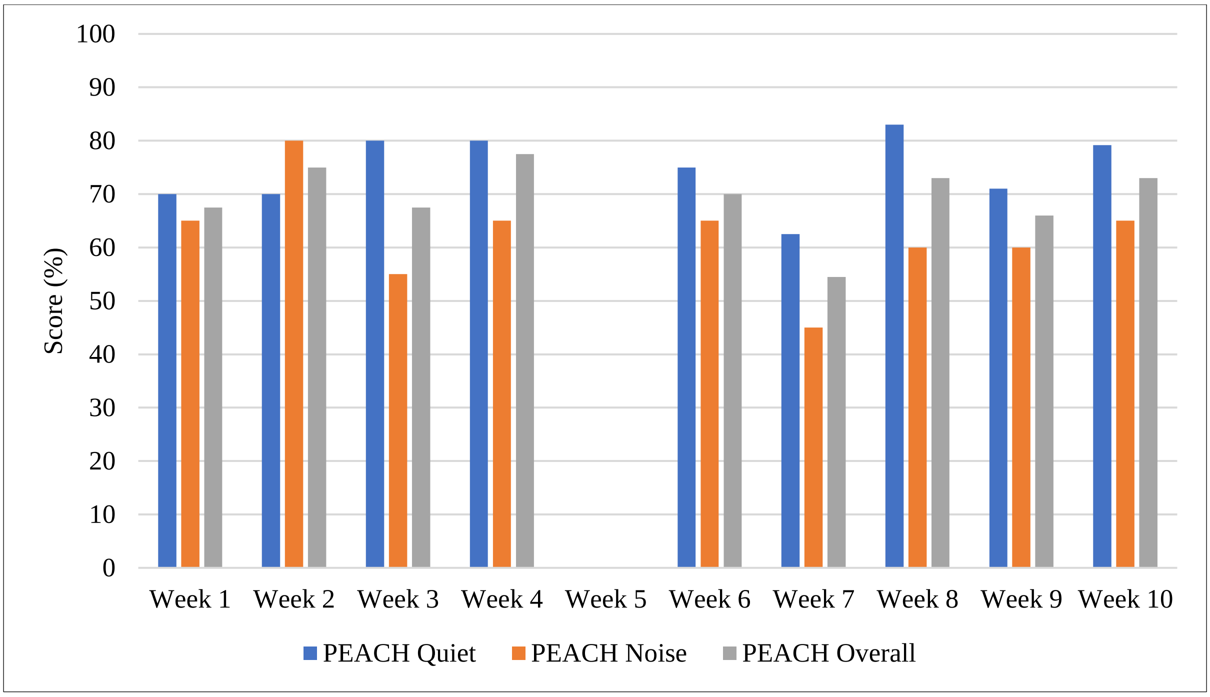

Figure 10. Sam's PEACH scores during the 10 week study

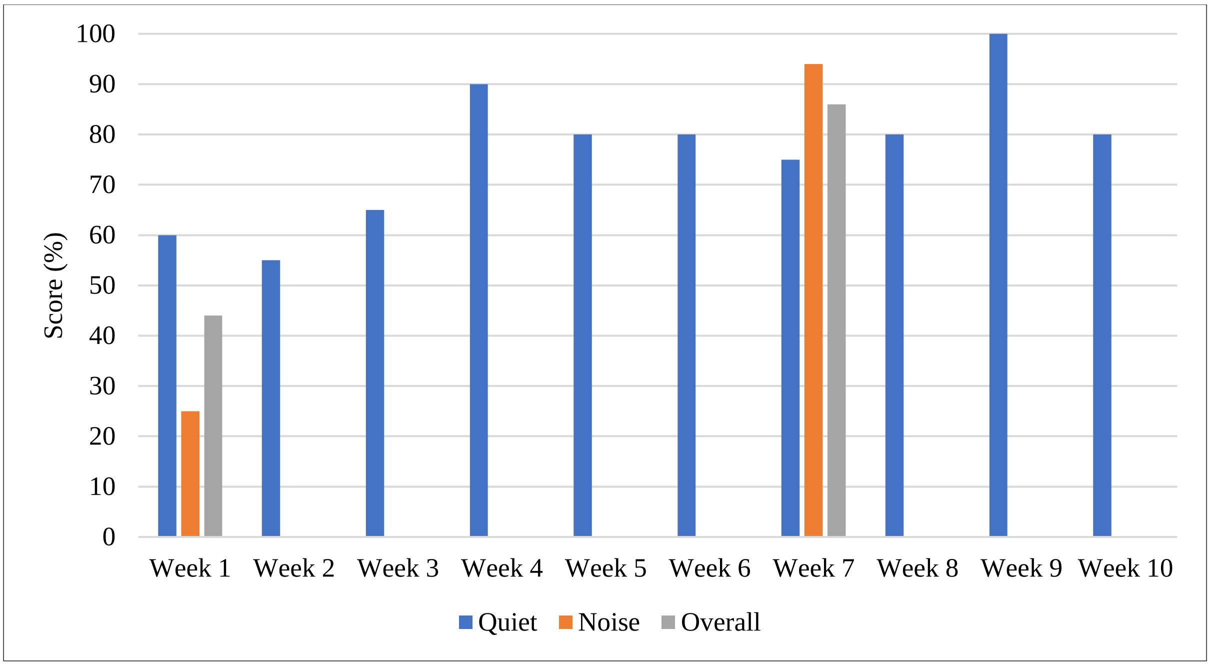

Figure 11. Sam's TEACH scores during the 10 week study

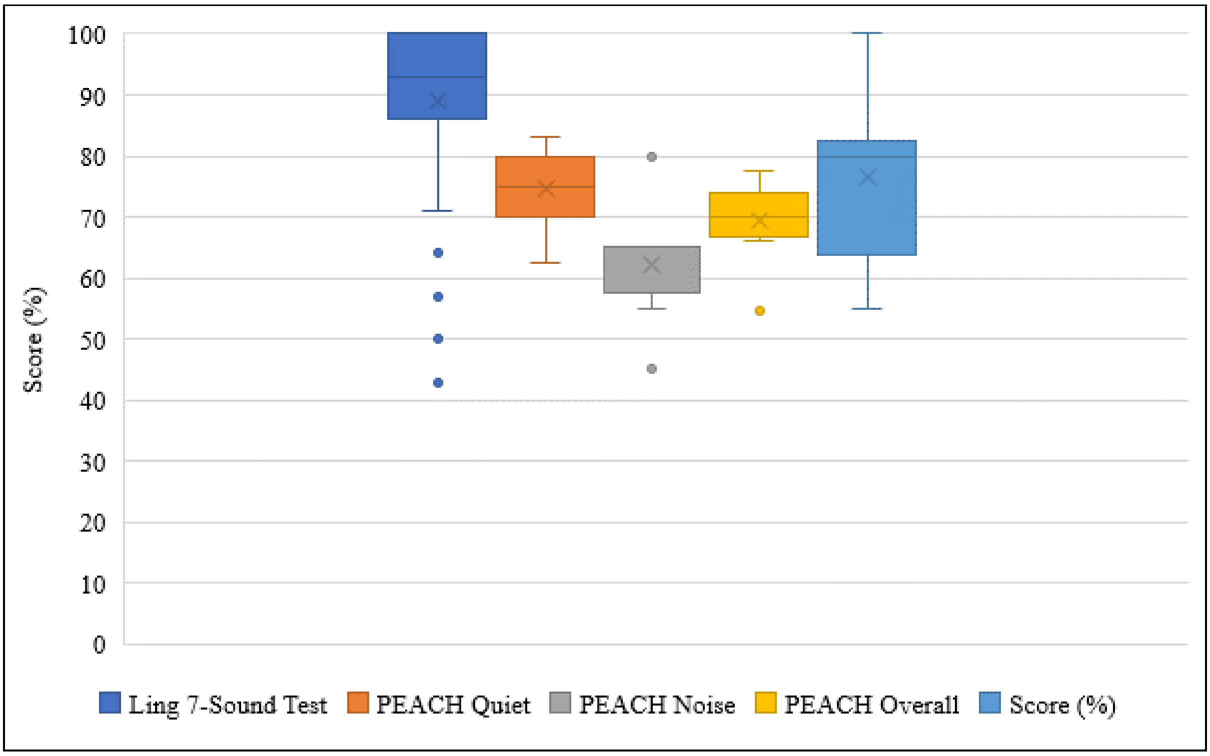

Figure 12. Sam's Ling 7-Sound Test, PEACH and TEACH scores during the 10 week study

3.3 Case study three: Drew

Drew was 4 years and 2 months old at the beginning of the study. Drew lives with both parents and his older sister. Drew uses non-verbal language, oral/aural language, augmentative and alternative communication (visuals and a core board) and New Zealand sign language to communicate. As well as attending the Champion Centre one morning a week with his mother, Drew is supported by a Champion Centre early intervention educator at an early childhood centre once a week.

3.3.1 Ear health and hearing history

Drew's hearing was first assessed under the Universal Newborn Hearing Screening programme which he passed. Drew's hearing was subsequently tested annually as part of the follow up pathway for children with Down syndrome. Drew had ventilation tubes inserted two months before the study began as a result of persistent otitis media with effusion. Because Drew had not had a hearing review at the hospital in the six months before the study began, a hearing assessment was completed one week prior to the study commencing. At this appointment, Drew presented with type B high tympanograms binaurally, consistent with patent grommets *in situ*. Additionally, play audiometry using supra aural headphones revealed air conduction hearing thresholds within normal limits from 500-4000 Hz although only two frequencies were assessed in each ear (2000 Hz and 500 Hz in the right ear and 4000 Hz and 1000 Hz in the left ear) as it proved too difficult to retain Drew's interest over the period of time required to complete the task and he was averse to having transducers in or on his ears. At this appointment Drew's mother reported that she was not concerned about his hearing at this time as he was hearing well with his grommets in.

3.3.2 Training

Drew was compliant throughout the practice and enjoyed holding a block to his ear, waiting for the sound and dropping the block in a plastic container when he heard the stimuli. Drew's mother and sister provided social reinforcement following Drew's correct responses. Drew was able to complete the Ling 7-Sound Test with little instruction during this first practice.

3.3.3 Study

Table 22. Drew's Results Week 1

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		82.5	
Tuesday	B high (right) B low (left)	93	/s/		
Wednesday		100			72
Thursday		100			
Friday		100			

Week 1: Drew's general health was good. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother completed the Ling 7-Sound Test each week day this week. Drew scored 100% each day except Tuesday when he scored 93% as he did not respond to the /s/ sound on one of the two presentations. Drew's mother reported that he paid attention "sometimes" during the Ling 7-Sound Test on the Tuesday and Thursday and "always" on the other days.
Drew scored an overall PEACH score of 82.5% (90% in quiet, 75% in noise) and an overall TEACH score of 72% (65% in quiet, 81% in noise).

Table 23. Drew's Results Week 2

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		82.5	
Tuesday	B high (right) B low (left)	100			
Wednesday		71	/u/ or /i/		
Thursday		100			
Friday		100			

Week 2: Drew was reported to be unwell over the weekend but was well enough to attend the Champion Centre on the Tuesday at which time he presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear. Drew and his mother completed the Ling 7-Sound Test each week day this week. Drew scored 100% each day except Wednesday when he scored 71% as he did not respond to the /u/ or /i/ sounds. Drew scored an overall PEACH score of 82.5% (90% in quiet, 75% in noise); a TEACH was not completed as Drew did not attend his early childhood centre this week due to illness.

Table 24. Drew's Results Week 3

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		82.5	
Tuesday	B high (right) B low (left)	100			
Wednesday		100			75
Thursday		86	/a/, /ɔ/		
Friday		100			

Week 3: Drew was reported to be slightly unwell with a mild cold this week. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother completed the Ling 7-Sound Test every day this week. Drew scored 100% on the Monday, Tuesday, Wednesday and Friday, and 86% on the Thursday. On the Thursday Drew did not respond to the /a/ or /ɔ/ sound.

Drew scored an overall PEACH score of 82.5% (85% in quiet, 80% in noise) and an overall TEACH score of 75% (70% in quiet, 81% in noise).

Table 25. Drew's Results Week 4

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		75	
Tuesday	B high (right) B low (left)	100			
Wednesday		100			86
Thursday		100			
Friday		100			

Week 4: Drew's general health was good. On the Tuesday, Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear. Drew and his mother completed the Ling 7-Sound Test each week day this week. Drew scored 100% on all days.

Drew scored an overall PEACH score of 75% (80% in quiet, 70% in noise) and an overall TEACH score of 86% (90% in quiet, 81% in noise).

Table 26. Drew's Results Week 5

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		85	
Tuesday	B high (right) B low (left)				
Wednesday		100			83
Thursday		100			
Friday		100			

Week 5: Drew's general health was good. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother completed the Ling 7-Sound Test each week day this week except Tuesday. Drew scored 100% each day the test was completed.

Drew scored an overall PEACH score of 85% (95% in quiet, 75% in noise) and an overall TEACH score of 83% (80% in quiet, 87.5% in noise).

Table 27. Drew's Results Week 6

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		82.5	
Tuesday	B high (right) B low (left)	100			
Wednesday		100			78
Thursday		100			
Friday		100			

Week 6: Drew's general health was good. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother completed the Ling 7-Sound Test each day this week during which Drew consistently scored 100%.

Drew scored an overall PEACH score of 82.5% (90% in quiet, 75% in noise) and an overall TEACH score of 78% (70% in quiet, 87.5% in noise).

Table 28. Drew's Results Week 7

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday				65	
Tuesday	Did not test				
Wednesday					Did not test
Thursday		100			
Friday		23 (did not complete test)			

Week 7: Drew's mother reported he had been sick with an upset stomach and cold this week. As a result, Drew did not attend the Champion Centre or his early childhood centre.

Therefore, tympanometry and the TEACH were not completed this week. Drew and his mother completed the Ling 7-Sound Test on Thursday and Friday this week, once he was feeling well. Drew scored 100% on the Thursday and 23% on the Friday. On the Friday Drew did not want to participate so his mother respected this and stopped testing after the first few sounds. Drew scored an overall PEACH score of 65% (65% in quiet, 65% in noise).

Table 29. Drew's Results Week 8

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday				80	
Tuesday	B high (right) B low (left)	100			
Wednesday		100			75
Thursday		100			
Friday		100			

Week 8: Drew's general health was good. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother completed the Ling 7-Sound Test each day this week except Monday during which Drew scored 100% each day the test was completed.

Drew scored an overall PEACH score of 80% (85% in quiet, 75% in noise) and an overall TEACH score of 75% (70% in quiet, 81% in noise).

Table 30. Drew's Results Week 9

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday		100		75	
Tuesday	Did not test	100			
Wednesday		100			80.6
Thursday		93	/a/ once		
Friday					

Week 9: Drew's mother reported he was unwell this week. Therefore, Drew did not attend the Champion Centre on Tuesday and tympanometry was not completed. Drew and his mother completed the Ling 7-Sound Test each day this week except Friday when Drew was particularly unwell and not willing to participate. Drew scored 100% on the Monday, Tuesday and Wednesday and 93% on the Thursday when he did not respond to the /a/ sound once.

Drew scored an overall PEACH score of 75% (80% in quiet, 70% in noise) and an overall TEACH score of 80.6% (75% in quiet, 87.5% in noise).

Table 31. Drew's Results Week 10

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)	TEACH Score (%)
Monday				62.5	
Tuesday	B high (right) B low (left)				
Wednesday					69.4
Thursday					
Friday					

Week 10: Drew's mother reported he was unwell with a virus. Drew presented with a type B high tympanogram in his right ear and a type B low tympanogram in his left ear on the Tuesday. Drew and his mother did not complete the Ling 7-Sound Test on the Monday because Drew was unwell. Drew scored an overall PEACH score of 62.5% (65% in quiet, 60% in noise) and an overall TEACH score of 69.4% (65% in quiet, 75% in noise).

3.3.4 Social validity

In the social validity interview, Drew's mother reported that prior to the study she was often concerned about Drew's hearing, particularly as she had no way of knowing if his grommets were patent or had extruded. Drew's mother reported that she was always confident to administer the Ling 7-Sound Test and it was often easy to engage Drew in the task provided he was feeling well because putting cars down the tunnel shoot was very motivating for him. Additionally, she reported that incorporating the Ling 7-Sound Test into their daily routine was often easy because it did not require much time and she found that having the score sheet on the bench and receiving the text message were useful in reminding her to do the Ling 7-Sound Test and PEACH. With regard to the usefulness of the Ling 7-Sound Test, Drew's mother reported that the test was useful to her when Drew had a cold because that is when she often suspects he may not be hearing well. However, Drew was often hearing better than she thought he would be which was useful for her to know. Drew's mother found the Ling 7-Sound Test more useful to monitor Drew's hearing than the PEACH questionnaire because the Ling 7-Sound Test results were instant, whereas the PEACH was about the past three days. Drew's mother reported that she was always confident in the results obtained from play audiometry and tympanometry, often confident in the results from the Ling 7-Sound Test and TEACH, and sometimes confident in the PEACH results.

3.3.5 Results summary – Case 3: Drew

Overall across the ten weeks Drew presented consistently with type B high tympanograms in his right ear and type B low tympanograms in his left ear. Drew's Ling 7-Sound Test scores were generally high and ranged between 23% and 100% (see Figure 13). There was one instance when he scored below 70%. Drew's overall PEACH scores ranged from 62.5% to 85% throughout the study (see Figure 14). Drew's overall TEACH scores ranged from 69.4% to 86% (see Figure 15). Drew's Ling 7-Sound Test results were on average higher than the results obtained from the questionnaires (see Figure 16).

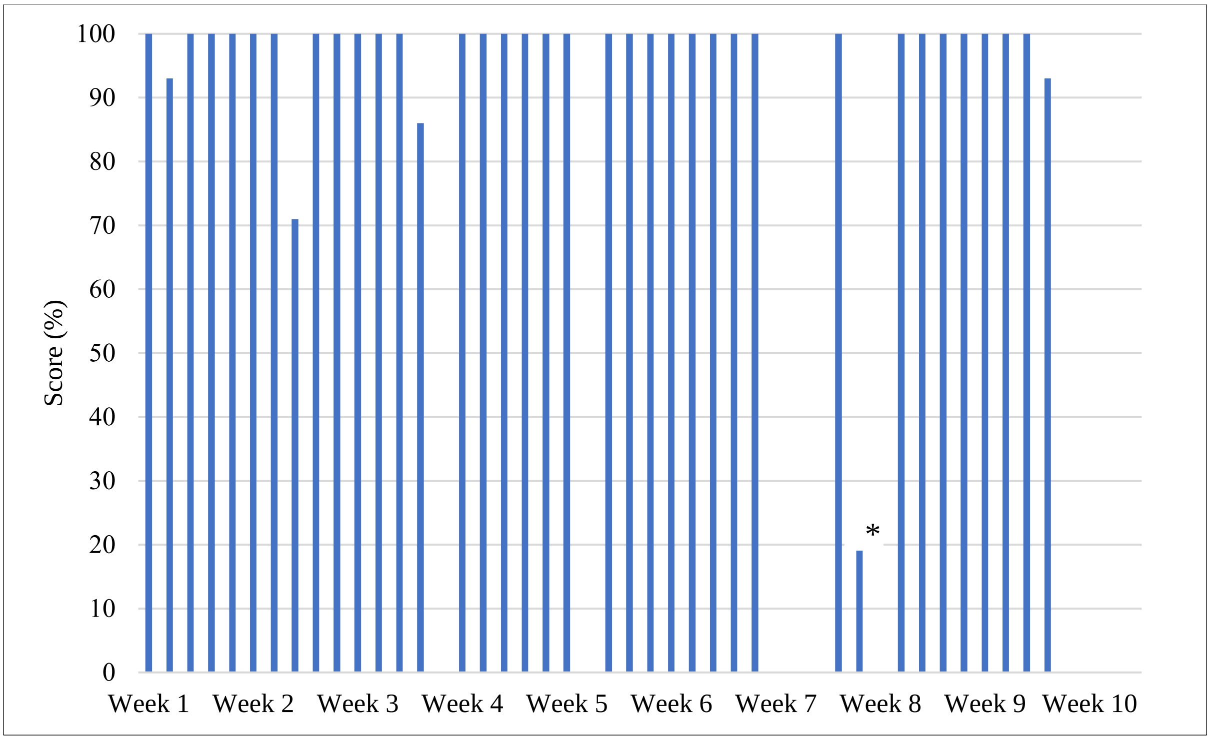

Figure 13. Drew's Ling 7-Sound Test results during the 10 week study

*Incomplete data

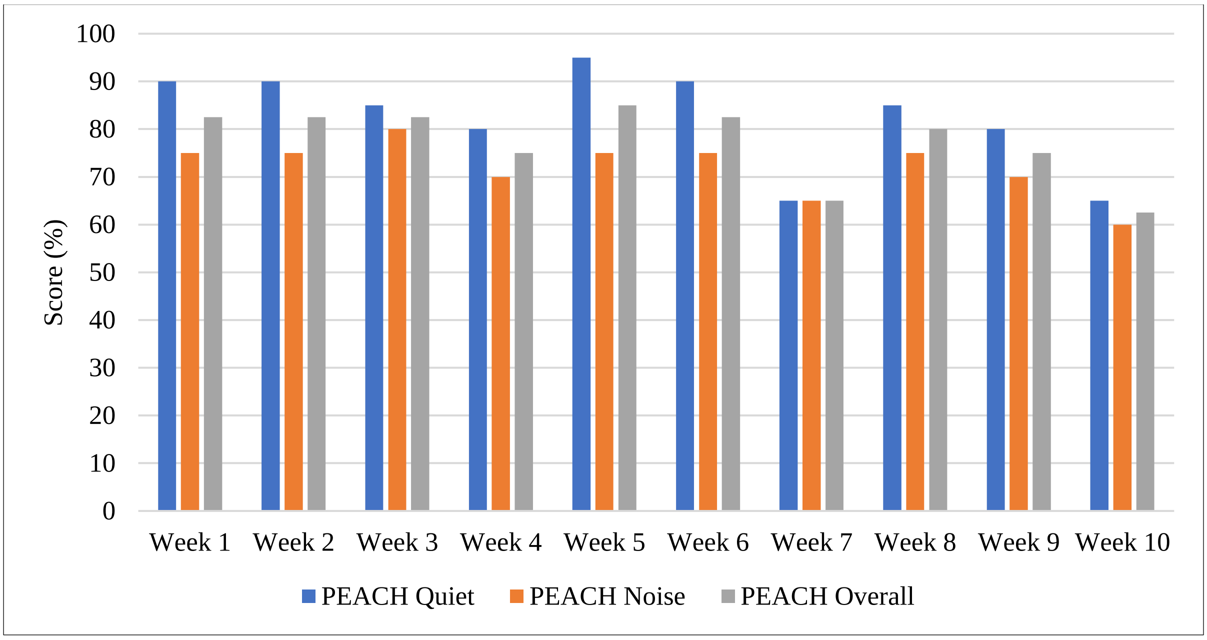

Figure 14. Drew's PEACH scores during the 10 week study

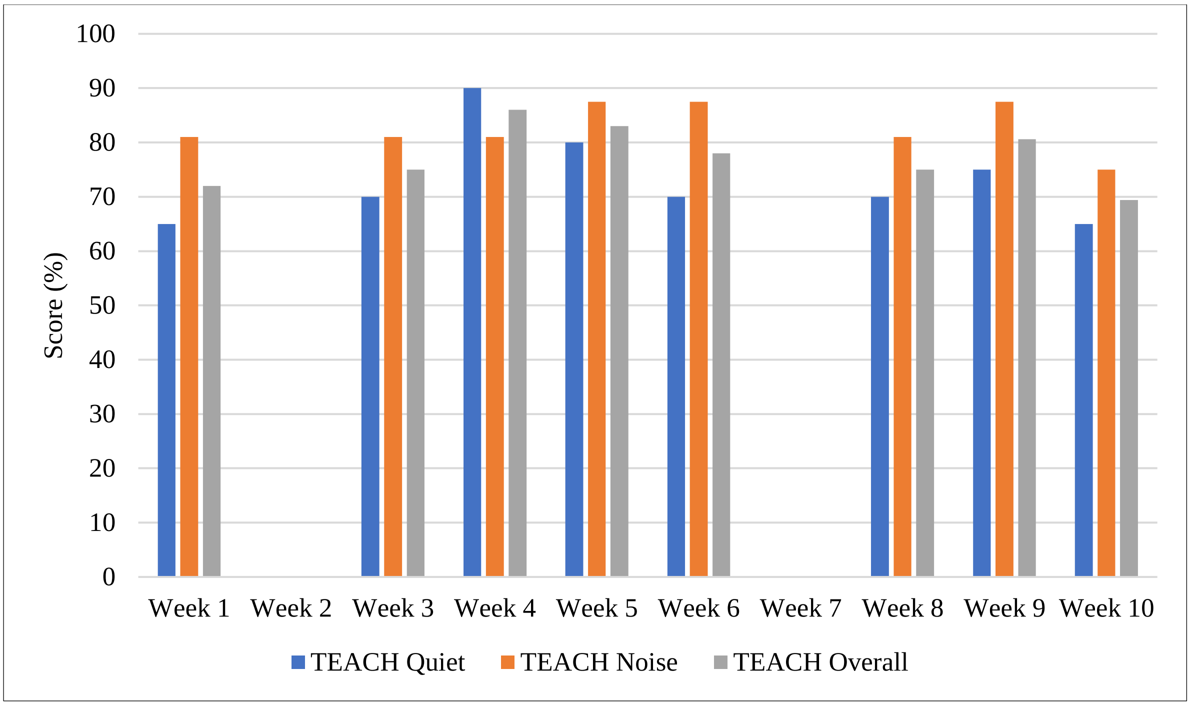

Figure 15. Drew's TEACH scores during the 10 week study

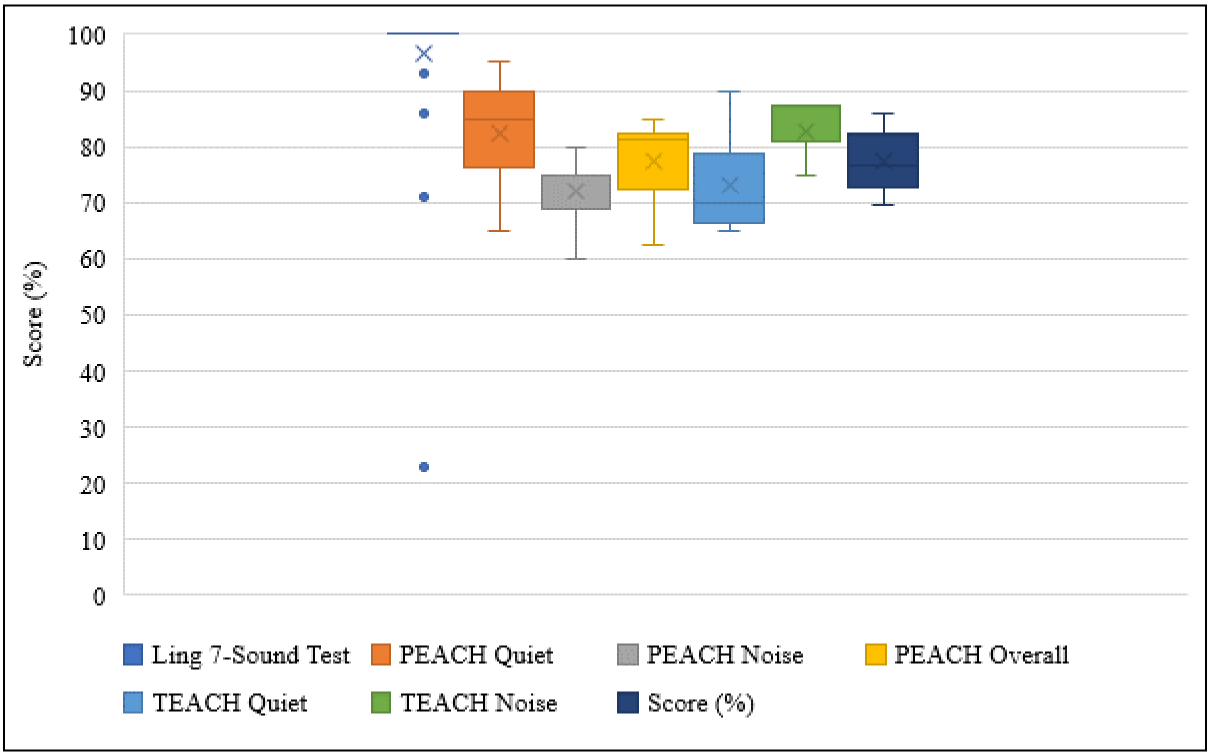

Figure 16. Drew's Ling 7-Sound Test, PEACH and TEACH scores during the 10 week study

3.4 Case study four: Rosie

Rosie was 4 years and 3 months old at the beginning of the study. Rosie lives with both parents and does not have any siblings. Rosie uses both aural/oral language (English, German and Japanese) and New Zealand sign language to communicate. As well as attending the Champion Centre one morning a week with her mother, Rosie attends an early childhood centre one day a week where she is supported by a Champion Centre early intervention educator. Unfortunately Rosie's early intervention educator was not able to participate in the current study. Therefore, results from the Ling 7-Sound Test, PEACH and social validity questionnaire and interview will be reported below.

3.4.1 Ear health and hearing history

Rosie was not born in New Zealand; as such there is no record of her Newborn Hearing Screening. However, since Rosie immigrated to New Zealand as an infant her hearing has been tested annually as part of the follow up pathway for children with Down syndrome. Rosie does not have a known history of otitis media with effusion and therefore has never had ventilation tubes inserted.

Rosie attended a hearing review appointment at the hospital one month before the study began. At this appointment, tympanometry revealed a type A shallow tympanogram consistent with in low middle ear admittance in the left ear and a type C tympanogram consistent with negative middle ear pressure in the right ear. Play audiometry revealed hearing thresholds within normal limits binaurally. Rosie's father reported no concerns about Rosie's hearing at this appointment.

3.4.2 Training

Rosie was compliant throughout the practice and enjoyed holding a plastic toy to her ear, waiting for the sound and posting the toy in a container when she heard the stimuli. Rosie's mother provided social reinforcement following Rosie’s correct responses. Rosie was able to complete the Ling 7-Sound Test with little instruction during this first practice.

3.4.3 Study

Table 32. Rosie’s Results Week 1

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		87.5
Tuesday	B low	29	/u/, /i/, /s/, /ʃ/	
Wednesday		29	/a/, /i/, /s/, /ʃ/	
Thursday		29	/u/, /a/, /i/, /s/	
Friday		29	/u/, /a/, /i/, /s/	

Week 1: Rosie was reported to be unwell with a cough this week. Rosie presented with type B low tympanograms binaurally on the Tuesday. It is possible that Rosie was developing otitis media with effusion at the time of her hearing review appointment, one month prior, when she presented with a type A shallow and a type C tympanogram. On the Tuesday the researcher also screened Rosie’s hearing at the Champion Centre and found Rosie had a mild to moderate hearing loss based on her air conduction thresholds. Bone conduction audiometry could not be completed due to the noise floor at the Champion Centre being too high. Rosie and her mother had completed the Ling 7-Sound Test on the Monday and Rosie had responded to all of the sounds. The researcher and Rosie’s mother decided that the Ling stimuli were too loud if she could still hear them when she had an identified hearing loss. Therefore, the intensity of the Ling stimuli was lowered from 45 dB to 40 dB. From Tuesday to Friday Rosie responded to 29% of the Ling sounds each day; she did not respond to the same sounds each day. On the Tuesday Rosie did not respond to the /u/, /i/, /s/ or /ʃ/ sounds, on the Wednesday she did not respond to the /a/, /i/, /s/ or /ʃ/ sounds, and on the Thursday and Friday she did not respond to the /u/, /a/, /i/ or /s/ sounds.

On the Monday, Rosie scored an overall PEACH score of 87.5% (90% in quiet, 85% in noise).

Table 33. Rosie's Results Week 2

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		97.5
Tuesday	B low	100		
Wednesday		86	/m/	
Thursday		100		
Friday		100		

Week 2: Rosie was reported to still have a slight cough this week, although she finished the course of antibiotics prescribed for this on Monday. Rosie presented with type B low tympanograms binaurally again on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day except Wednesday when she did not respond to the /m/ sound and scored 86%.

Rosie scored an overall PEACH score of 97.5% (100% in quiet, 95% in noise).

Table 34. Rosie's Results Week 3

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) A (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 3: Rosie's general health was good this week. Rosie presented with a type B low tympanogram in her right ear and a type A tympanogram in her left ear on the Tuesday.

Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 35. Rosie's Results Week 4

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		97.5
Tuesday	C	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 4: Rosie's general health was good this week. Rosie presented with type C tympanograms binaurally on the Tuesday. The researcher screened Rosie's hearing at the Champion Centre and found Rosie's hearing had improved, however, she still had a mild loss in her left ear. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise),

Table 36. Rosie's Results Week 5

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) A (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 5: Rosie's general health was good this week except for some hay fever. Rosie presented with a type B low tympanogram in her right ear and a type A tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day.

Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 37. Rosie's Results Week 6

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	C (right) A (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 6: Rosie's general health was good this week. Rosie presented with a type C tympanogram in her right ear and a type A tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 38. Rosie's Results Week 7

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) A (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 7: Rosie was reported to have hay fever and a cold this week. Rosie presented with a type B low tympanogram in her right ear and a type A tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored

100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 39. Rosie's Results Week 8

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) C (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 8: Rosie's general health was good this week. Rosie presented with a type B low tympanogram in her right ear and a type C tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 40. Rosie's Results Week 9

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) C (left)	100		
Wednesday		100		
Thursday		100		
Friday		100		

Week 9: Rosie's general health was good this week. Rosie presented with a type B low tympanogram in her right ear and a type C tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test each day. Rosie scored 100% every day. Rosie scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

Table 41. Rosie’s Results Week 10

Day	Tympanometry	Ling 7-Sound Test Score (%)	No response	PEACH Score (%)
Monday		100		100
Tuesday	B low (right) A (left)			
Wednesday				
Thursday				
Friday				

Week 10: Rosie’s general health was good this week. Rosie presented with a type B low tympanogram in her right ear and a type A tympanogram in her left ear on the Tuesday. Rosie and her mother completed the Ling 7-Sound Test on the Monday and Rosie scored 100%. Rosie also scored an overall PEACH score of 100% (100% in quiet, 100% in noise).

3.4.4 Social validity

In the social validity interview, Rosie’s mother reported that she did not have any concerns about Rosie’s hearing before the study because she reports Rosie’s hearing is very sensitive as she picks up very quiet sounds from long distances. With regard to the Ling 7-Sound Test, Rosie’s mother was able to incorporate it into their daily routine easily, although they did not necessarily complete the test at the same time each day and she reported the text message and record sheet were useful to remind her to complete both the Ling 7-Sound Test and PEACH. As with other parents, Rosie’s mother found the information she got from the Ling 7-Sound Test more useful than the information from the PEACH. Rosie’s mother reported that it was surprising to her that Rosie was not hearing well in the first week of the study, as Rosie was not demonstrating any behaviours such as touching her ears that might suggest she had otitis media with effusion. Rosie’s mother was equally confident in the results obtained from all of the measures used in the current study.

3.4.5 Results summary – Case 4

Overall across the ten weeks Rosie's tympanograms fluctuated. Except for during the first two weeks when Rosie had type B low tympanograms binaurally, Rosie had at least one ear that did not show any indication of middle ear fluid. Rosie's Ling 7-Sound Test scores ranged from 29% to 100% (see Figure17). Her lowest scores (29%) were obtained during the time she had tympanometry results that indicate fluid behind the eardrum and play audiometry results that revealed a mild-moderate hearing loss. Rosie's PEACH scores were consistently high throughout the study with overall scores ranging from 87.5% to 100% (see Figure 18). Rosie's scores in all of the measures were high throughout the study, except for during the first two weeks (see Figure 19).

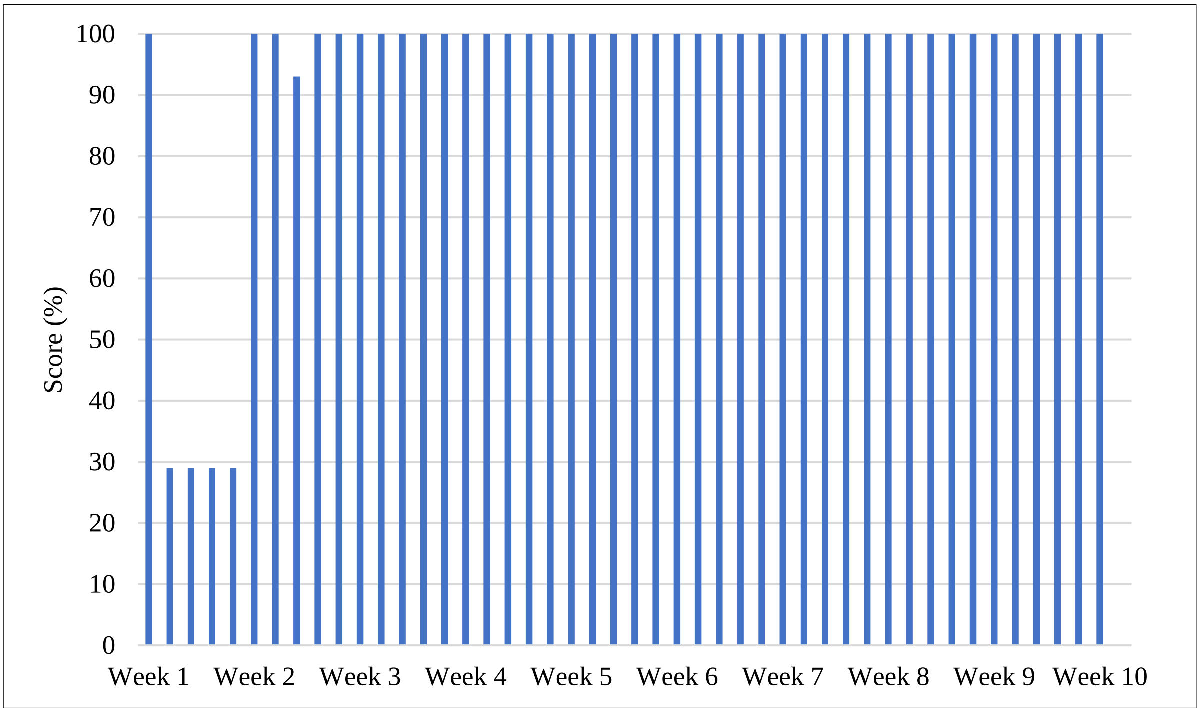

Figure 17. Rosie's Ling 7-Sound Test results during the 10 week study

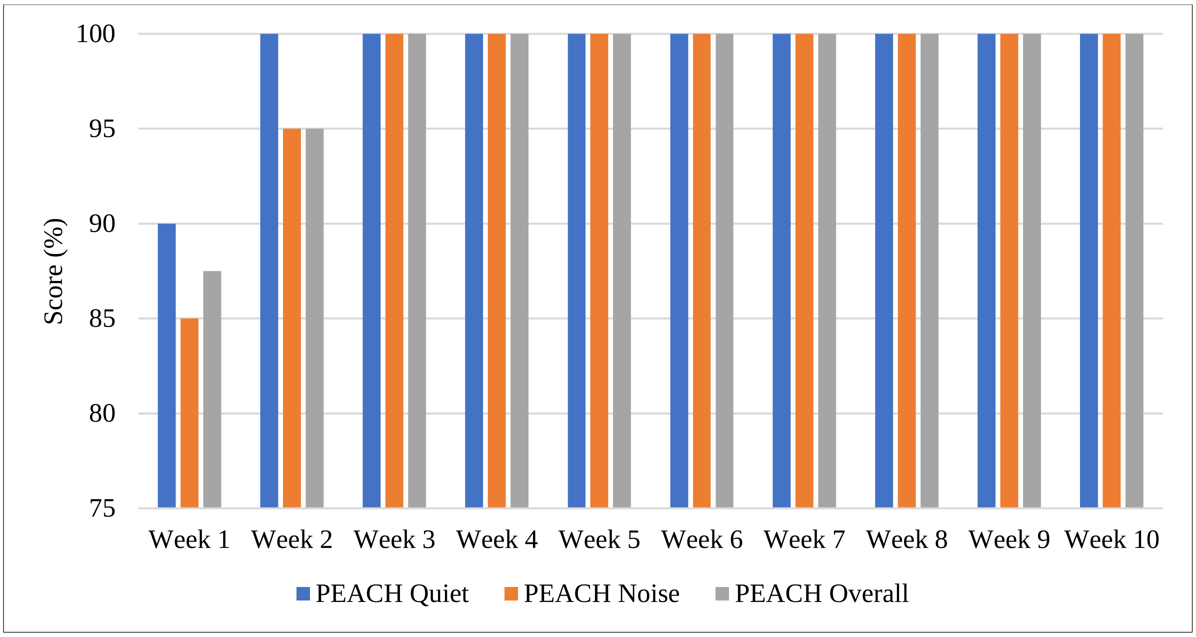

Figure 18. Rosie's PEACH scores during the 10 week study

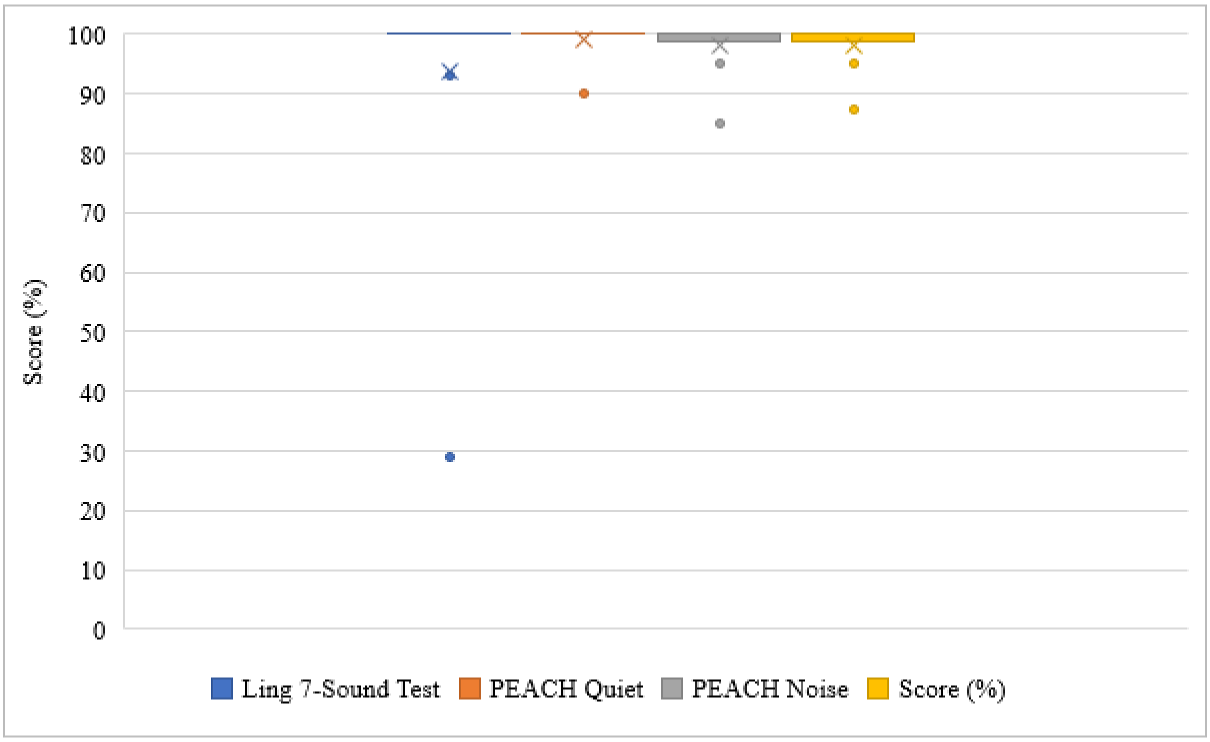

Figure 19. Rosie's Ling 7-Sound Test and PEACH results during the 10 week study

3.5 Early intervention team leader social validity interview results

The early intervention team leader is a speech therapist who works with the participating children and parents once a week at the Champion Centre. Her role involves overseeing all the therapists who work with the children at the Champion Centre. As such she has an in depth holistic understanding of each child and their families.

In the social validity interview the early intervention team leader reported that prior to the study she was often concerned about the hearing of children in her programme because the research reports that children with Down syndrome are at risk of hearing loss and there have been instances when parents have informed her that their child has an ear infection. It was also reported by the early intervention team leader that it is always important for parents, therapists and early intervention educators to know about the children's hearing status because hearing is the basis for communication and it underpins the children's wellbeing. Additionally, she reported that when the team was aware that a child was not hearing well or had an ear infection, they could be more flexible with the child because the therapists understood why the child's behaviour may have changed. With regard to the Ling 7-Sound Test, the early intervention team leader reported that it was always a practical tool for parents to use to monitor their children's hearing as it was easy and straightforward to complete. With regard to the PEACH and TEACH questionnaires, the early intervention team leader reported that the informal observations of the therapists' may be more accurate than those of the parents'.

3.6 Overall summary of results

3.6.1 Ear health and hearing history

Prior to the study, three of the four children in the present study had required at least one set of grommets as a result of persistent otitis media with effusion. No children in the current study had permanent sensorineural hearing loss. Despite these results being available, it was reported that behavioural testing in the past for two of the children had not been as successful as was hoped.

3.6.2 Training

For each of the families the training phase of the current study was completed in one session taking no longer than 45 minutes. By this time the parents were confident to administer the Ling 7-Sound Test to their child, complete a PEACH questionnaire and the children had been successfully conditioned to respond appropriately to the Ling 7-Sound Test stimuli.

3.6.3 Study

Throughout the current study all children presented with tympanograms indicative of normal middle ear functioning in at least one ear, except for Rosie who had type B low tympanograms binaurally during the first two weeks of the study. All families successfully incorporated the Ling 7-Sound Test into their daily routines with all families completing the test at least 3 times a week except during exceptional circumstances when the child was unwell. Isobel's family completed the Ling 7-Sound Test on 22 of the 46 days (48%), Drew's family on 38 (83%) of the days and both Sam and Rosie's families on all 46 days (100%). Because it was found that lower intensity Ling stimuli were sensitive to changes in hearing thresholds for Rosie, the researcher trialled the use of the softer sounds with the other three children. However, the quieter stimuli were not intense enough to exhibit a consistent response. Therefore, the intensity of the stimuli remained at 45 dB A for the other three children. Overall, the children obtained high scores in the Ling 7-Sound Test when they had at least one ear with a tympanogram indicating normal middle ear functioning. When a child obtained a lower score than would be indicated from tympanometry, this was always accompanied by parent report of poor health or difficulty maintaining attention.

Similarly with the PEACH questionnaires, all parents completed these on the majority of the weeks, with the children receiving similar scores week to week unless they were unwell. All three early intervention educators who were able to participate in the current study completed the TEACH questionnaire each week that the child attended preschool. Comparison of the PEACH and TEACH scores indicated that one child achieved lower TEACH than PEACH scores, while the other two children had close agreement between the two scores.

3.6.4 Social validity

All four parents and the early intervention team leader participated in a social validity questionnaire and semi-structured interview exploring the responses in the questionnaire. When asked if they received adequate support from the researcher during training and throughout the study, all parents responded with “always”. Two parents reported that they were “always” confident to administer the Ling 7-Sound Test; one reported they “often” were. All four parents reported that it was” often” easy to incorporate the Ling 7-Sound Test into their daily routine, with one parent reporting that the information from this test was “sometimes” useful, one reporting it was “often” useful and two reporting it was “always” useful (see Table 43 for a summary of the social validity findings). The same result was reported for the usefulness of the information from the PEACH. Overall, parents reported most confidence in the audiometry and tympanometry results with slightly lower confidence in the Ling 7-Sound Test followed by the PEACH then the TEACH (see Table 42).

Table 42. Parent reported confidence in the measures used in the current study

Measure	Always	Often	Sometimes	Rarely	Never	N/A
Audiometry	4					
Tympanometry	4					
Ling 7-Sound Test	3	1				
PEACH	3		1			
TEACH	2	1				1

Table 43. Parent response in the social validity questionnaire

Question	Always	Often	Sometimes	Rarely	Never
I was confident to administer the Ling 7-Sound Test	3	1			
It was easy to engage my child in the Ling 7-Sound Test	2	1	1		
It was easy to incorporate the Ling 7-Sound Test into our family's routine		4			
The record sheet was useful in reminding me to complete the Ling 7-Sound Test	1	1	2		
The text message was useful in reminding me to complete the Ling 7-Sound Test	3		1		
The Ling 7-Sound Test was useful as a hearing monitoring device	4				

Overall, all four parents had some concern about their child's hearing and successfully incorporated the Ling 7-Sound Test into their family's routine during the 10 week study period. The parents reported that the use of this test provided them with information about their child's hearing that was useful to them.

CHAPTER 4 DISCUSSION

4.1 Introduction

The current study aimed to investigate parent use of the Ling 7-Sound Test to monitor the hearing of young children with Down syndrome. The study used tympanometry, the Ling 7-Sound Test, Parents' Evaluation of Aural/ Oral performance in Children (PEACH), Teachers' Evaluation of Aural/ Oral performance in Children (TEACH) and a social validity questionnaire and semi-structured interview to collect both quantitative and qualitative data to investigate four research questions:

1. What is the extent to which the children with Down syndrome experience fluctuating conductive hearing loss as a result of otitis media with effusion?
2. What are the experiences of parents when administering the Ling 7-Sound Test?
3. What is the relationship between tympanometry, play audiometry, the Ling 7-Sound Test, the PEACH, TEACH and social validity measures?
4. What is the extent to which the Ling 7-Sound Test, PEACH and TEACH are sensitive to conductive hearing loss?

4.2 What is the extent to which the children with Down syndrome experience fluctuating conductive hearing loss as a result of otitis media with effusion?

Three of the four children in the study had a history of grommet surgery used to manage chronic otitis media with effusion. This is consistent with the findings described by Kreicher et al. (2018) who found 75.4% of the children with Down syndrome had experienced chronic otitis media with effusion. The one child who did not have a history of grommet surgery was the only child who presented with type B low tympanograms, consistent with middle ear fluid, for the first two weeks of the study. Two weeks prior to the study commencing, her hearing was assessed, during which she had a type As and type C tympanograms. Therefore it can be concluded that she experienced otitis media with effusion for 2-4 weeks. Fortunately for the hearing health of the children, the incidence of fluctuating conductive hearing loss as a result of otitis media with effusion was this low during the study. It is likely indicative of effective medical management of the hearing health of these children. A high rate of normal hearing was also noted in a previous study by Shott et al. (2001) when children with Down syndrome were seen for audiologic evaluation every 3 to 6 months. It is possible that if the current study assessed the children over a longer period of time, or included only children who did not have patent grommets, that the incidence of bilateral type B low tympanograms would be higher. Despite the reported difficulty of obtaining complete audiological records of two of the children in the current study, they both had received grommet surgery in the past. As N. Thompson and Yoshinaga-Itano (2014) reported, there can be challenges associated with formal hearing assessments of children with developmental disabilities, usually due to the assessment environment and requirements not being matched to the child's ability, and the novel environment such assessments take place in being unfamiliar and unpredictable. The result of this mismatch is that parents and professionals working with children with Down syndrome may not have a complete understanding of the child's hearing, which may have negative implications if more information is required for the employment of active management strategies. This difficulty in formally assessing the children's hearing along with the invisible nature of hearing loss means that parents are often unsure if their child is hearing well or not (Cole & Flexer, 2015; Marriage et al., 2017). Similarly, even for children who have grommets *in situ*, parents may not be able to determine if the grommets are patent, occluded or extruded. Deferral of treatment for hearing loss may exacerbate the delays in speech and language development that children with Down syndrome are at risk for (Roberts et al., 2007). This highlights the importance of thorough and timely diagnosis of hearing loss in this population.

4.3 What are the experiences of parents when administering the Ling 7-Sound Test?

Overall, parents were able to successfully administer the Ling 7-Sound Test to their child as part of their daily routine, and reported value in doing so. All four parents reported that they were “often” or “always” confident to administer the Ling 7-Sound Test, and that it was “often” easy to incorporate the Ling 7-Sound Test into their family’s routine.

There are several factors that may have contributed to these findings, including the family centred approach, the audiology presence within the multi-disciplinary team at the early intervention centre the families attended and parent perceived need to monitor their child’s hearing.

Family centred practice is an approach to working with families that has sound rationale and many benefits (Espe-Sherwindt, 2008). Within this service delivery model, professionals share responsibility and work collaboratively with parents, enhance family functioning and provide intervention that is individualised and strengths focused (Trivette, Dunst, & Sandall, 2000). Because the most essential context to a child is their family/whānau, part of the training for the Ling 7-Sound Test was carried out within the home environment. Completing training in this setting allowed for families to feel supported in making decisions about how they could use the Ling 7-Sound Test in a way that aligned with the routine and values of their family. Aspects of the Ling 7-Sound Test also allowed flexibility. As such, parents could choose the time, environment, frequency of administration (although encouraged to complete daily) and the activity within which the Ling 7-Sound Test would be completed. As parents learned to integrate the new task of completing the Ling 7-Sound Test into their routines, a relationship of dignity was fostered and the values and choices of each family were respected. Family centred practice has many documented benefits related to family self-efficacy, wellbeing, confidence, competence, functioning and child behaviour and may have contributed to the results in parent ability to complete the Ling 7-Sound Test (Trivette, Dunst, & Hamby, 2010).

Another factor that may have contributed to the results was having an audiology presence within the multi-disciplinary team at the early intervention centre the families attended. Because the present study took place within the context of a multi-disciplinary, family centred early intervention centre, the researcher could discuss the participants' hearing with a range of professionals to gain a holistic understanding of each child's health and development. As with the context of the present study, it is common for professionals from a wide range of disciplines to work with young children with Down syndrome due to the complexity and variety of presentations associated with the syndrome (Alliston, 2007). While early intervention is not necessarily delivered in a multi-disciplinary service model, working as a team to provide family-centred care to young children with Down syndrome can have many advantages. Clinicians working with the children in the study could come together and share their individual expertise and observations with each other which enhanced all team members' holistic understanding of the child they were supporting (Madell & Flexer, 2014). Sharing knowledge across disciplines can expand team members' knowledge base and shed light on the impact a hearing loss may have across activities, people and settings (Ruhstaller, Roe, Thürlimann, & Nicoll, 2006). Such advantages as an improved holistic understanding of the child and continuing education are reliant on professionals and families being united in their philosophy and regular, effective communication with each other and were apparent in the current study (McNamara & Richard, 2012). For example, the researcher was available at the centre each week during the 10 week study so that families could share their experiences and access support and education when they needed it. Additionally, the researcher could share findings from the tympanometry, Ling 7-Sound Test, PEACH and TEACH questionnaires with the multi-disciplinary team working with each child and similarly, other professionals could share their observations of the children with the team and researcher resulting in a holistic understanding of each child.

All of the families in the study incorporated the Ling 7-Sound Test into their daily routines. Two families completed the Ling 7-Sound Test 5 days a week throughout the study, one family approximately four times a week and one 2-3 times a week. Parents were encouraged to complete the Ling 7-Sound Test every week day during the study. However, it was also explained that there may be times when the Ling 7-Sound Test was not a priority for the family and that that would be respected. Parents of children with hearing loss are frequently encouraged by their audiologist to complete the Ling sound test as a biological device check each day (Smiley, Martin, & Lance, 2004). Therefore, it was decided that the researcher would ask for the same frequency of administration in the current study. The results indicate that administering the Ling 7-Sound test 5 times a week was reasonable for two of the four families. It is possible that even the two families who completed the Ling 7-Sound Test each week day during the study would decrease the frequency of administration if they were required to complete the test over a longer period of time than that of the current study. This is because although they may enjoy the activity itself, the intrinsic motivation (Gagné & Deci, 2005) to complete the test may lower when other priorities arise. Additionally, the extrinsic motivation (Gagné & Deci, 2005) may lower without the group setting, presence of the researcher to encourage participation, if the parent does not perceive that their child has a hearing loss and if their child does not have a device that needs to be checked. Therefore, the parents may see fit that a decreased frequency of administration is sufficient to monitor their child's hearing or perhaps that it is useful only on days when they hypothesise that their child has a hearing loss.

The frequency of participation in the current study may be a result of parents' concerns of speech and language delays, prior challenges with formal audiological assessment, and the incidence of audiology appointments that children with Down syndrome are offered. Firstly, parents of children with Down syndrome have been reported to be more concerned about otitis media with effusion because of the negative impact it may have on speech and language development rather than to hearing itself (Fortnum et al., 2014). Therefore, the level of participation in the current study may be related to the level of concern each family had in terms of their child's speech and language development and the extent to which they attribute this delay to fluctuating hearing. Secondly, it is documented in the literature that audiologists have experienced difficulty in formally assessing the hearing of young children with developmental disabilities (N. Thompson & Yoshinaga-Itano, 2014). It was reported in two of the children's audiology notes that prior audiological assessments had not yielded complete results due to difficulties maintaining the child's attention and the children being averse to wearing transducers. This prior difficulty that has been experienced may contribute to the interest of parents in the current study to an alternative method to inform them about their child's hearing. Lastly, in New Zealand, young children with Down syndrome are typically seen for audiological evaluation once each year (Ministry of Health, 2016). It is possible that the children will experience fluctuation in their hearing within this one year period. Therefore, an increased frequency of hearing assessment may be valued by parents to provide more frequent information.

In summary, parent concern regarding the relationship between otitis media with effusion and speech and language delays, prior difficulty with formal hearing assessments, having up to one year between assessments as well as a small sample size of children with heavily invested parents all may have contributed to the Ling 7-Sound Test being valued as a method to monitor the hearing of their young children with Down syndrome.

4.3.1 Early intervention team leader

The results from the early intervention team leader indicated that the children's hearing is a concern and that having up to date information about each child's hearing is important for her practice as a team leader and speech therapist. The early intervention team leader reported that the informal observations of the therapists' may be more accurate than those of the parents' because the therapists only see the children once a week and often place high demands on the children to attend and respond to verbal information so a change in listening behaviour may be more apparent to the therapists.

There are benefits to therapists being regularly informed of the children's hearing status and having audiology services on site at the early intervention centre. Such benefits include supporting the multi-disciplinary team to broaden their holistic understanding of each child which could result in adaptations to practice such as the use of communication strategies to ensure each child is given the support they need to listen.

4.4 What is the relationship between tympanometry, play audiometry, the Ling 7-Sound Test, the PEACH, TEACH and social validity measures?

4.4.1 Isobel

Isobel presented with type B high tympanograms binaurally throughout the study indicative of a high ear canal volume and consistent with patent grommets or tympanic membrane perforations. Due to this result and because Isobel had no history of sensorineural hearing loss, it is reasonable to conclude that Isobel did not experience hearing loss during the study except potentially monaurally in week 7 and 8 when one of her ears was discharging.

Overall, Isobel's Ling 7-Sound Test results ranged from 71% to 100%. Isobel's lowest score obtained during the time she had an ear infection. During this time she also scored 100%. This fluctuation in her Ling 7-Sound Test scores may be explained by other stimuli in her environment being more alerting to her. Isobel's early intervention educator reported that Isobel is visually active and can lose focus on auditory information if the environment is visually busy. This observation may indicate that other stimuli may be more alerting than auditory stimuli to Isobel. Therefore, Isobel not attending to auditory stimuli may be due to factors other than hearing loss.

Isobel's TEACH scores ranged from 28-58%. In week two of the study, Isobel's TEACH score was 55.5%. Isobel's early intervention educator commented that the TEACH evaluation may not be reflective of her performance overall because Isobel celebrated her fifth birthday at her early childhood centre that week. As such Isobel had lots of family members in attendance and was very excited, therefore not demonstrating her typical listening abilities. Isobel's score wasn't particularly low as a result of this change in routine. During week four of the study, Isobel's TEACH score was 28% which was her lowest TEACH score during the study. Isobel's early intervention educator reported that Isobel had started a new early childhood centre and she was quieter than usual, which may be reflective of the increased listening demands in a novel environment with increased stimulation.

Isobel's Ling 7-Sound Test scores were on average higher than the scores obtained through the questionnaires. The higher scores may be a result of the Ling 7-Sound Test occurring within the most controlled environment of the three assessments because it was completed at a time of day when the child was at their best and ready to pay attention, the expectations were known to the child, and the stimuli and motor task were minimally demanding.

When compared to her PEACH scores which ranged from 67-75%, more variability was present in her TEACH scores, although the scores obtained during week 4 may well be a reflection of the new preschool environment rather than a change in her listening ability.

Both Isobel's TEACH and PEACH scores in quiet were higher than her TEACH and PEACH scores in noise. This result may be a reflection of the increased challenges of listening in background noise compared to a quiet environment. Similarly, Isobel's TEACH scores were consistently lower than her PEACH scores, which may be reflective of a more challenging listening environment at preschool than at home.

Overall, Isobel did not present with tympanometry results consistent with otitis media with effusion during the study. The above results demonstrate the variability in Isobel's listening behaviour during a time when it is unlikely that her hearing thresholds fluctuated. This variability highlights the importance of having a holistic understanding of the child when considering their hearing. It was apparent that the environment and changes in health resulted in fluctuations in Ling 7-Sound Test performance and both parent and teacher observations of Isobel's hearing.

4.4.2 Sam

During the study Sam presented consistently with type B high tympanograms in his right ear, consistent with a patent grommet *in situ*. In his left ear, Sam's tympanograms varied. Due to Sam's tympanometry and prior audiometry results, it is reasonable to conclude that Sam did not experience hearing loss during the study except potentially monaurally in week 1 when he had a type B low tympanogram.

Sam's Ling 7-Sound Test scores ranged between 43% and 100%. There were 5 instances when he scored below 70%. Two of his lowest scores were obtained on the first two days of the study. During this time Sam had a type B low tympanogram in one of his ears, so it is possible that he was not hearing optimally. Alternatively, Sam may have been getting used to the task during this time. The next two instances were when the test was completed in a novel environment and the fifth instance was during a time when Sam was unwell. Interestingly, Sam's mother made the observation that Sam had the most difficulty responding to the /s/ and /ʃ/ sounds, which she hypothesised may be because the /ʃ/ sound in particular is associated with being told to be quiet. Perhaps increasing positive social reinforcement following the behavioural response would have resulted in Sam responding to this sound more frequently.

Additionally, teaching the children that the sounds are associated with picture cards may avoid such a response. For example, teaching the child that the /ʃ/ sound is associated with the card showing a baby being soothed by their mother.

Sam's overall PEACH scores ranged from 62.5% to 83% throughout the study. Sam's PEACH scores in quiet were consistently higher than his scores in noise, reflecting the increased challenges of listening in a noisy environment, even when his hearing is within normal limits. Similarly to his Ling 7-Sound Test scores, the outlier from the overall PEACH scores was in week 7 when Sam had been unwell.

As with Isobel, more variability was present in Sam's TEACH scores which ranged from 55-100% in the quiet category. Overall there was close agreement between Sam's PEACH and TEACH scores. Additionally, Sam's TEACH scores in quiet were similar, albeit slightly higher than his PEACH scores in quiet which may be due to both assessments being completed in a quiet home environment, rather than an early childhood centre.

Similarly to Isobel, Sam did not present with tympanometry results consistent with binaural otitis media with effusion during the study. Generally, when Sam was unwell, all of his results were lower than they had been when he was well, indicating all measures were sensitive to changes in his health. The above results demonstrate the variability in Sam's listening behaviour during a time when he constantly had type B high tympanograms in at least one ear. This variability highlights the importance of having a holistic understanding of the child when considering their hearing, as it was apparent that a novel task, the environment and Sam's health resulted in fluctuations in his Ling 7-Sound Test performance and both parent and teacher observations of Sam's hearing.

4.4.3 Drew

Based on Drew's tympanometry results and prior audiological results, it is reasonable to conclude that he did not experience any binaural hearing loss during the study. During the study Drew presented with type B high tympanograms in his right ear, consistent with a patent grommet *in situ,* and type B low tympanograms in his left ear, consistent with middle ear fluid each week. Drew's Ling 7-Sound Test scores ranged between 23% and 100%. On the Wednesday of week two Drew scored 71% because he did not respond to the /u/ or /i/ sounds. These two sounds were the last to be presented on the Wednesday by which time Drew no longer wanted to participate. The one instance when Drew scored below 70% was in week 7 when the test was discontinued because Drew was unwell and did not want to participate resulting in the 23% score. Turning to the PEACH scores, Drew's overall scores in this assessment ranged from 62.5% to 85% throughout the study. Drew's PEACH scores in quiet were consistently higher than his scores in noise, reflecting the increased challenges of listening in a noisy environment. Similarly to his Ling 7-Sound Test scores, the lowest overall PEACH score was in week 7 when Drew had been unwell. Drew's overall TEACH scores ranged from 69.4% to 86%, a similar range to his overall PEACH scores. Drew's TEACH scores in noise were consistently higher than his TEACH scores in quiet. This is difficult to interpret, because based on Drew's tympanometry results, it is more likely that he would experience more listening difficulty in noise. While this may be a difference in perception of the teacher, it could also be that increased auditory stimulation is more engaging for Drew in the preschool environment. As the study progressed, closer agreement between Drew's PEACH and TEACH scores was achieved, possibly due to Drew's mother and early intervention educator becoming more consistent in their reflections of Drew's listening. In contrast to Drew's TEACH scores in noise, his TEACH scores in quiet were on average lower. Despite periods of ill health in weeks two, three, seven, and nine, Drew, his mother and early intervention teacher were able to complete the measures at least some days during these weeks. During this time Drew achieved high scores (except for week seven) in the Ling 7-Sound Test, PEACH and TEACH, demonstrating that poor health is not necessarily an indicator of poor hearing. Despite consistently presenting with a type B low tympanogram in his left ear, the above results show that Drew was able to achieve high scores in all of the measures. Advantages of binaural hearing such as better understanding of speech in noise and sound localisation (Avan, Giraudet, & Büki, 2015) would not be observed in the Ling 7-Sound Test because the task was binaural and completed in a quiet environment at 180 degrees azimuth. Drew's PEACH scores in noise were consistently lower than his PEACH scores in quiet, which may be a reflection of unilateral otitis media with effusion. However, it would be expected that a similar pattern would be observed in Drew's TEACH scores in quiet when compared to his TEACH scores in noise, when in fact the opposite was reported.

The above results demonstrate the variability present in Drew's scores when his tympanometry results were constant throughout the study. Variability is present across days, settings, examiners and listening environments.

4.4.4 Rosie

During the study Rosie's tympanograms fluctuated. Other than in the first two weeks (when Rosie had type B low tympanograms binaurally), Rosie had at least one ear that did not show any indication of middle ear fluid. When play audiometry was completed in the first week and revealed a mild to moderate hearing loss, Rosie's mother reported that this result did not match her informal observations of Rosie's hearing or her reflections she reported in the PEACH questionnaire. Rosie's mother had reported that Rosie appears to have very sensitive hearing and often pays attention to very quiet sounds, sometimes from a long distance. On the first two days of the study Rosie responded to all 7 Ling sounds at 45 dB A despite having a mild to moderate hearing loss. Therefore, it was determined that 40 dB A stimuli would be used with Rosie for the remainder of the study. Rosie's reported sensitive hearing may explain why a lower intensity of Ling sounds was more appropriate and sensitive to changes in her middle ear status. This highlights the issue that one intensity level may not be appropriate for use with all children due to differences in hearing sensitivity and attention. In future, a reasonable sensation level could be determined for each child given air conduction thresholds obtained in the past. Rosie's Ling 7-Sound Test scores ranged from 29% to 100%. Her lowest scores (29%) were obtained during the time she had tympanometry results that indicate fluid behind the eardrum and play audiometry results that revealed a mild-moderate hearing loss. Rosie's PEACH scores were consistently high, in agreement with her Ling 7-Sound Test scores in all weeks except week 1.

4.4.5 Summary

Three of the four children in the study had a history of grommets while one child experienced otitis media with effusion during the study. The historical high incidence of otitis media with effusion and parental concern about their child's hearing may have contributed to parents successfully administrating the Ling 7-Sound Test to their child as part of their daily routine, and the reported value in doing so. Parents were able to reflect and report as to why fluctuations in scores occurred. The children generally scored highly across all measures when they were well and paying attention to the task. Of the three measures, the children all obtained the highest scores in the Ling 7-Sound Test. Comparison of the PEACH and TEACH scores indicated one child scored achieved lower TEACH than PEACH scores, while the other two children had close agreement between the two scores. This result may be due to the similarity or lack thereof between the home environments the PEACH and TEACH were completed in.

4.5 What is the extent to which the Ling 7-Sound Test, PEACH and TEACH are sensitive to conductive hearing loss?

The Ling 7-Sound Test at a 40 dB A intensity produced results consistent to tympanometry and play audiometry in one child during the study. Rosie presented with bilateral tympanograms consistent with otitis media with effusion during two weeks of the study. For this child, the Ling 7-Sound Test stimuli at 45 dB A were not sensitive to conductive hearing loss. However, when the intensity of the stimuli was lowered to 40 dB A, her Ling 7-Sound Test results dropped and showed a change in performance. Rosie's PEACH results did not show any indication of hearing loss in the first two weeks, and unfortunately Rosie's early intervention educator was not able to participate in the study so it cannot be concluded as to whether or not a change in hearing was apparent in the early childhood centre during this time. The researcher trialled the use of the softer sounds with the other three children, but found the stimuli was not intense enough to exhibit a consistent response. Therefore, the intensity of the stimuli remained at 45 dB A for the other three children.

Three of the four children presented with fluctuation in their Ling 7-Sound Test, PEACH and TEACH results. Such fluctuation occurred without binaural change in tympanometry results, so it is unlikely that their hearing changed binaurally. The fluctuation that occurred is an important consideration because listening is dependent on both the integrity of the auditory system and higher order cognitive and linguistic skills (Larsby, Hällgren, Lyxell, & Arlinger, 2005). Therefore, it may be beneficial to counsel parents to use communication strategies even when hearing thresholds may be within normal limits (Rosenfeld et al., 2016). Certain environments, such as those that are noisy, place a higher demand on children and in such environments simple adaptations to the communication partner's communication could result in greater engagement from the child and less frustration and fatigue (Hicks & Tharpe, 2002). It is worth noting that even tympanometry is not 100% sensitive to otitis media with effusion. For example, Takata et al. (2003) reported that 81% of the children with otitis media with effusion tested in their study presented with Type B tympanograms. Therefore, pure tone audiometry with both air conduction and bone conduction transducers needs to be completed to determine if a conductive hearing loss is present, even if tympanometry yields a type A or C tympanogram.

4.6 Clinical implications

The findings of the current study support the following approach to monitoring the hearing of young children with Down syndrome.

- Active management and monitoring because the delay in speech and language development is reported to have an impact beyond the time of resolution of the hearing loss (Laws & Hall, 2014).
- Inclusion of an audiologist in the multi-disciplinary early intervention team
- Increased frequency of audiological evaluations or screenings to provide parents and therapists with current information about the child's ear health and hearing
- Completing audiological evaluations at the early intervention centre or child's home. This is reliant on access to portable audiological equipment such as audiometers and tympanometers
- Educating parents of children at risk of otitis media with effusion about ear health, hearing, listening and hearing loss
- Empowering parents and teachers to monitor a child's hearing
- Placing value in having a holistic understanding of each child's hearing. As such, valuing reflections from all people who interact regularly with the child
- Utilising hearing and listening assessments, such as the Ling 7-Sound Test, PEACH, TEACH and tympanometry, that allow for flexibility so that the child both enjoys the experience and can perform at their best. This includes assessments that can be completed in environments in which the child is comfortable, with people who the child has a relationship with, using stimuli and response methods that are engaging and familiar to the child

- Employing a strengths-focussed approach to audiological evaluation. For example, using visual schedules displaying sequences for children with Down syndrome
- Recommending low cost communication strategies that are easily employed by parents to mitigate the negative effects of otitis media with effusion.

4.7 Limitations

The current study aimed to investigate parent use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome. However, the extent to which this was achieved was limited by the low incidence of hearing fluctuation in the participants. Although not designed to be a population study, the current study would have benefitted from an increased number of participants and those who did not have grommets and were therefore less likely to experience otitis media with effusion. While the small number of participants and case study design provided a depth of understanding, findings would not have portrayed the full range of variability likely to be present and cannot be generalised to all young children with Down syndrome (Yin, 2017). Additionally, it would have been beneficial to determine the optimal stimulus level for each participant prior to the study commencing.

A limitation of the current study was that parents were not blinded to the results obtained from the weekly tympanometry. Parent perception of their child's listening that they reported in the PEACH questionnaire may have been influenced by tympanometry results. Similarly, the results the parents obtained during the Ling 7-Sound Test could have also influenced the reflections in the PEACH questionnaire.

Additionally, parent and teacher report measures may exhibit results that are more positive rather than true observations of the child's listening due to the strengths based approach the Centre has to early intervention. Therefore, the findings from the PEACH and TEACH may suggest greater listening abilities than a more objective measure by a person of no relation to the child.

Another limitation of the current study is that the researcher administered the interviews. The current study was deliberately set up in a strength and relationship based service and for the researcher to develop a relationship over time with the families in the early intervention setting. While reasonably confident that the relationships were such that parents were comfortable to be open and honest, parents may have reported more positive, socially desirable responses to the researcher.

While precautions were taken to control the variables in the Ling 7-Sound Test, the nature of a parent administered assessment at home means it is possible that variables such as distance from the child and presentation order could have changed during the study without the researcher's knowledge.

4.8 Directions for future research

The current study demonstrated that the approach utilised was appropriate for a centre-based weekly early intervention programme with young children with Down syndrome. Future research to determine if such an approach works equally well when parents are engaging in one on one speech therapy or home based early intervention is warranted. Furthermore, a similar approach with school age children with Down syndrome would be of interest.

It is not necessarily common for an audiologist to be a member of a multi-disciplinary early intervention team working with young children with Down syndrome.

The current study demonstrates the value of an audiologist engaging with such a team. As such, further research involving audiologists in early intervention teams working with children who have complex disabilities would be beneficial.

Further, the follow through and management of fluctuating conductive hearing loss in young children with Down syndrome is another area in which research is warranted.

CHAPTER 5 CONCLUSION

The purpose of the present study was to investigate the parent use of the Ling 7-Sound Test to monitor the hearing of young children with Down syndrome. The measures used in the present study were tympanometry, the Ling 7-Sound Test, Parents' Evaluation of Aural/ Oral performance in Children (PEACH), Teachers' Evaluation of Aural/ Oral performance in Children (TEACH) and a social validity questionnaire with a semi-structured interview.

All four families in the study successfully incorporated the Ling 7-Sound Test into their daily routines and reported that the information the test provided was useful. One of the four children presented with binaural type B low tympanograms and a mild to moderate hearing loss, consistent with middle ear fluid. The Ling 7-Sound Test at 45 dB A was not sensitive to this change in hearing while the 40 dB A Ling stimuli were. The children generally scored highly in the Ling 7-Sound Test when they were well and paying attention to the task. Comparison of the PEACH and TEACH scores indicated that one child scored achieved lower TEACH than PEACH scores, while the other two children had close agreement between the two scores. The qualitative nature of the comments provided by parents in the PEACH, Ling 7-Sound Test, questionnaires and interviews increased understanding of each child's health, attention and listening behaviour in relation to their ear health and hearing throughout the 10 week study period.

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APPENDICES

Appendix A: Ethical approval

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

HUMAN ETHICS COMMITTEE

Secretary, Rebecca Robinson
Telephone: +64 03 369 4588, Extn 94588
Email: human-ethics@canterbury.ac.nz

Ref: 2018/16/ERHEC

23 May 2018

Beth Rees
Communication Disorders
UNIVERSITY OF CANTERBURY

Dear Beth

Thank you for providing the revised documents in support of your application to the Educational Research Human Ethics Committee. I am very pleased to inform you that your research proposal "Use of the Ling 6-Sound Test to Monitor Hearing in Young Children with Down Syndrome" has been granted ethical approval.

Please note that this approval is subject to the incorporation of the amendments you have provided in your email of 12th May 2018.

Should circumstances relevant to this current application change you are required to reapply for ethical approval.

If you have any questions regarding this approval, please let me know.

We wish you well for your research.

Yours sincerely

pp R. Robinson

Dr Patrick Shepherd
Chair
Educational Research Human Ethics Committee

Please note that ethical approval relates only to the ethical elements of the relationship between the researcher, research participants and other stakeholders. The granting of approval by the Educational Research Human Ethics Committee should not be interpreted as comment on the methodology, legality, value or any other matters relating to this research.

Ethical approval for amendment to include social validity questionnaire and interview

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

HUMAN ETHICS COMMITTEE

Secretary, Rebecca Robinson
Telephone: +64 03 369 4588, Extn 94588
Email: human-ethics@canterbury.ac.nz

Ref: 2018/16/ERHEC Amendment 1

13 June 2018

Beth Rees
Communication Disorders
UNIVERSITY OF CANTERBURY

Dear Beth

Thank you for your request for an amendment to your research proposal "Use of the Ling 6-Sound Test to Monitor Hearing in Young Children with Down Syndrome" as outlined in your email dated 8th June 2018. I am pleased to advise that this amendment has been considered and approved by the Educational Research Human Ethics Committee.

Please note that should circumstances relevant to this current application change you are required to reapply for ethical approval.

If you have any questions regarding this approval, please advise.

We wish you well for your continuing research.

Yours sincerely

pp R. Robinson

Dr Patrick Shepherd
Chair
Educational Research Human Ethics Committee

Please note that ethical approval relates only to the ethical elements of the relationship between the researcher, research participants and other stakeholders. The granting of approval by the Educational Research Human Ethics Committee should not be interpreted as comment on the methodology, legality, value or any other matters relating to this research.

F E S

Appendix B: Informed consent

Information sheet for participants

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

June 11, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Information sheet for parents/caregivers.

I am a second year Master of Audiology student at the University of Canterbury. I am researching parent use of an assessment tool, called the Ling 7-Sound Test, to monitor changes in hearing in preschool children with Down syndrome. The purpose of the research is to see whether the Ling 7-Sound Test is as effective as an objective measure, tympanometry, in the detection of hearing loss due to fluid in the ear. The research will occur over a school term, where parents will administer the Ling 7-Sound Test at home and the children will be screened using tympanometry once a week at the Champion Centre. The results from the Ling 7-Sound Test and tympanometry will be compared.

If you choose to take part in this study, your involvement in this project will be attending an information and training session in week 9 of term 2, and in term 3, administering the Ling 7-Sound Test to your child at home each day. The children will also be screened once a week using a tympanometer during their session at the Champion Centre in term 3. You will complete the Parents' Evaluation of Aural/Oral Performance of Children (PEACH) questionnaire each week. Similarly, your child's early intervention educator will complete the Teachers' Evaluation of Aural/Oral Performance of Children (TEACH) questionnaire

Upon completion of the study, at the end of term 3, you will complete a short questionnaire about your experiences during the study.

If you choose to take part in this study, you give permission to the researcher to access existing clinical notes on your child's health, language and audiology records from their Champion Centre file. Your child's team leader and early intervention educator at the Champion Centre will have access to the raw data collected from the Ling 7-Sound Test, tympanometry, hearing test, and PEACH/TEACH questionnaires.

During the training session, you will be taught how to administer the Ling 7-Sound Test to your child, and to record the results. The Ling 7-Sound Test involves the parent playing 7 sounds while standing behind their child and seeing if the child responds to the sounds by turning their head. The researcher will also screen the children using a tympanometer, so that they can get familiar with this. The children do not need to do anything for this test, it involves the researcher placing a soft tip into the child's ear for a few seconds that will measure the movement of their ear drum.

During term 3, you will administer the Ling 7-Sound Test to your child at home each day. You will record the results on a provided check sheet that you will return to the Champion Centre each week. I will check your child's ear health each week at the Champion Centre using a tympanometer as described above. You will be informed of any clinically significant results obtained from this, and what the recommended management is. Data from the Ling 7-Sound Test and tympanometry will be collected weekly and stored securely at the Champion Centre until analysis occurs following the completion of the study at the end of term 3.

It is estimated that the training session will take no longer than 30 minutes. It is estimated that the Ling 7-Sound Test will take no more than 5 minutes to administer each day and the questionnaire will take no longer than 5 minutes to administer each week. The tympanometry will take no longer than 5 minutes to administer each week at the Champion Centre. If your child's tympanometry result suggests fluid in the ear, I will conduct a short hearing screening at the Champion Centre to confirm hearing loss as a result of this. It is expected that this will take no longer than 10 minutes.
As a follow-up to this investigation, you will be asked to complete a short questionnaire about your experiences of administering the Ling 7-Sound Test on a regular basis to your child and the impact this may have.

The children may decline to participate in the Ling 7-Sound Test and/ or tympanometry and/or the hearing screening at any time, and this will be respected. There may also be times during the study that this task is not a priority for your family and that will also be respected.

To reduce any risks of anxiety or worry in relation to your child's ear health, you can be reassured that you will be advised if the research reveals any clinical concerns. You will also be provided with strategies and suggestions to address this. If a significant deterioration in hearing is detected, children will be referred to the Hospital Paediatric Audiology Department or University of Canterbury Speech and Hearing clinic for a full diagnostic hearing test and appropriate management.

Participation is voluntary, and you have the right to withdraw at any stage without penalty. Your services received at the Champion Centre will not be affected.
You may ask for your raw data to be returned to you or destroyed at any point. If you withdraw, I will remove information relating to you. However, once analysis of raw data starts on October 1, 2018 it will become increasingly difficult to remove the influence of your data on the results.

The results of the project may be published, but you may be assured of the complete confidentiality of data gathered in this investigation. To ensure anonymity and confidentiality, only the researcher, team leader and supervisors will have access to the raw data. All data will be stored securely in password protected facilities at the Champion Centre, as part of your child's file. A thesis is a public document and will be available through the UC Library.

Please indicate to the researcher on the consent form if you would like to receive a copy of the summary of results of the project.

The project is being carried out as a requirement for a Master of Audiology by Beth Rees under the supervision of Anne van Bysterveldt and Susan Foster-Cohen who can be contacted at anne.vanbysterveldt@canterbury.ac.nz. They will be pleased to discuss any questions you may have about participation in the project.

This project has been reviewed and approved by the University of Canterbury Educational Research Human Ethics Committee, and participants should address any complaints to The Chair, Educational Research Human Ethics Committee, University of Canterbury, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

If you agree to participate in the study, you are asked to complete the consent form and return to Beth Rees at the Champion Centre by June 22.

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

June 11, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Information sheet for staff.

I am a second year Master of Audiology student at the University of Canterbury. I am researching parent use of an assessment tool, called the Ling 7-Sound Test, to monitor changes in hearing in preschool children with Down syndrome. The purpose of the research is to see whether the Ling 7-Sound Test is as effective as an objective measure, tympanometry, in the detection of hearing loss due to fluid in the ear. The research will occur over a school term, where parents will administer the Ling 7-Sound Test at home and the children will be screened using tympanometry once a week at the Champion Centre. The results from the Ling 7-Sound Test and tympanometry will be compared.

If you choose to take part in this study, your involvement in this project will be completing a short questionnaire called the Teachers' Evaluation of Aural/Oral Performance of Children (TEACH) questionnaire about the child's listening each week during term 3.

The child's parents will be completing the Ling 7-Sound Test each morning with their child which involves the parent saying 7 sounds while standing behind the child and seeing if the child responds to the sounds by turning their head.
The researcher will also screen the children using a tympanometer once a week. The children do not need to do anything for this test, it involves the researcher placing a soft tip into the child's ear for a few seconds that will measure the movement of their ear drum. If the child's tympanometry result suggests fluid in the ear, I will conduct a short hearing screening at the Champion Centre to confirm hearing loss as a result of this. It is expected that this will take no longer than 10 minutes.

It is estimated that the questionnaire will take no longer than 5 minutes to complete each week.

There may be times during the study that this task is not a priority during your work and that will also be respected.

In the performance of completing the questionnaire there are no risks foreseen. Parents will be informed of any clinically significant results relating to their child's ear health, and strategies to implement to address these results.

Participation is voluntary, and you have the right to withdraw at any stage without penalty.

The results of the project may be published, but you may be assured of the complete confidentiality of data gathered in this investigation. To ensure anonymity and confidentiality, only the researcher, team leader and supervisors will have access to the raw data. Analysed data will be stored securely in password protected facilities at the Champion Centre, as part of your child's file. A thesis is a public document and will be available through the UC Library.

Please indicate to the researcher on the consent form if you would like to receive a copy of the summary of results of the project.

The project is being carried out as a requirement for a Master of Audiology by Beth Rees under the supervision of Anne van Bysterveldt and Susan Foster-Cohen who can be contacted at anne.vanbysterveldt@canterbury.ac.nz. They will be pleased to discuss any concerns you may have about participation in the project.

This project has been reviewed and approved by the University of Canterbury Educational Research Human Ethics Committee, and participants should address any complaints to The Chair, Educational Research Human Ethics Committee, University of Canterbury, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

If you agree to participate in the study, you are asked to complete the consent form and return to Beth Rees at the Champion Centre by June 22.

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

September 17, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Information sheet for parents/caregivers.

I am approaching you because you and your child have taken part in a study investigating the use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome. I would like to invite you to complete a questionnaire and take part in a brief interview at the end of this study.

If you choose to take part in this study, your involvement in this project will be completing a short questionnaire about your experiences of administering the Ling 7-Sound Test on a regular basis to your child and the impact this may have. You can do this independently or we can complete it together. Following this there will be a short interview. The questionnaire should take around 5-10 minutes, and the interview approximately 20 minutes. If it doesn't suit you to do this in person we can do it over the phone or I can send you the questions to complete in a written format. The face to face or phone interview will be audio recorded and notes will be taken and transcribed by me the researcher. The transcripts will be emailed to you to review. You can make any changes to these. If you change your mind about something you said you can remove it or add something new you may have forgotten. When you are happy these are accurate please return them to me by email.

Participation is voluntary, and you have the right to withdraw at any stage without penalty. Your services received at the Champion Centre will not be affected.

You may ask for your raw data to be returned to you or destroyed at any point. If you withdraw, I will remove information relating to you. However, once analysis of raw data starts on October 1, 2018 it will become increasingly difficult to remove the influence of your data on the results.

The results of the project may be published, but you may be assured of the complete confidentiality of data gathered in this investigation. To ensure anonymity and confidentiality, only the researcher, team leader and supervisors will have access to the raw data. All data will be stored securely in password protected facilities at the Champion Centre, as part of your child's file. A thesis is a public document and will be available through the UC Library.

Please indicate to the researcher on the consent form if you would like to receive a copy of the summary of results of the project.

The project is being carried out as a requirement for a Master of Audiology by Beth Rees under the supervision of Anne van Bysterveldt and Susan Foster-Cohen who can be contacted at anne.vanbysterveldt@canterbury.ac.nz. They will be pleased to discuss any questions you may have about participation in the project.

This project has been reviewed and approved by the University of Canterbury Educational Research Human Ethics Committee, and participants should address any complaints to The Chair, Educational Research Human Ethics Committee, University of Canterbury, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

If you agree to participate in this aspect of the study, you are asked to complete the consent form and return to Beth Rees at the Champion Centre by September 25.

Information sheet for the early intervention team leader regarding amendment to study design

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

September 17, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Information sheet for early intervention team leader.

I am approaching you because parents and children in your early intervention program have taken part in a study investigating the use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome. I would like to invite you to complete a questionnaire and take part in a brief interview at the end of this study.

If you choose to take part in this study, your involvement in this project will be completing a short questionnaire and interview upon completion of the study regarding your experiences during the study. You can complete the questionnaire independently or we can complete it together. Following this there will be a short interview. The questionnaire should take around 5-10 minutes, and the interview approximately 20 minutes. If it doesn't suit you to do this in person we can do it over the phone or I can send you the questions to complete in a written format. The face to face or phone interview will be audio recorded and notes will be taken and transcribed by me the researcher. The transcripts will be emailed to you to review. You can make any changes to these. If you change your mind about something you said you can remove it or add something new you may have forgotten. When you are happy these are accurate please return them to me by email.

Participation is voluntary, and you have the right to withdraw at any stage without penalty. Your services received at the Champion Centre will not be affected.

You may ask for your raw data to be returned to you or destroyed at any point. If you withdraw, I will remove information relating to you. However, once analysis of raw data starts on October 1, 2018 it will become increasingly difficult to remove the influence of your data on the results.

The results of the project may be published, but you may be assured of the complete confidentiality of data gathered in this investigation. To ensure anonymity and confidentiality, only the researcher and supervisors will have access to the raw data. All data will be stored securely in password protected facilities at the Champion Centre, as part of your child's file. A thesis is a public document and will be available through the UC Library.

Please indicate to the researcher on the consent form if you would like to receive a copy of the summary of results of the project.

The project is being carried out as a requirement for a Master of Audiology by Beth Rees under the supervision of Anne van Bysterveldt and Susan Foster-Cohen who can be contacted at anne.vanbysterveldt@canterbury.ac.nz. They will be pleased to discuss any questions you may have about participation in the project.

This project has been reviewed and approved by the University of Canterbury Educational Research Human Ethics Committee, and participants should address any complaints to The Chair, Educational Research Human Ethics Committee, University of Canterbury, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz).

If you agree to participate in this aspect of the study, you are asked to complete the consent form and return to Beth Rees at the Champion Centre by September 25.

Parent consent form

Department of Communication Disorders
Telephone: +64 3 369 3533
Email:
beth.rees@pg.cantebury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Consent form for parents/caregivers.

- ☐ I have been given a full explanation of this project and have had the opportunity to ask questions.
- ☐ I understand what is required of me if I agree to take part in the research.
- ☐ I understand that participation is voluntary and I may withdraw at any time without penalty. Withdrawal of participation will also include the withdrawal of any information I have provided should this remain practically achievable.
- ☐ I understand that any information or opinions I provide will be kept confidential to the researcher and supervisors and that any published or reported results will not identify the participants or the Champion Centre. I understand that a thesis is a public document and will be available through the UC Library.
- ☐ I understand that all data collected for the study will be kept in locked and secure facilities and in password protected electronic form as part of the child's file at the Champion Centre. The data will be backed up in password protected electronic form on the University of Canterbury server and will be destroyed after 5 years.
- ☐ I understand the risks associated with taking part include experiencing anxiety or worry in relation to my child's ear health and hearing. I will be advised if the research reveals any clinical concerns and I will be provided with strategies and suggestions to address this. . If a significant deterioration in hearing is detected, my child will be referred for a full diagnostic hearing test and appropriate management.
- ☐ I understand that I can contact the researcher, Beth Rees, or supervisors Anne van Bysterveldt and Susan Foster-Cohen for further information. If I have any complaints, I can contact the Chair of the University of Canterbury Educational Research Human Ethics Committee, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz)
- ☐ I would like a summary of the results of the project.
- ☐ By signing below, I agree to participate in this research project.

Name:____________________________Signed:___________________________Date:_________

Email address *(for report of findings)*:

____

Please return this consent form to Beth Rees at the Champion Centre by June 22, 2018

Staff consent form

Department of Communication Disorders
Telephone: +64 3 369 3533
Email:
beth.rees@pg.cantebury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Consent form for staff.

- ☐ I have been given a full explanation of this project and have had the opportunity to ask questions.
- ☐ I understand what is required of me if I agree to take part in the research.
- ☐ I understand that participation is voluntary and I may withdraw at any time without penalty. Withdrawal of participation will also include the withdrawal of any information I have provided should this remain practically achievable.
- ☐ I understand that any information or opinions I provide will be kept confidential to the researcher and supervisors and that any published or reported results will not identify the participants or the Champion Centre. I understand that a thesis is a public document and will be available through the UC Library.
- ☐ I understand that all data collected for the study will be kept in locked and secure facilities and in password protected electronic form as part of the child's file at the Champion Centre. The data will be backed up in password protected electronic form on the University of Canterbury server and will be destroyed after 5 years.
- ☐ I understand the risks associated with taking part include parents experiencing anxiety or worry in relation to their child's ear health and hearing and that they will be advised if the research reveals any clinical concerns and will be provided with strategies and suggestions to address this.
- ☐ I understand that I can contact the researcher, Beth Rees, or supervisors Anne van Bysterveldt and Susan Foster-Cohen for further information. If I have any complaints, I can contact the Chair of the University of Canterbury Educational Research Human Ethics Committee, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz)
- ☐ I would like a summary of the results of the project.
- ☐ By signing below, I agree to participate in this research project.

Name:____________________________Signed:___________________________Date:__________

Email address *(for report of findings)*:

____

Please return this consent form to Beth Rees at the Champion Centre by June 22, 2018.

Parent consent form following amendment to study design

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

UC UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

September 17, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Consent form for parents/caregivers.

- ☐ I have been given a full explanation of this project and have had the opportunity to ask questions.
- ☐ I understand what is required of me if I agree to take part in the research.
- ☐ I understand that participation is voluntary and I may withdraw at any time without penalty. Withdrawal of participation will also include the withdrawal of any information I have provided should this remain practically achievable.
- ☐ I understand that any information or opinions I provide will be kept confidential to the researcher and supervisors and that any published or reported results will not identify the participants or the Champion Centre. I understand that a thesis is a public document and will be available through the UC Library.
- ☐ I understand that all data collected for the study will be kept in locked and secure facilities and in password protected electronic form as part of my child's file at the Champion Centre. The data will be backed up in password protected electronic form on the University of Canterbury server and will be destroyed after 5 years.
- ☐ I understand the interview will be audio recorded and notes will be taken by the researcher during the interview which will be transcribed and sent to me to review.
- ☐ I understand the risks associated with taking part include experiencing anxiety or worry in relation to my child's ear health and hearing. I will be advised if the research reveals any clinical concerns and I will be provided with strategies and suggestions to address this.
- ☐ I understand that I can contact the researcher, Beth Rees, or supervisors Anne van Bysterveldt and Susan Foster-Cohen for further information. If I have any complaints, I can contact the Chair of the University of Canterbury Educational Research Human Ethics Committee, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz)
- ☐ I would like a summary of the results of the project.
- ☐ By signing below, I agree to participate in this research project.

Name:______________________________Signed:____________________________Date:__________

Email address *(for report of findings, if applicable)*:

____

Please return this consent form to Beth Rees at the Champion Centre by September 25, 2018

Early intervention team leader consent form following amendment to study design

UC UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Department of Communication Disorders
Telephone: +64 3 369 3533
Email: beth.rees@pg.cantebury.ac.nz

September 17, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.
Consent form for early intervention team leader.

☐ I have been given a full explanation of this project and have had the opportunity to ask questions.

☐ I understand what is required of me if I agree to take part in the research.

☐ I understand that participation is voluntary and I may withdraw at any time without penalty. Withdrawal of participation will also include the withdrawal of any information I have provided should this remain practically achievable.

☐ I understand that any information or opinions I provide will be kept confidential to the researcher and supervisors and that any published or reported results will not identify the participants or the Champion Centre. I understand that a thesis is a public document and will be available through the UC Library.

☐ I understand that all data collected for the study will be kept in locked and secure facilities. The data will be backed up in password protected electronic form on the University of Canterbury server and will be destroyed after 5 years.

☐ I understand the interview will be audio recorded and notes will be taken by the researcher during the interview which will be transcribed and sent to me to review.

☐ I understand that I can contact the researcher, Beth Rees, or supervisors Anne van Bysterveldt and Susan Foster-Cohen for further information. If I have any complaints, I can contact the Chair of the University of Canterbury Educational Research Human Ethics Committee, Private Bag 4800, Christchurch (human-ethics@canterbury.ac.nz)

☐ I would like a summary of the results of the project.

☐ By signing below, I agree to participate in this research project.

Name:______________________________Signed:_____________________________Date:__________

Email address *(for report of findings, if applicable)*:______________________________________

Please return this consent form to Beth Rees at the Champion Centre by September 25, 2018

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome.

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Assent form for children.

I know that:

	Beth has told me about her project about listening.
	I will play a listening game with Mum or Dad every day.
	Beth will check my ears each week at the Champion Centre.
	I can say no if I don’t want to.
	My parents said it’s okay.

Choose one of the faces:

YES, I want to be in the project. **NO, I don’t want to.**

☺ ☹

Appendix C: Ling 7-Sound Test scoresheet

Name: ___________________________ **Ling 7-Sound Test Score Sheet**

☐ TV/radio off ☐ Quiet room

Place: ☐ Same ☐ Different. If so, please describe: ________________________________

✓ = response — = no response

Week of:	Health	My child paid attention:	Background noise:	oo	ah	ee	sh	s	or	m
Tuesday	Very well Somewhat well Not well	Always Sometimes Never								
Wednesday	Very well Somewhat well Not well	Always Sometimes Never								
Thursday	Very well Somewhat well Not well	Always Sometimes Never								
Friday	Very well Somewhat well Not well	Always Sometimes Never								
Monday	Very well Somewhat well Not well	Always Sometimes Never								

Please provide comments about your experience:

____

____

____

____

____

____

____

____

____

Tips:

-Have the speaker 1m from your child

-choose a time when your child is ready to pay attention

-if possible, play the sounds in exactly the same place each day

-make sure it is as quiet as possible

-try to vary the time between sounds

Appendix D: Social validity measures

Parent social validity questionnaire

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome - Parent questionnaire.

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Please circle or add your answer as necessary.

1. Prior to the study, I was concerned about my child's hearing

   Never Rarely Sometimes Often Always

2. During training, I felt I had adequate support from the researcher:

   Never Rarely Sometimes Often Always

3. Throughout the process, I felt I had adequate support from the researcher:

   Never Rarely Sometimes Often Always

4. What could have improved the training?

   ____

5. Following the training, I was confident to administer the Ling 7-Sound Test

   Never Rarely Sometimes Often Always

6. It was easy to engage my child in the Ling 7-Sound Test

   Never Rarely Sometimes Often Always

7. It was easy to incorporate the Ling 7-Sound Test into our family's routine

   Never Rarely Sometimes Often Always

8. The record sheet was useful in reminding me to complete the Ling 7-Sound Test

   Never Rarely Sometimes Often Always

9. The text message was useful in reminding me to complete the Ling 7-Sound Test

   Never Rarely Sometimes Often Always

10. The information I got from the Ling 7-Sound Test was useful

   Never Rarely Sometimes Often Always

11. When I found my child wasn’t hearing well in the Ling 7-Sound Test, I used communication strategies:

Never Rarely Sometimes Often Always

12. When I used the communication strategies, they had a positive impact:

Never Rarely Sometimes Often Always

13. The Ling 7-Sound Test results matched my informal observations of my child’s hearing:

Never Rarely Sometimes Often Always

14. The Ling 7-Sound Test results were informative:

Never Rarely Sometimes Often Always

15. The PEACH questionnaire results were informative:

Never Rarely Sometimes Often Always

16. The Ling 7-Sound Test was useful as a hearing monitoring device:

Never Rarely Sometimes Often Always

17. The PEACH questionnaire was useful as a hearing monitoring device:

Never Rarely Sometimes Often Always

18. I was confident in the results from the Ling 7-Sound Test:

Never Rarely Sometimes Often Always

19. I was confident in the results from the PEACH questionnaire:

Never Rarely Sometimes Often Always

20. I was confident in the results from the TEACH questionnaire:

Never Rarely Sometimes Often Always

21. I was confident in the results from the tympanometry:

Never Rarely Sometimes Often Always

22. I was confident in the results from the hearing test:

Never Rarely Sometimes Often Always

Use of the Ling 7-Sound Test to monitor hearing in young children with Down syndrome – Early intervention team leader questionnaire.

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Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

Please circle or add your answer as necessary.

1. Prior to the study, I was concerned about the children's hearing

   Never Rarely Sometimes Often Always

2. The Ling 7-Sound Test was a practical tool for parents to use to monitor their child's hearing

   Never Rarely Sometimes Often Always

3. Throughout the process, the parents had adequate support from the researcher:

   Never Rarely Sometimes Often Always

4. It is important for therapists to regularly know about the children's hearing

   Never Rarely Sometimes Often Always

5. It is important for early intervention educators to regularly know about the children's hearing

   Never Rarely Sometimes Often Always

6. It is important for parents to regularly know about their children's hearing

   Never Rarely Sometimes Often Always

7. Communication strategies are an effective tool for parents to use to support their children when they are not hearing well

   Never Rarely Sometimes Often Always

8. Communication strategies are an effective tool for Early Intervention Educators to use to support the children when they are not hearing well

   Never Rarely Sometimes Often Always

9. Parents' informal observations are accurate in detecting when their children have hearing loss

   Never Rarely Sometimes Often Always

10. Therapists' informal observations are accurate in detecting when children have hearing loss

    Never Rarely Sometimes Often Always

11. The information I got from the Ling 7-Sound Test was useful to my speech language therapy practice

Never	Rarely	Sometimes	Often	Always

12. The information parents got from the Ling 7-Sound Test was valued

Never	Rarely	Sometimes	Often	Always

Appendix E: Visual schedules

Tympanometry visual schedule

Sitting down	
Still like a statue	
Look in ears	
Ear tickles	
Finished	

Play audiometry visual schedule

Sitting down	
Headphones on	
Quiet	
Listen	
Game	
Finished