The Development and Evaluation of the Hearing Intervention Battery in Arabic (HIBA) for Auditory Perception in Children with Cochlear Implants

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DECLARATION

I, Hanin Hassan H Rayes, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis. Portions of this thesis have been published in conference proceedings and one peer-reviewed article has been published in Journal of Speech Language and Hearing Research.

DEDICATION

I dedicate this work to my father whose courage and persistence shaped my future and my mother whose love and resilience nurtured my soul.

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وَمَا تَوْفِيقِي إِلَّا بِاللَّهِ عَلَيْهِ تَوَكَّلْتُ وَإِلَيْهِ أُنِيبُ

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CONFERENCE PARTICIPATION AND PUBLICATIONS

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GLOSSARY

AC	Alternating Current
AT	Auditory Training
BCT	Behaviour change technique
CHL	Conductive Hearing Loss
CI(s)	Cochlear Implant(s)
CIS	Continuous Interleaved Sampling
CF	Characteristic Frequency
DC	Direct Current
DPOAE	Distortion Product Otoacoustic Emission
HIBA	Hearing Intervention Battery in Arabic
IHCs	Inner Hair Cells
LAD	Language Acquisition Device
OHCs	Outer Hair Cells
RCT	Randomized Control Trial
SNHL	Sensorineural Hearing Loss
WHO	World Health Origination

ABSTRACT

The Hearing Intervention Battery in Arabic (HIBA), is a multi-modal auditory training intervention, that was developed based on the recommendations from our published systematic review of the literature on the effectiveness of auditory training (AT) for children with cochlear implants (CIs). HIBA was primarily intended to help improve speech and pitch perception in Arabic-speaking children with CIs.

Due to the lack of auditory and speech assessment tools for the Arabic language, the A-CAPT, an Arabic version of the English Chear Auditory Perception Test (CAPT) was developed. The A-CAPT was validated prior its use in this project with 26 children with typical hearing. There was a strong agreement between the test and retest measures and normative data and the critical difference values were calculated which were similar to the British English CAPT.

A randomized control trial (RCT) to evaluate the HIBA training programme was conducted with 14, 5- to 13-year-old Arabic-speaking children with CIs. The control group received art training following step-by-step drawing and face-paint exercises while the HIBA multi-modal training group received games involving communication interactions (DiaPix), speech cue discrimination (Alefbata.com), and pitch discrimination (musical discrimination using a keyboard). All tasks were interactive and designed to be completed by the children together with their parents or caregivers. There was a double baseline measurement, followed by a 4-week intervention period before a post intervention assessment.

There was a significant improvement in consonant perception for children who received the HIBA multi-modal training intervention but this was not observed in the active control group. There was some evidence of generalization of learning, as observed by improvements in the non-trained task (phoneme discrimination) for the intervention group but not for controls. It was unclear if one particular element of the HIBA led to these improvements.

Parents were actively involved in the multi-modal training group and their feedback indicated that the most preferred part of multi-modal training was the communication interaction tasks using the Diapix. To understand which element of the HIBA led to improvements in speech perception and whether the duration of training and sample size masked any gains, a trial forward in a larger scale should be conducted. In addition, to improve the quality of evidence of the study, collaboration is need to achieve a double blinded study and minimize bias.

Findings of this project may suggest that children with CIs and their parents can benefit from regular and sustained access to age-appropriate auditory training materials and activities. In addition, findings would extend the current understanding of the impact of auditory training on CI outcomes in children and provide inspiration for a more comprehensive rehabilitation scheme for CI users.

IMPACT STATEMENT

In Saudi Arabia, children with CIs make up a growing but underserved population due to scarce (re)habilitation services. The need for raising awareness and enforcing aural (re)habilitation programmes is dire at best. Hence, the culmination of the series of projects, showcased here, is the development of HIBA, an evidence-based multi-modal parent-led auditory training programme.

An evidence-based approach was taken to the design of HIBA using a systematic review to critically appraise the literature of auditory training in children with CIs. The review evaluated a variety of research designs and summarized essential measures that would increase the reliability of the studies when utilized. It also highlighted the importance of selecting the appropriate outcome measures. Driven by the limited number of validated and published tests for Arabic-speaking children in Saudi Arabia, the A-CAPT was developed to be the gold standard outcome measure. Based on the English CAPT, the A-CAPT will assess the discrimination of speech cues and provide clinicians with frequency-specific information with high reliability. In fact, since it is developed in simple-worded Standard Arabic, we predict that the A-CAPT will be an essential tool for clinicians working with all Arabic speaking patients, including in non-Arabic speaking countries, regardless to dialect.

After participating in a four-week intervention, benefits from this multi-modal auditory training were observed for complex pitch discrimination and for generalisation to speech cue discrimination. We predict that longer training periods would further improve outcomes but a future study is required to corroborate this. Furthermore, we confirmed that parents are ready to implement home-based interventions when appropriately trained and provided with easy to use materials. We also showed that children with CIs can benefit from regular and sustained access to age-appropriate auditory training materials and activities. By making HIBA readily accessible, families will be empowered to facilitate their children's speech and language development, while clinicians can monitor their progress and compliance and provide on-going support.

Finally, this research extends new opportunities for further research and development. To list a few, studies are needed to validate the current version of the A-CAPT in background noise, to validate the current version with Arabic-speaking children in countries other than Saudi Arabia, and to develop and validate a modified version of the A-CAPT in a variety of Arabic dialects. Technically, the methodology used to develop the A-CAPT, based on the English version of the same test, can be utilized to develop the test in other languages. A larger scale trial of HIBA is also needed to further enhance its effectiveness through adjustment of its parameters, such as the optimum age for the intervention, the duration of the intervention, and the dynamic progression of the intervention's complexity according to subject's own development.

CHAPTER 1: INTRODUCTION

1.1 BACKGROUND

The current gold standard treatment for severe to profound deafness in children is cochlear implantation. There is strong support in the literature for the use of aural (re)habilitation post implantation to maximize CI outcomes (Kennedy et al., 2006; Kral, 2013; Moeller, 2000; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998). However, not all children with CIs have access to such services. In Saudi Arabia, in particular, follow-up intervention services for children with hearing loss are scarce (Alyami, Soer, Swanepoel, & Pottas, 2016; Milaat, Ghabrah, Al-Bar, Abalkhail, & Kordy, 2001). In fact, a study conducted before the implementation of new-born hearing screening programme in 2010 in Saudi Arabia indicated that most of the people with disabilities, including deaf and hard of hearing individuals, did not have access to essential services such as aural rehabilitation, psychological, and educational support at an early age (Hanafi, 2007). Such lack of access to essential services negatively affected the language and academic achievements of children with hearing impairment.

Although progress has been reported post the implementation of the new-born hearing screening programme, these programmes are mostly available in hospitals found in metropolitan cities and thus still only reaching a limited number of the population. A more recent study reported that children residing in Riyadh, the capital of Saudi Arabia, were fitted with hearing devices and enrolled into early intervention services earlier than those living outside of the city. The study emphasized on the needs for making rehabilitation services accessible to all families throughout the country (Alyami et al., 2016).

Even though this project looked at ways to address this issue specifically in Saudi Arabia, the approach is relevant to many countries facing the same difficulties with service delivery for rehabilitation. We developed a multi-modal approach for delivering rehabilitation informed by a systematic review of the literature. We used a parent-delivery approach to maximize family engagement. The training battery is called HIBA (the Hearing Intervention Battery in Arabic) and it was specifically designed for school-aged children with CIs.

In this section, hearing loss and its impact on children is discussed together with ways to potentially improve outcomes.

1.2 HEARING LOSS

Hearing loss is the most common sensory disorder in humans (Wolrd Health Organization, 2018). According to the World Health Organization (WHO) report on deafness and hearing loss in 2018, more than 5% of the world's population, approximately 466 million people, has disabling hearing loss, 34 million of which are children. This number is estimated to rise above 900 million by 2050; by then, almost one in every ten individuals will have disabling hearing loss. Disabling hearing loss is defined as hearing loss above 40 dB HL in the better hearing ear for adults and above 30 dB HL in the better hearing ear for children.

Hearing loss can be described as a partial or total absence of auditory sensitivity due to peripheral and/or central auditory deficits. The degree of hearing loss refers to the severity of the loss and is classified in children as mild (20-40 dB HL), moderate (41-70 dB HL), severe (71-95 dB HL) or profound (more than 91 dB HL) (British Society of Audiology, 1988).

Hearing loss can be caused by complications that occur when sound waves cannot travel through the outer ear canal, tympanic membrane, or middle ear ossicles due to a dysfunction in the pathway, dampening sounds and making it difficult to hear; this type of hearing loss is called conductive hearing loss (CHL). CHL can be caused by a number of factors some of which can be as simple as impacted cerumen or presence of a foreign body, while others can be more serious, such as fluid in the middle ear, poor eustachian tube function, perforated tympanic membrane, or infections in outer or middle ear. Other more complex issues that could cause CHL include damage to the middle ear structures, absence or malformation of the outer ear, ear canal, or middle ear, benign tumors, and skull fractures due to head trauma. Generally, this type of hearing loss often can be corrected by medical or surgical intervention (ASHA, 2020).

Hearing loss can also occur due to defects in the cochlea (sensory) or the VIIIth nerve (neural) and is called sensorineural hearing loss (SNHL). More specifically, SNHL affects at least 1 in 500 newborns, with over 50% of these being caused by hereditary factors (Wolrd Health Organization, 2018). Factors that cause congenital hearing loss includes intrauterine infections including rubella, cytomegalovirus, herpes simplex virus, complications associated with the Rh factor in the blood, prematurity, maternal diabetes, toxemia during pregnancy, and lack of oxygen (anoxia) (ASHA, 2020). Furthermore, SNHL can be acquired after birth through viral and bacterial infections, or aminoglycoside and cisplatin ototoxicity (Wolrd Health Organization, 2018).

Despite the tremendous effort that has been put into gene therapy and medical surgical approaches, yet a treatment that re-instates hearing abilities has not been found. There are major obstacles limiting the effectiveness of such approaches, including addressing the blood-labyrinth barrier, producing more specific and effective delivery approaches for therapies, and optimizing surgical access (Ren, Landegger, & Stankovic, 2019). Typically, SNHL is treated with hearing devices either with or without surgical intervention. However, for patients with SNHL who cannot benefit from acoustical hearing aids, implantable hearing devices are used with CIs being the most frequently worn by those with greater degrees of loss.

1.3 SOUND AND NATURAL HEARING

1.3.1 Sound Transmission

Sound waves travel through the external ear canal, strikes the tympanic membrane, and generates vibration that is transferred to the middle ear. The vibration then moves the ossicles, small bones that are found in the middle ear space, to generate pressure waves within the cochlea and to vibrate the basilar membrane. High frequency sounds induce maximum vibration toward the base of the cochlea while low frequency sounds produce maximal vibration toward the apex. Each place has a characteristic frequency (CF), which is characterized by a maximal vibration at a given place on the basilar membrane. A wide frequency range of sound energy is efficiently transferred from the air cavity to the cochlear fluids via this mechanical coupling mechanism. Then, mechanoelectrical transduction, a process of detection and conversion of sound stimulus to an equivalent electrical waveform, occurs in the sensory hair cells placed on the basilar membrane (Fettiplace, 2017; Moore, 2010).

1.3.2 Hair Cells

Human cochlea contains two types of hair cell, inner hair cells (IHCs) and outer hair cells (OHCs). The main function of OHCs, which form three rows of 12,000 hair cells along the basilar membrane, is to amplify the sound signal by mechanically amplifying the sound-induced vibrations of the cochlear partition. This active mechanism process is achieved by changing the OHCs stiffness and length in response to the vibration on the basilar membrane, increasing the amplitude of vibration and sharpening the tuning on the basilar membrane, and enhance the frequency selectivity. This specific selectivity pertains to the ability of the auditory system to precisely distinguish between the different frequencies that are present in complex sounds (i.e., speech and music) (Fettiplace, 2017; Hudspeth, 1997; Moore, 2010). The IHCs, which form a single row of 3,500 hair cells along the basilar membrane, are innervated by up to 20 afferent nerve fibres each, and are the actual sensory receptors. The function of the IHCs is to transfer the signal into electrical impulses that are transmitted tonotopically to the auditory nerve fibres up to the auditory pathways and auditory cortex (Fettiplace, 2017; Hudspeth, 1997; Moller, 2006).

The depolarization potential of the hair cells occurs when the hydromechanical energy moving the basilar membrane upward in response to the sensory input, causing the stereocilia of the hair cells to move away from the modiolus of the cochlea and trigger depolarization. In the IHCs, depolarization potential is generated when potassium influx triggers calcium influx from the presynaptic, causing a release of the neurotransmitter glutamate. Glutamate is then picked up by the nerve endings and stimulates the group of nerve fibers or spiral ganglions to generate action potential and transfer electrical impulses to the auditory cortex (Bear, 2006). This transmission has high temporal precision and a wide dynamic range of intensity coding of acoustic sounds (Fuchs, 2005; Moser, Neef, & Khimich, 2006; Palmer & Evans, 1982). Intact sensory receptors are essential to relay acoustic signals through the auditory pathways.

1.3.2.1 Hair Cell Damage

Damage to the hair cells within the cochlea is associated with cochlear hearing loss. OHCs damage lessens the vibration of the basilar membrane and impairs the active mechanism of the cochlea. Clinically, this dysfunction is manifested as elevated hearing thresholds. Damage to the IHCs impairs the process of stimulation of the auditory nerve and causes greater elevation of hearing thresholds. Often in cases with greater degrees of cochlear loss, complete dysfunction to the IHCs at certain places on the basilar membrane occurs effecting the auditory neurons in contact with those areas; these places are called dead regions (Moore, 2001). Basilar membrane vibration that occurs within a dead region cannot be detected by the neurons connected there, however, a region closer to the CF of the signal can be stimulated if basilar membrane vibration were sufficient; this phenomena is called “off-place or off-frequency listening.” (Moore, 2010).

Synaptic damage or losses between IHCs and auditory nerve fibers at the presynaptic site (the base of the hair cell) or the postsynaptic site (terminal dendrite of the spiral ganglion) leads to deficits including temporal and intensity coding discrepancy and are associated with different perceptual hearing difficulties (Bharadwaj, Masud, Mehraei, Verhulst, & Shinn-Cunningham, 2015). Depending on the degree of hearing deficits, this type of hearing loss can be managed by hearing devices (i.e. hearing aids and/or CIs).

1.4 SOUND AND HEARING DEVICES

1.4.1 Hearing Aids

Hearing aids are essentially sound amplifiers that pick-up sounds using built-in microphones and transform them from analogue sound waves to digital electronic signals and then transmit them to the processor. The processor analyses and adjusts the incoming sound signal based on specific parameter settings and signal processing algorithms. Typically, the input signal is divided into multiple frequency bands and differing degrees of amplification applied to each according to a target gain formula. The signal is then converted back to an acoustic signal and travels through the auditory pathway just as described above.

The neural activity produced by hearing aids is not the same as for normal hearing. The signal processing does not return hearing to normal, partly due to the distortion caused by damage to the structures in the ear and due to the capability of the hearing aid to deliver the appropriate gains. Accordingly, hearing aids can improve audibility but not necessarily the fidelity of sound (Kuk, Lau, Korhonen, & Crose, 2015).

1.4.2 CIs

CIs are considered the most successful neuroprosthesis with more than 700,000 recipients worldwide (European Association of Cochlear Implant users, 2017; Oehlerking, 2020). When hearing aids are not an option it is primarily a result of damaged IHCs and the associated synapses. In the majority of cases the neurons remain viable although there can be sections of neural damage, typically the nerve fibres survive in sufficient numbers to be stimulated electrically with CIs (Seyyedi, Eddington, & Nadol, 2011). CIs bypass the

IHCs and its associated synapses and directly stimulate the nerve fibres transmitting the electrical signal to the brain, at which point the brain may interpret the signal as a sensory input or “sound” (Kral, Kronenberger, Pisoni, & O’Donoghue, 2016).

The health of the hair cells and its surrounding structure including the synapse will not affect CI outcomes. In contrast, the integrity of the auditory nerve, nerve fibres, or any structure or conjunctions in the auditory pathways in the nervous system (i.e., synapses between the spiral ganglion and the cochlear nucleus, midbrain, or auditory cortex) may disturb the electrical transmission of sound provided by the CIs and could reduce CI outcomes (Shearer & Hansen, 2019).

1.4.2.1 Components of CIs

CIs consist of external and internal components. The external parts are made up of sound processer usually placed behind the ear and a transmitting coil placed on the head. The surgically implanted internal parts are the receiver, stimulator, and electrode array. The external processor picks up sounds from the environment via microphones and convert it into electrical signals using a built-in micro-computer. The electrical signal is then transmitted through the skin as a radio-signal via the external coil to the receiver and stimulator components, which process the input and transmit it to the electrode array placed in the scala tympani in the cochlea.

1.4.2.2 Electrodes in the CI

CI have between 12–22 electrodes or stimulation contacts depending on the device and are arranged longitudinally along the electrode array similar to the frequency layout in the cochlea, preserving the tonotopic organisation of the auditory nerve (Kral et al., 2016). When the CI receives sound, it passes it to a filter bank to separates the signal into different frequency bands. An envelope detector receives the output of each filter to estimate the energy and evolution in time for each band. Then, acoustic dynamic range for each channel is transformed into an electrical dynamic range, each assigned to one electrode; such transformation is individualized for each patient and is different for each electrode. At this point, the processor generates the stimulation pulses, based on the stimulation rate assigned, representing the current level to be presented at each electrode and at each time instant (Saleh, 2013).

1.4.2.3 Speech Processing Strategies

An ideal speech processing strategy is one that most accurately mimics or reproduces the original sound spectrum and provide CI users with clearer sounds. Stimulating strategies main function is to transform sound waveform into a series of electric impulses that determines which electrodes should be activated in each cycle. There are several signal processing strategies that are available for CI systems one of which is continuous interleaved sampling (CIS). CIS is widely used as it is available on all major CI systems. CIS is a non-simultaneous pulsatile strategy that stimulates all active electrodes for each cycle (Wilson, Finley, Lawson, Wolford, & Zerbi, 1993). The sound signal is passed through a number of band-pass filters, then temporal envelope for each of those waveforms is extracted using either half-wave or full-wave-rectification technique. These extracted envelopes are mapped into a narrow electric dynamic range (Loizou, 1998; F. G. Zeng et al., 2002) that is used to modulate biphasic pulses to deliver the signal at a set rate ranges from hundreds to thousands per second (Loizou, 1998; F. G. Zeng, Rebscher, Harrison, Sun, & Feng, 2008). CIS generates the stimulation pulses in a way such that at only one channel is active at a given moment. This allow CIS to prevent electrode interaction which may cause smearing of envelope cues (Gang Zeng, 2004; Zeng et al.,

2008) and avoid electrical-field interference that is caused by simultaneous activation. Other signal processing strategies, such as advanced combinational encoder (ACE) HiRes120 and Hybrid stimulating strategy, are not covered here as they are out of scope of this work. However, detailed information are available (Loizou, 1998; Rouiha, Bachir, & Ali, 2008; Saleh, 2013).

1.4.2.4 Factors Affecting Outcomes with CIs

1.4.2.4.1 Subject-dependent Factors

The main factors predicting CI outcomes in children are: age of implantation, residual hearing before implantation, parent-child interactions, socioeconomic status, maternal education (Baker & Hazan, 2011; Connor, Craig, Raudenbush, Heavner, & Zwolan, 2006; Miyamoto, Colson, Henning, & Pisoni, 2017; Niparko et al., 2010; Yoshinaga-Itano, Sedey, Wiggin, & Mason, 2018), and language acquisition status prior to cochlear implantation (prelingual or postlingual) (Kane, Schopmeyer, Mellon, Wang, & Niparko, 2004). Other general factors predicting CI recipients' speech or language performance post implantation include electrode coupling (Mens & Berenstein, 2005; Pfingst, Franck, Xu, Bauer, & Zwolan, 2001), signal processing approach (Nogueira, Litvak, Saoji, & Büchner, 2015; M. W. Skinner et al., 2002; Wilson et al., 1988), and quality of CI fitting (Holden, Vandali, Skinner, Fourakis, & Holden, 2005; M. W. Skinner, 2003)

1.4.2.4.2 Electrode-dependent Factors

There are many electrode-dependent factors that could degrade CI performance, one of which is the number of active electrodes. Electrodes on the electrode array receive frequencies related to the information in the band pass filters. Technically each channel can be set to correspond to a single electrode (1-to-1 mapping), however, practically the number of electrodes does not always correspond to the number of spectral channels provided by the CI (Blamey et al., 1992; Zwolan et al., 1997; Fu et al., 1998 and Friesen et al., 2001). Two or more electrodes can stimulate the same auditory nerve ending and produce only one sound percept, thus different sound percepts often cannot be produced by closely spaced electrodes. Electrodes that do not provide distinct sound percept's may lead to poor perception since cycles of information in a CIS would deliver duplicate signal effecting perception, these electrodes could be considered problematic and may require intervention (Saleh, 2013). In contrast, utilizing more electrodes could be beneficial particularly when dead regions exist. When more than one electrode is activated, adjacent healthy regions can be stimulated using a different subset of electrodes and override the unresponsive regions (Fu and Nogaki, 2004 and Dorman and Spahr, 2006).

Since the number of electrodes and the range of bandwidth assigned to each electrode is limited, pitch perception is restricted. In addition, spatial coding cannot be coded in the same level of accuracy (F. Zeng & Fay, 2013) leading to cross-channel interference and undetailed spectral information (Kral et al., 2016). Also, the range of intensities that CI can offer is limited (F. Zeng & Fay, 2013). Fortunately, speech perception and recognition are still possible with the use of CI even under degraded conditions, thanks to the robustness of the brain and its ability to perceive speech (Shannon, Zeng, Kamath, Wygonski, & Ekelid, 1995).

1.4.2.5 Quality of Sound in CIs

CIs deliver sound to the brain in a very different way to natural hearing, CI users with Signal Sided Deafness (SSD) can probably more accurately compare the quality of acoustical signal in the healthy ear to the electrical signal in the implanted ear. A recent study was conducted to assess the sound quality of CI users with SSD requested from the participants to rate the similarity between two signals, one was a pure speech signal that was presented to the ear with CI and the other was a CI-like signals presented to the ear with normal hearing (Dorman, Natale, Butts, Zeitler, & Carlson, 2017). CI-like signals were synthesized using noise vocoded signals, sine vocoded signals, frequency shifted, and band-pass filtered. Vocoders, devices that were used originally with normal hearing listeners to investigate speech transmission systems for telephones, were used in the research in effort to create signals that mimic the sound quality produced by CIs (Dudley, 1939). Rating the responses of the participants using a scale from 1-10 with 10 being a complete match to the CI signals, the median rating of eight listeners to the sound of the CI for noise vocoded signals was 1.9; for sine vocoded signals 2.9; for frequency upshifted signals, 1.9; and for band pass filtered signals, 5.5. In another experiment, three listeners rated combinations of band pass filtering and spectral smearing signal to be 10. The study concluded that sounds produced by noise and sine vocoders do not actually correspond to the sound quality of CI. Instead, natural speech signals that have been muffled by band pass filtering and/or spectral smearing may offer a closer match to CI sound quality. Although this study does not give a definitive answer about how hearing through CI sound like, it demonstrates that hearing using CI does sound distorted (Dorman et al., 2017). There are many factors that affect the performance of CI some of which were described in section 1.4.2.4. Fortunately, the degraded signal of sound is processed and interpreted by the brain as meaningful sound.

1.5 DEVELOPMENT OF AUDITORY SYSTEM AND CRITICAL PERIODS

1.5.1 Development of Auditory System

In humans, the auditory system begins to develop in utero. By 15 weeks of gestation, the structural parts of the cochlea and the middle ear are well formed and they become anatomically functional around 20 weeks of gestation. Between 25 to 29 weeks of gestation, the auditory system becomes functional when the ganglion cells of the cochlear nucleus connects the IHCs to the brainstem and temporal lobe of the cortex (Hall, 2000). The earliest evidence of an auditory evoked response can be observed at 16 weeks of gestation when a physiologic response is produced by the ganglion cells in the cochlea that are connected to nuclei in the brainstem (Graven & Browne, 2008). Around 25 to 26 weeks of gestation, autonomic function such as heart rate, blood pressure, respiratory pattern, gastrointestinal motility, and oxygenation can be modified in response to loud noises in utero (Morris, Philbin, & Bose, 2000). The neural connections to the temporal lobe of the cortex are functional starting at 28 to 30 weeks of gestation. This begins the development of tonotopic columns in the auditory cortex (Morris et al., 2000). The auditory cortex develops with tonotopic cell columns that represent different CFs, where adjacent columns are organized from low frequency to high frequency moving posteriorly to anteriorly; this is consistent with the tonotopic representation found in the cochlea and auditory pathways (Graven & Browne, 2008).

Table 1-1: Summary of the Major Stages in Human Cochlear Development

Developmental event	**Time of emergence in weeks after conception**
Hair cell differentiation	not known

Afferent eighth nerve fibres in cochlear epithelium	10
Inner hair cells histologically distinguished	10 to 12
Outer hair cells histologically distinguished	12+
Synapses between hair cells and afferent eighth nerve fibres	11 to 12
Development of stereocilia on inner hair cells	12
Development of stereocilia on outer hair cells	12+
Efferent eighth nerve fibre endings below inner hair cells	14
Maturation of inner hair cells eighth nerve synapse	15
Onset of hearing function by structural criteria	20
Efferent synapses with outer hair cells	22
Maturation of outer hair cells eighth nerve synapse	22
Stereocilia maturation (inner and outer hair cells)	22
Outer hair cells and related structures appear mature	30
Normal (mature) auditory sensitivity and frequency resolution	not known
Adapted from Hall, (2000); Pujol & Lavigne-Rebillard (1992)	

To develop a healthy auditory system, auditory stimulation is needed starting at the 28 to 40 weeks of gestation and continuing for several years after birth. Around 28 to 29 weeks after conception, the hair cells and their connections in the cochlea are sufficiently mature to begin tuning for specific frequencies. The hair cells for the lower-frequency sounds are tuned while the hair cells for the high-frequency develops later after birth. The fetus is protected from most high-frequency in utero sounds as the environment in the uterus filters out high frequency ones, and allows for the recognition and response to internal and external sounds (Graven & Browne, 2008). While in the womb, the fetus can learn different sounds including mother's voice and simple melodies, and is able to discriminate it from others after birth. This has been demonstrated at as early as 32 weeks of gestation (Moon & Fifer, 2000).

1.5.2 Development of Speech Perception

Children acquire certain aspects of language in the first days of life and even before birth (Dehaene-Lambertz, Hertz-Pannier, Dubois, & Dehaene, 2008; Hirnholz & Benacerraf, 1983; Mehler et al., 1988; Querleu, Renard, Versyp, Paris-Delrue, & Crèpin, 1988). Mehler et al. (1988) showed that infants expressed a preference for languages they were exposed to in the uterus. In fact, studies have shown that new-borns' speech perception is developed in many aspects including phoneme categorization and identification of abstract phonemes, in addition to their ability to discriminate between languages belonging to different rhythmic families (Dehaene-Lambertz et al., 2008; Nazzi, Bertoncini, & Mehler, 1998; Nazzi & Ramus, 2003; Querleu et al., 1988). As infants grow, counts of neural synapsis is the highest between the first year and the fourth year of life (Huttenlocher & Dabholkar, 1997), perhaps to support the process of experience-expectant learning seen in acquisition of spoken language (Maurer & Werker, 2014; Werker, 2012). Even though cortical development accelerates rapidly following birth, neural pathways in cortical circuits start forming shortly before birth and continues until adolescence (Kral et al. 2016), while myelin sheaths maturation period extends into adulthood (Lin, Mula, & Hermann, 2012)

1.5.2.1 Effects of Sensory Loss

The brain is highly sensitive to sensory input and sensory loss since several processes influenced by the environment starts in utero (Hübener & Bonhoeffer, 2014; Kral, 2013). Since the human cochlea is functional from weeks 24–26 after conception, loss of cochlear receptors could lead to damage of subsequent auditory neurons in the brainstem (Tong et al., 2015), affecting the functional integrity of auditory pathways as it appears to be dependent on the age at onset of sensory deficits, jeopardizing higher order functions in the brain (Kral et al. 2016). During the critical period where that neuronal connections peaks, hearing deficits may reduce functional maturation, postpones cortical synaptogenesis, and accelerate synaptic elimination (Kral & O'Donoghue, 2010; Kral & Sharma, 2012). Eventually, disturbing central functions such as intensity coding, cortical column functioning, cochleotopic representation, representation of auditory space, and corticocortical interactions including top-down control and auditory object formation (Kral, 2013).

1.5.3 Neuroplasticity

The brain is a dynamic self-organising system that is shaped by recurrent exposure to stimulation from the environment (Kral et al., 2016). Neuroplastic changes occur following experience with a sensory stimulus that starts in utero and continues after birth. Experience helps to optimise connectivity in the brain and is dependent on synaptic plasticity, by which existing neural connections are strengthened or weakened and new synapses are formed or eliminated (Kral et al., 2016). During these neuroplastic changes, the brain may delete neural connections that are no longer useful (pruning) and strengthen the necessary ones. Neuroplasticity changes have been measured in response to auditory training (AT) and have shown that neural pathways and synapses can be affected by training. In fact, studies have shown that neural responses to sound change through rigorous listening (Tremblay, Kraus, McGee, Ponton, & Otis, 2001; Tremblay, Shahin, Picton, & Ross, 2009), suggesting that AT may optimise neural activation and in turn improve auditory perception and listening skills and reduce functional deficits (Kraus & Chandrasekaran, 2010).

1.6 Effect of Hearing Loss on Children's Speech and Literacy

Speech impairment in children is often associated with delays in the development of phonological awareness and printed word-decoding skills (Bird and Bishop1992, Larivee and Catts 1999, Carroll and Snowline 2004). This is because those children have difficulty accessing the underlying phonological representations of words (Bird et al. 1995, Swan and Goswami 1997, Rvachew et al. 2003, Nathan et al. 2004). Phonological representation refers to the storage of phonological information, phoneme or combination of phonemes that comprise a word, in long-term memory. A well-specified and easily accessible phonological representation leads to accurate phoneme production skills in addition to phonological awareness and reading skills e.g. unfamiliar words (Elbro 1996).

Hearing loss can hinder auditory and speech development. Inability to access sound to accurately map between the acoustic signals and their corresponding phonemes to create accurate phonological representations of words leads to delays in vocabulary growth, the development of syntax and matching of phonemes and graphemes in alphabetic languages. Children with hearing loss tend to have poorer literacy skills than their normal-hearing peers (Werfel, 2017). Hearing devices help children with hearing loss to access sound and cochlear implantation have shown to improve children's spoken communication skills, educational achievements and quality of life when children were implanted before the age of 5 years (Stacey, Fortnum, Barton, & Summerfield, 2006). Nevertheless hearing-impairment school-leavers read at a median of 3rd to 4th grade level (8-9 years old) at the age of 18 years, this level did not changed since the 1970s (Qi & Mitchell, 2012).

Studies also have shown that children with CI performed poorer than their hearing peers on decoding (Geers, 2003; Geers & Hayes, 2011; Wass et al., 2019), vocabulary (Coppens, Tellings, Verhoeven, & Schreuder, 2013; Dillon, de Jong, & Pisoni, 2012; Fagan & Pisoni, 2010; Geers, Moog, Biedenstein, Brenner, & Hayes, 2009; Walker, Sapp, Oleson, & McCreery, 2019), spoken language comprehension (Geers et al., 2009), and phonological and complex working memory (Wass et al., 2008). Furthermore, more than half of adolescents with CIs scored lower than normal limits on measures of reading and writing (Geers & Hayes, 2011). Geers, Moog, Biedenstein, Brenner, & Hayes, (2009) investigated vocabulary and language skills in 153 children with CIs (range of age 4.11-6.11 years) who received auditory-oral intervention during the preschool years. The study revealed that only 50% of the children received age-appropriate scores on measures of receptive vocabulary, 58% on measures of expressive vocabulary, 46% on measures of verbal intelligence, 47% on measures of receptive language, and 39% on measures of expressive language. The study have shown that a bout 50% children with CIs were delayed (by 1 standard deviation or more) compared to their age-matched normal-hearing counterparts regardless of the language skill assessed. This result should be interpreted as meaning that half of the children who were born with severe-to-profound hearing loss had languages abilities in the lowest 15th percentile rankings for the measure (Young & Kirk, 2016). Such level of literacy negatively influences the quality of life of those children and place their future at risk as they struggle to successfully achieve their potential.

Low literacy achievement would have a strong negative effect on children’s future. Adults with low literacy achievement are less likely to have full-time jobs compared to those with average literacy achievement (Kutner et al., 2007). In fact, it has been reported that only half of individuals with hearing loss are employed post high school (Newman, Wagner, Cameto, Knokey, & School, 2009).

1.7 LEARNING FROM PAST EXPERIENCE OR EXPOSURE

1.7.1 Hebbian Learning

Hebbian learning (Hebb, 2005) is one of the learning models which hypothesizes that the strength of the connection between neurons in the brain will be stronger after initial communication as the neural network would be reorganized post initial and recurrent exposures. Considering this on a sensory input model, a stimulus would generate a particular pattern of activity in sensory input neurons which in turn will provoke a particular percept in neurons further downstream. In addition, the connections from the sensory input neurons to the activated downstream neurons should be stronger post initial stimulation. Multiple exposure to the same stimulus will increase the tendency for a familiar input to elicit the same output on a succeeding event.

Perception of /r/ and /l/ in native Japanese speakers is an example of this phenomena. The English sounds /r/ and /l/ are all mapped to the same (/l/-like) percept, thus the English /r/ or /l/ input in Japanese speakers would elicit the same percept (/l/-like) (Miyawaki et al., 1975). When native Japanese speakers underwent adaptive AT for discrimination between the sound /r/ – /l/, subjects showed clear evidence of learning using neuroimaging, and several indicators suggested that training affects speech perception rather than simply auditory processes (McClelland, Fiez, & McCandliss, 2002). This example also can be explained by the statistical learning theory linked to language acquisition in infants (Romberg & Saffran, 2010). Like the Hebbian learning model, the statistical learning theory relies on the ability of humans to extract statistical regularities from the environment around them and learn from their past experiences or exposures. Both theories reinforce the concept of neuroplasticity and the effects of feedback on learning.

1.7.2 Principles Guiding AT

The transfer-appropriate processing (TAP), a meaning-based orientation, and the affective filter hypothesis are the three main theoretical underpinnings for AT (Tye-Murray, 2019, p. 109). TAP states that the greater the overlap between the training task and the desired outcome, the greater the benefits of the AT intervention. The meaning-based orientation theory suggests using tasks that activate the regions of the brain that process semantic contents as it requires the participants to engage in meaning-related processing, similar to real-word communication. The affective filter hypothesis recognizes that emotions play a critical role in new language acquisition. It states that affective filter may block input that would allow an individual to progress in acquiring new language. Similarly, if an individual with hearing loss is suffering from high level of anxiety due to continuous failure in recognition of casual speech or increased listening effort during everyday conversation, listening skills are unlikely to improve. In this case its recommended to individualize their AT to reduce anxiety and enhance their self-esteem.

1.8 AURAL (RE)HABILITATION

Processing and interpretation of the auditory input differs between CI recipients and can be influenced by number of factors some of which are recipients-dependent while others are technology-dependent (section 1.1.4.4). Another important factor that also has shown to influence outcomes of CI is aural (re)habilitation. Aural (re)habilitation includes services for adults and children with hearing impairments and their family that involve a variety of disciplines including audiology, education, and speech-language pathology (Tye-Murray, 2019, p. 10). The aim of those services is to maximize patients' outcomes; it starts with diagnosing hearing loss and fitting of hearing devices and it expands to AT, patient and family counseling, and patients and family psychosocial support.

Effective aural rehabilitation plans for children with hearing loss takes a holistic approach that starts with appropriate diagnoses of hearing loss then fitting the child with the needed hearing device to provide effective auditory, speech and literacy interventions. As a result, children can enhance their quality of life and avoid deficiency in language and reading comprehension, which have shown to increase the risk of emotional and behavioural difficulties at school during teenage years (Stevenson et al., 2018).

Aural rehabilitation in the context of this thesis focuses specifically on AT intervention. Studies have shown that AT can improve auditory and speech perception in children with CI (Rayes, Al-Malky, & Vickers, 2019) and can promote auditory plasticity in children with CIs by optimizing neural activation, and in turn improving auditory perception, listening skills and reducing functional deficits (Kraus & Chandrasekaran, 2010).

1.8.1 AT to Improve Speech Outcomes

AT aims to teach the brain how to make sense of sounds and to map discriminable components to internal representations of specific sounds. It requires listeners to actively engage in processing the auditory input, and through a process of repetition and variation of stimuli, it should promote the development of sound representations and in turn improve auditory perception (Schow & Nerbonne, 2006). AT have shown to help children with CI to access subtle spectral−temporal cues (Fu & Galvin, 2008), improve speech-in-noise performance (Mishra, Boddupally, & Rayapati, 2015), and develop sound identification and discrimination perception including phonetic discrimination skills (Roman, Rochette, Triglia, Schön, & Bigand, 2016).

AT can follow a bottom-up (analytic) or top-down (synthetic) methodology. Bottom-up method uses context-free acoustic or phonetic signals to train the listener to decode the auditory input without any context. Stimuli that are using in bottom-up training include none-sense words, syllabic structure, vowels, and initial or final consonant difference. Bottom-up approaches help the listener to discriminate and identify small differences between sounds, for example, pitch changes, or different phonemes. Many authors have had success with this approach for training specific distinctions (Fu, Galvin, Wang, & Nogaki, 2004; Stecker et al., 2006).

In contrast, top-down training methods utilize the listeners' linguistic knowledge to fill in the gaps in the sensory input provided by their hearing device. Examples of top-down training include matching spoken words to pictures which requires access to the stored representations of words in their lexicon, speech or reading comprehension, and music training.

Benefits of this approach was demonstrated in many studies and were even suggested to be more beneficial than bottom-up approach (Rubinstein & Boothroyd, 1987; Sweetow & Palmer, 2005). However, recent studies (Rayes et al., 2019) have shown benefits of both approaches help AT to develop both auditory and cognitive processes in children with CI to improve their listening and communication abilities.

1.8.2 Parental Engagement

Active family involvement and parent engagement affect children's auditory perception and language development and should be a major component of AT intervention (Moeller, 2000; Yoshinaga-Itano, 2003). Regardless to the mode of communication, effective interaction between parents and their deaf children is essential to the children's emotional and language development (Meadow-Orlans, Sass-Lehrer, & Mertens, 2003). Moeller (2000) examined the relationship between age of enrolment in an intervention programme and language outcomes in children with hearing loss and their findings were consistent with previous studies (Yoshinaga-Itano et al., 1998), which revealed that children with hearing loss who received intervention at earlier ages had better language outcomes than children who started intervention at later ages. The authors observed that parents or caregivers who were actively engaged in the early intervention process resulted in children with the highest language scores even if the identification of hearing loss or intervention services started relatively late. Moreover, a meta-analysis (Roberts & Kaiser, 2012) that investigated the effectiveness of parent-led language interventions on children's language skills revealed a significant positive impact on receptive and expressive language skills of children.

1.9 Children with Hearing loss in Saudi Arabia

Saudi Arabia has a higher prevalence of hearing loss in paediatric population than global prevalence. Global prevalence of hearing impairment ≥35 dB HL in children was estimated to be 1.2%, while prevalence of sensorineural hearing loss (SNHL) was 1.5% and severe to profound hearing loss was 0.7% (Al-Shaikh & Zakzouk, 2003; Al-Shaikh, Zakzouk, Metwalli, Dasugi, & Metwalli, 2002). Inevitably, the incidence rates of hearing loss are increasing over time (A Al-Abduljawad & Zakzouk, 2003) and the number of paediatric candidates for hearing devices is growing.

There are many studies that reported an association between hearing loss and degraded speech perception and literacy development in children (section 1.6). Such relationship was also shown to influence children's psychological stability and well-being. In fact, early detection and access to intervention improve the speech and language outcomes of children with hearing loss (Kennedy et al., 2006; Moeller, 2000; Yoshinaga-Itano et al., 1998, 2018). Generally, aural rehabilitation services, and AT specifically, have shown to have such effects. In addition, appropriate parental counselling including comprehensive understanding of the diagnostic tests is essential as it was shown to influence acceptance of hearing devices and improve compliance of parents with recommended interventions. (Mehta, Mahon, Watkin, Marriage, & Vickers, 2019).

There is no sufficient information about outcomes of hearing loss on children in Saudi Arabia. In general, epidemiological data on communication disabilities including hearing loss in Saudi Arabia is lacking (Manal A Khoja & Sheeshah, 2018). A survey that was conducted in Makkah province estimated the prevalence of functional disabilities among children to be 3.6%, where speech disorders was ranked amongst the most common disabilities (Milaat et al., 2001). Parents participated in the survey reported a lack of accessibility of (re)habilitation services for their children, and only one-third of the families participated in the survey received some sort of intervention. Accordingly, aural rehabilitation services in Saudi Arabia for children with hearing loss needs attention. It is critical to take an action to optimize speech and literacy outcomes for children with hearing loss to promote higher academic and professional success. In efforts to improve outcomes of hearing loss in Saudi Arabia, newborn hearing screening programmes have been recognized as a mandatory service that need to offered to all newborns but early intervention and rehabilitation services post implantation or hearing devices fittings are still not mandated. This project demonstrates a step toward enhancing outcomes for children with hearing loss who use CI in Saudi Arabia by developing an AT intervention that would be available for all and assessing its outcomes.

1.10 RATIONALE AND SIGNIFICANCE OF THE STUDY

Hearing loss can have a significant impact on children’s speech and language development and quality of life. Even though, CIs have significantly contributed to minimizing the burden of hearing impairment by allowing deaf children to access sound. There are limitations in the sounds delivered through CIs due to restrictions caused by the signal processing. The brain adapts to some extent to optimise the encoding of the signal but the outcomes for children with CIs does not reach the levels of their normal hearing counterparts. This research looks at an approach for aural intervention to help the brain maximise the information provided. The research was specifically focussed on children with CIs in Saudi Arabia. Fortunately, children in Saudi Arabia have good access to CIs at an early age ever since the Saudi High Authority mandated national neonatal hearing screening in 2014. However, follow up and aural rehabilitation services post identification of hearing loss are lacking (Alyami et al., 2016; Milaat et al., 2001). Hence, the motivation behind this project was to develop an intervention that could be used by children with CIs and their families. The intervention is called the Hearing Intervention Battery in Arabic (HIBA) and it is an approach that can be implemented at home helping to overcome the difficulties of accessing aural rehabilitation services.

There are several essential areas that can be enhanced by this research. Firstly, HIBA can be available for clinicians as a validated (re)habilitative tool that can be recommended to patients enforcing aural rehabilitation practice with Arabic speaking paediatric CI recipients. Secondly, HIBA is not dialect-dependent intervention and that makes it applicable for all Arabic speaking children. Thirdly, HIBA can offer many research opportunities in aural rehabilitation and in mandating aural intervention services in Saudi Arabia. Finally, this project produced a validated outcome measure.

1.11 AIMS AND RESEARCH QUESTIONS

The aim of this project was to investigate whether HIBA, a multi-modal auditory training programme, can improve auditory outcomes in Saudi-Arabic speaking children with CIs. To address this, three projects were conducted. The first was a systematic review of the literature to assess the efficacy of AT interventions for paediatric CI recipients and answered the following questions:

a) Does AT lead to improvements in speech and language, cognition, and/or quality of life in children with CIs?
b) Which AT approaches (top-bottom, bottom-up or combined approach) is more effective for improving outcomes in implanted children?
c) Are improvements in speech and language, cognition, and/or quality of life retained over a period of time post AT intervention?

d) Do improvements in trained tasks generalize to other domains or transfer to untrained tasks?

The second project led to the development of a phoneme discrimination test because of the limited range of validated speech tests available in Arabic. This project answered the following questions.

a) Are the phoneme discrimination test materials that were developed appropriate for the target population to assess speech perception skills?
b) Can the speech materials be organised to create multiple lists with different levels of difficulty?
c) Is the developed test reliable?

The third project was the evaluation of the HIBA multi-modal auditory training programme in a randomised control trial with an active control condition (art activities). It answered the following questions:

a) Would HIBA lead to greater improvements in speech and pitch perception in the intervention group compared to the control group?
b) Would the benefits of HIBA transfer to untrained tasks (speech-in-noise perception and phoneme discrimination)?
c) Would improvements, if any, remain post intervention?

1.12 OVERVIEW OF THE THESIS

This thesis is organized into five chapters. Following the introduction, chapter 2 presents a systematic review of AT in paediatric CI recipients that was published in the journal of speech, language and hearing research (Rayes et al., 2019). It was conducted to understand whether AT is a potential intervention that may lead to improved auditory and speech outcomes. This review critically appraised the AT literature and was beneficial in terms of designing the intervention, developing the training materials, determining the delivery approaches, and selecting outcome measures that could be used to appropriately assess potential benefit.

Chapter 3 outlines the development and validation of a new Arabic speech discrimination test. There is a scarcity of auditory assessment tools in Arabic so an Arabic version of the CAPT (Vickers et al., 2018) was developed. This test produced two validated closed-set lists that varied in difficulty that can reliably assess consonant perception in children. The process of developing and validating this outcome measure is explained.

Chapter 4 described the HIBA intervention study. This research took place in Jeddah Saudi Arabia. Children with CI were recruited via speech and hearing clinics and social media. After parents responded to the adverts and agreed to participate in the study, baseline assessments were conducted twice (double-baseline) to account for learning effects in the outcome measures. The children were randomly assigned to an active control (art and craft activities) or an experimental (HIBA) group. Both groups took part in a four-week intervention programme. Assessments were repeated following the four weeks training period.

Chapter 5 discuss the main findings of this thesis, its impact and limitations, and describe future extended work.

CHAPTER 2: SYSTEMATIC REVIEW OF AUDITORY TRAINING IN PAEDIATRIC CI RECIPIENTS

Rayes H, Al-Malky G, Vickers D. Systematic Review of Auditory Training in Pediatric Cochlear Implant Recipients. *Journal Speech Language Hearing Research*. 2019;62(5):1574-1593. doi:10.1044/2019_JSLHR-H-18-0252

2.1 OVERVIEW

This chapter presents our published systematic review which evaluated the published research in AT for paediatric CI recipients. This review evaluated a variety of research designs and summarized essential considerations for a high-quality intervention study. The purpose of the research was to investigate whether AT in children with CIs leads to improvements in speech and language development, cognition, and/or quality of life; and whether improvements, if any, remain over time post AT intervention. In this chapter, benefits of AT were illustrated through the improvement in trained tasks in all the reviewed studies. Transfer of improvement to other domains and also retention of benefits post AT were evident when assessed, although rarely done. However, higher quality evidence to further examine outcomes of AT in paediatric CI recipients is still needed.

2.2 INTRODUCTION

Audibility or access to sound is only the first step of many that result in effective communication for hearing device users (Sweetow & Palmer, 2005). Kiessling et al. (2003) noted that audition is an essential component in aural communication, but it does not guarantee effective interaction. Instead they suggested sequential stages that lead to successful communication namely, hearing, listening, comprehension and finally communication.

CIs have been an extremely successful intervention for children with severe-to-profound hearing loss, helping to restore access to sound (Markman et al., 2011; Pulsifer, Salorio, & Niparko, 2003). However, large variability in auditory, speech, and language outcomes post implantation has been observed (Kane, Schopmeyer, Mellon, Wang, & Niparko, 2004; Niparko & Blankenhorn, 2003; Niparko et al., 2010). Average speech recognition outcomes are reported to be similar across different CI systems, however within-device-variation can be large across individuals (Firszt et al., 2004); suggesting that observed variation is recipient-dependent (Blamey et al., 2015; Finley et al., 2008). There are various factors that affect speech and language outcomes post-implantation. The main factors that have been identified for predicting word recognition scores in adult CI recipients are duration of deafness and duration of CI device use, where the shortest duration of deafness and longest CI device use lead to highest word recognition scores (Blamey et al., 1996; Friedland, Venick, & Niparko, 2003; Rubinstein, Parkinson, Tyler, & Gantz, 1999). For paediatric CI recipients, the main factors predicting CI outcomes are age of implantation, residual hearing before implantation, parent-child interactions, socioeconomic status (Niparko et al., 2010), and language acquisition status prior to cochlear implantation (prelingual or postlingual) (Kane et al., 2004). Children with CI progressed exceptionally well when they were postlingualy deaf, implanted at younger age, had residual hearing before implantation, and belonged to supportive and highly-motivated parents who were amongst the higher socioeconomic families. Other general factors predicting CI recipients' speech or language performance post implantation include electrode coupling (Mens & Berenstein, 2005; Pfingst, Franck, Xu, Bauer, & Zwolan, 2001), signal processing approach (Nogueira, Litvak, Saoji, & Buchner, 2015; Skinner et al., 2002; Wilson et al., 1988), quality of CI fitting (Holden, Vandali, Skinner, Fourakis, & Holden, 2005; Skinner, 2003), and age at implantation (Blamey et al., 1996; Connor, Craig, Raudenbush, Heavner, & Zwolan, 2006). Other factors, known to vary across subjects but not yet shown to influence speech recognition ability significantly, include spiral ganglion cell survival (Khan et al., 2005; Nadol, Young, & Glynn, 1989; Seyyedi, Viana, & Nadol, 2014) or morphological changes in surviving ganglion cells (Briaire & Frijns, 2006), and compromised central pathways (Kral, Kronenberger, Pisoni, & O'Donoghue, 2016; Shepherd & Hardie, 2001; Shepherd, Hartmann, Heid, Hardie, & Klinke, 1997)

Some of these factors such as CI-fitting approach and parameters for the sound-processing strategy have the potential to be improved; however other factors are out of the control of the clinician e.g. home language and family engagement. In addition, in some cases the sound may be delivered through the auditory system but the individual needs support to make effective use of the sound, and to this end AT programmes may help.

2.2.1 Auditory Training

AT is a sound based habilitative intervention aimed at improving individuals' speech and hearing skills through varied listening exercises (Sweetow & Sabes, 2006). AT aims to teach the brain to make sense of sound contrasts through repetition and variation of stimuli together with effective feedback. This way the listener habitually learns to distinguish between sound contrasts (Schow & Nerbonne, 2007).

AT is a potential intervention that can be used to maximize benefit from hearing devices. Although hearing devices may help people with hearing loss to access sound, it cannot enhance their ability to listen and comprehend what they hear. Changes in brain organisation to some extent can lead to improvements over time but the rate of change and potentially the maximum level of performance achieved can be modified with AT (Sharma, Purdy, & Kelly, 2009). Outcomes of AT have been assessed by measuring improvement in trained tasks and by improvement in different tasks that were not included in the training session. A review of AT research in adult CI users reported improvements in trained tasks, however generalisation of the trained tasks to other learning domains that were not targeted within an intervention, and retention of any benefits thereafter remain unproven (Henshaw & Ferguson, 2013).

2.2.2 Analytic (bottom-up) and Synthetic (top-down)

Approaches of AT are mainly divided into two types, bottom-up (analytic) and top-down (synthetic). Analytic approach uses a context-free acoustic-phonetic signal; it trains the listener to decode the speech signal without any context, such as syllabic structure, vowels, and initial consonant difference. Whereas the synthetic approach relies on the listeners' linguistic knowledge (e.g. semantic, syntactic, lexical, and phonological) to fill in the gaps in the sensory information provided by their hearing device. An example of synthetic AT includes connected discourse tracking (De Flippo & Scot, 1978).

One of the earliest studies in AT (Rubinstein & Boothroyd, 1987) where a group of adults with mild-to-moderate sensorineural hearing loss received only synthetic training and another group received both synthetic and analytic training reported that the inclusion of analytical training did not lead to further improvement in listening skills since a significant improvement was found with synthetic training alone. Furthermore, Sweetow and Palmer (2005) reviewed studies between 1970-1996 to evaluate AT in adults with hearing loss and assessed its effectiveness in improving communication and concluded that synthetic training could enhance speech recognition abilities, whereas the effectiveness of analytic training was not clear. Contrary to such views, Fu and colleagues (Fu, Galvin, Wang, & Nogaki, 2004; Fu & Galvin, 2008; Galvin, Fu, & Shannon, 2009; Zhang, Dorman, Fu, & Spahr, 2012), conducted many experiments using analytic training approaches with adults with CIs, and demonstrated significant improvements in the subjects' phonemic contrast scores and word recognition after training. Recent evidence recommends combining the two approaches to achieve maximum benefit (Amitay, Irwin, & Moore, 2006). Tye-Murray et al. (2012) used both approaches for AT with stimuli ranging from basic phonemic discrimination to comprehension of extended passages and they reported significant improvement in all trained tasks. Overall, a trend toward combining analytic and synthetic training is evolving throughout the literature as a means to achieve maximum benefit from this intervention.

2.2.3 Trained-task Performance and Generalization of Benefits

Reports of improvement in trained tasks post AT intervention in both hearing aid and CI users are positive. Henshaw and Ferguson (2013) systematically reviewed AT studies published from 1996 up to 2011 for adults with hearing loss. Their review stated that improvement in trained tasks was consistently reported whenever they were assessed. Only one study, which trained adult-CI recipients, reported a trend in improvement on the trained task rather than showing a significant improvement (Stacey et al. 2010).

Reports of learning transfer or generalization of benefits post AT are varied. Henshaw and Ferguson (2013) reported a significant but small improvement in generalisation of learning to untrained measures including speech intelligibility, cognition and self-reported hearing abilities. For example, Burk et al. (2006) reported that word-training programmes generalised to improvements in untrained words and to untrained speakers of trained words but did not generalise to trained words used in sentences. Zhang et al. (2012) also reported post-training improvements in the intelligibility of untrained vowels, consonants and words, but not in untrained sentences; the degree of improvement was larger in subjects with normal hearing compared to those with hearing loss. When training communication strategies along with syllable recognition, Kricos and Holmes (1996) observed improved performance post active-listening training, and skills were transferred to speech-in-noise conditions that were not included in the training. Communication strategies that were included in the training programme include encouraging active listening, showing interest while others are talking, using eye contact and body language, filling in the gaps for words not heard clearly based on the context of the conversation, replying with a statement summarizing whatever the speaker said, and accepting corrections readily.

2.2.4 Retention of Benefits Post AT

Retention of benefits or maintaining improvements over time is measured by comparing the performance of the subjects at baseline and after the training regimen has ceased on trained tasks and/or non-trained tasks. Henshaw and Ferguson (2013) indicated that 8 out of the 13 articles that were reviewed assessed retention at follow up assessments ranging from 4 days to 7 months post training. For instance, Burk, Humes, Amos, and Strauser (2006) reported that word recognition performance was significantly improved six months after training compared to baseline, whereas Oba, Fu, and Galvin (2011) reported sustained performance on digit recognition up to one-month post training. In addition, Stecker et al. (2006) and Burk and Humes (2008) reported significant improvements on a Nonsense-Syllable Test (NST) (Dubno & Levitt, 1981) and both easy and hard real-word recognition tests up to 7 weeks post AT.

Retention was not only limited to trained tasks, it was also measured in other tasks that were not included in the training intervention. For example, Sweetow and Sabes (2007) reported that post training improvements were maintained for all measures including Quick Speech–in-Noise Test (QuickSIN) (Killion, Niquette, Gudmundsen, Revit, & Banerjee (2004), and Hearing-in-Noise Test (HINT) (Nilsson, Soli, & Sullivan (1994), Hearing-Handicap Inventory for Elderly (HHIE) (Ventry & Weinstein,1982), Hearing-Handicap Inventory for Adults (HHIA) (Newman, Weinstein, Jacobson, & Hug,1990), and the Communication Scale for older Adults (CSOA) (Kaplan, Bally, Brandt, Busacco, & Pray,1997) questionnaires up to four weeks post training. However, this improvement can be attributed to test-retest affect as alluded to by the authors. In a different study, Oba et al. (2011) controlled for this confound by comparing subjects' performance immediately post training and at 4 weeks follow up and reported no significant change. Therefore, Oba et al. (2011) suggested that subjects improved performance in both HINT and Institute of Electrical and Electronics Engineers (IEEE) (IEEE, 1969) sentences in steady noise and in multi-talker babble were a clear evidence of AT retention.

2.2.5 Brain Plasticity as Evidence of AT

Neuroplasticity changes have been investigated as evidence of AT and have shown that neural pathways and synapses can be affected by training. In fact, studies have shown that neural responses to sound change through rigorous listening (Tremblay, Kraus, McGee, Ponton, & Otis, 2001; Tremblay, Shahin, Picton, & Ross, 2009), suggesting that

AT may optimise neural activation and in turn improve auditory perception, and listening skills and reduce functional deficits (Kraus & Chandrasekaran, 2010).

The question is which parts of the brain are being affected by AT? Electroencephalography (EEG) and magnetoencephalography (MEG) have been used to explain how AT exercises might affect the brain. These techniques determine the time course and the occurrence of cortical and sub-cortical modulations as a response to a stimulus, which is related to the particular AT goal (Barrett, Ashley, Strait, & Kraus, 2013; Brattico, Tervaniemi, & Picton, 2003; Shahin, 2011; Tremblay, Inoue, McClannahan, & Ross, 2010; Tremblay et al., 2009).

The P1-N1-P2 waves of the cortical auditory-evoked response (AEP) measured with EEG consistently showed increased gain in P2 amplitude post AT (Shahin et al., 2003; Kuriki et al., 2007; Seppänen et al., 2012; Kühnis et al., 2013). Despite the emerging evidence that improved perception is reflected by increased amplitude of the P2 wave of the P1-N1-P2 complex, not much is known about the neural generators of the auditory P2 response. Ross and Tremblay (2009) showed that the centre of activity for P2 to be in the anterior auditory cortex, but how it relates to learning is still unidentified (Tremblay, Ross, Inoue, McClannahan, & Collet, 2014).

Other studies have examined P1 cortical AEP latencies in relation to cortical maturation in response to sound (Bauer, Sharma, Martin, & Dorman, 2006; Ponton, Don, Eggermont, Waring, & Masuda, 1996). The auditory thalamic and cortical sources generate P1 responses that vary with chronological age. Accordingly, P1 latency has been used to infer the maturational status of auditory pathways (Bauer et al., 2006; Sharma et al., 2005). The rapid decrease in P1 latency post cochlear implantation is speculated to reflect central auditory plasticity (Sharma, Dorman, Spahr, & Todd, 2002; Sharma, Dorman, & Spahr, 2002; Sharma et al., 2004).

Anderson and Kraus (2013) established that there are brain plasticity changes in two distinct ways: short and long-term plasticity. Language reflects long-term plasticity whereas AT exercises relate to short-term plasticity. Jeng et al.'s (2011) study investigated the difference between Chinese and American speakers' pitch representation at the level of the brainstem. The study revealed that brainstem encoding of linguistic pitch contours was enhanced in Chinese adults compared to American adults reflecting the outcome of long-term linguistic experience in each group. The study also suggested that tuning features of neurons along the pitch axis with enhanced sensitivity to linguistically relevant variations in pitch are sharpened by long-term experience (Krishnan, Xu, Gandour, & Cariani, 2005). Another example of neuroplasticity is bilingualism. Krizman, Marian, Shook, Skoe, and Kraus (2012) showed that a greater brainstem encoding of the fundamental frequency (F0), a feature known to underlie pitch perception and grouping of auditory objects, was greater in bilinguals compared to monolinguals.

An example of short-term brain plasticity has been observed in musical-training programmes. Growing evidence especially for normal-hearing listeners suggests that intersecting networks in the brain process acoustic features heard in music and speech, suggesting that musical training may generalize to neural encoding of speech, language and music (Anvari, Trainor, Woodside, & Levy, 2002; Besson, Schon, Moreno, Santos, & Magne, 2007; Herholz & Zatorre, 2012; Kraus, Skoe, Parbery-Clark, & Ashley, 2009; Patel, 2011). In Deaf children, a recent study showed evidence of improvements in executive function following a five week music-training intervention (Manson, 2017).

Further evidence confirmed that music skills significantly correlate with phonological awareness and reading (Anvari et al., 2002; Culp, 2017). It was proposed that actively listening to music by utilizing greater perceptual demands might further fine-tune the auditory system (Herholz & Zatorre, 2012; Ingvalson & Wong, 2013; Patel, 2011). Not only listening to music but also exploration of sound and singing was linked to improved pitch discrimination, speech perception in noise and singing competency in children with normal hearing and children with hearing loss (Welch et al., 2015).

2.2.6 Research Aims

The primary aim of this systematic review was to investigate whether AT is effective at improving performance scores for paediatric CI recipients. Performance measures were considered for speech and language, cognition, and quality of life abilities. Secondary aims were to evaluate the impact of different AT approaches (analytic versus synthetic) and to determine if improvements generalize to untrained tasks and assess the retention of benefit post AT. Ultimately, outcomes of this review will potentially help clinicians to make informed decisions related to AT with paediatric patients using CIs and provide researchers with the latest AT findings for paediatric CI recipients. This research aimed to answer the following questions:

- Does AT lead to improvements in speech and language, cognition, and/or quality of life in children with CIs?
- Is analytic or synthetic AT more effective for improving outcomes in implanted children?
- Do improvements in speech and language, cognition, and/or quality of life remain over a period of time post AT intervention?

- Do improvements in trained tasks generalize to other domains or transfer to untrained tasks?

2.3 METHODS

A systematic review protocol was prepared and registered with PROSPERO (2017: CRD42017057346) (Appendix I), the International prospective register of systematic reviews. Inclusion and exclusion criteria were established based on the Participants, Intervention, Control, Outcomes, and Study designs (PICOS) strategy (Richardson, Wilson, Nishikawa & Hayward, 1995) (See Appendix II).

Methods for the review were clearly stated in advance of the review and followed to ensure transparency and to avoid bias. The search was conducted using seven electronic bibliographic databases (MEDLINE, EMBASE, The Cochrane Library (Cochrane Database of Systematic Reviews, Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Methodology Register), CINAHAL, Scopus, PubMed, and Web of Science (Science and Social Science Citation Index)). Only studies published in English were included with no publication-period restrictions. Study designs that were included in this review were RCTs, non-RCT, cohort studies with control, or repeated measures. All AT interventions involving human or computer-based delivery in clinic, home, school or laboratory were included. Keywords used included: cochlear Implant, cochlear prosthesis, auditory training, auditory learning, and rehabilitation.

To minimize the risk of bias, two of the authors independently extracted and analysed the data based on several measures, including: randomization, blinding, controls, power calculation, selective reporting of outcome measures, training feedback, participants’ self-assessment and generalization of improvements if any. The third author was the moderator who reviewed the extracted and analysed data and discussed any inconsistencies or concerns. All retrieved papers went through three main stages: identification, screening and eligibility assessment. A total of 96 articles were extracted from the selected databases and from references therein. After removing duplicates, review articles and studies addressing different outcomes, only 19 remained. The 19 articles were carefully reviewed and only 9 matched the PICO criteria and were included in the review. The other ten articles (Barton & Robbins, 2015; Chen et al. 2010; De Bruyn et al., 2011; Fu, Galvin, Wang, Wu, 2015, Kant & Adhyaru, 2009; Rochette & Bigand, 2009; Rochette, Moussard, & Bigand, 2014; Perin da Siliva, Comerlatto Junior, Andreoli Balen, & Bevilacqua, 2012; Vongpaisal, Caruso, & Yuan, 2016; Zhou, Chai Sim, Tan, & Wang, 2012) were not included in this review due to failing to meet the inclusion criteria including irrelevant outcome measure, study design , or lack of controls. The articles were further evaluated and graded to assess their levels of evidence and control for bias (Figure 2-1).

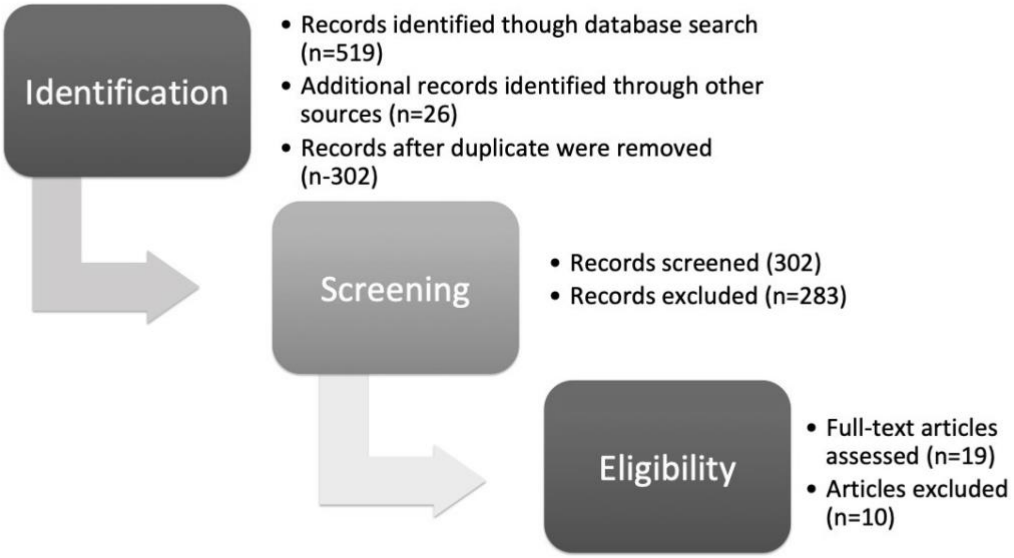


Figure 2-1: Process of paper selection that was followed in this systematic review

2.3.1 Quality of the Articles

All the selected studies were evaluated and graded to assess their levels of evidence following the guidelines from the 2004 Grading of Recommendations Assessment, Development and Evaluation (GRADE) Working Group guide (See Appendix III) (Atkins et al., 2004). Measures and criteria used in assessing the quality of the studies were adopted from Henshaw & Ferguson (2013). The level of evidence of each study was established based on a sum of scores that was given to each category within the general scientific measures and AT specific measures. General scientific measures include looking at the approaches for randomization and control groups, and explanation of the power calculation, blinding, and outcome measure reporting. AT specific measures include looking at the applicability of outcome measures selection, providing training feedback, assessing ecological validity (i.e. the location where AT was conducted e.g. in the home which better represents normal listening environment compared to an unnatural laboratory setting), complying with training protocols, and assessing retention of improvements. The score for each measure is either 0, 1 or 2. A score of 0 indicates faulty or lack of information to make an informed judgment, a score of 1 indicates weak information or absence of detail, and a score of 2 refers to proper use and comprehensive reporting. Scores for each study were summed to produce an individual study quality score, which is used to convey the level of evidence credited to each study. A low-level of evidence indicates that the results of the study are not repeatable, whereas a high-level of evidence suggests greater confidence in the findings (Henshaw & Ferguson, 2013).

2.3.2 Synthesis of Results

All the extracted data including study design, participant details, training protocol, outcome measures, and main findings were tabulated; then a summarised table was produced to answer the research questions, assess levels of evidence, quality of research and bias. Ideally, combined data would be subjected to a meta-analysis but as there was no commonality across studies for training stimuli, training protocols and outcome measures it meant that this was not possible.

2.4 RESULTS

The analysed studies and their findings were summarised in two tables. Table 2-1 describes the study design (design, number of subjects, participants' age, and training location), training stimuli, frequency of training sessions, outcome measures, and main findings. Table 2-2 summarizes the main findings including, improvement, retention, generalization of learning, and compliance.

2.4.1 Characteristics of the Studies

The participants in all of the studies were children with severe-to-profound hearing loss. Seven of the nine studies included only children with CIs or bimodal devices (CI and hearing aid), and only two studies (Welch et al., 2015; Wu, Yang, Lin, & Fu, 2007) included both children with CIs, bimodal devices and also children using hearing aids. Overall, the studies represented results from 89 CI and bimodal users and six hearing-aids users. Although our initial inclusion criteria were restricted to studies with CI users, it was necessary to relax this criterion to include a larger group of papers for analysis.

Participant sample sizes ranged from 9 subjects (Kronenberger, Pisoni, Henning, Colson, & Hazzard, 2011) to 29 subjects (Welch et al., 2015), (mean=19.67, SD=7.03). Only three studies utilized a repeated measures design (Kronenberger et al., 2011; Welch et al., 2015; Wu et al., 2007), and only one study (Welch et al., 2015) included children with normal hearing as a control group. The remaining studies utilized non-repeated-measure design that used two independent groups one as an experimental group and the other as a control group.

There were two RCTs (Ingvalson, Young, & Wong, 2014; Roman, Rochette, Triglia, Schön, & Bigand, 2016), four non-RCTs (Good et al., 2017; Hagr et al., 2016; Mishra, Boddupally, & Rayapati, 2015; Yucel, Sennaroglu, & Belgin, 2009) and three repeated measures (Kronenberger et al., 2011; Welch et al., 2015; Wu et al., 2007).

2.4.2 Quality of the Studies

Quality of the studies in addition to their level of evidence is listed in Table 2-3. Scores for each study were calculated based on a number of scientific measures and AT specific measures. The scientific measures include randomisation, controls, power calculation, blinding, and outcome measure reporting. Whereas, AT related measures included generalisation of learning, outcomes used, evaluation of functional benefit in real-world listening, training feedback, ecological validity measurement of compliance with training protocols, and long-term follow-up of improvements. This rigorous evaluation revealed that the level of evidence of all studies but one (Mishra et al., 2015) were low. A major factor affecting the quality of the studies was failing to meet the requirements for randomization, power calculation and/or blinding. An attempt to randomize was evident in four studies (Good et al., 2017; Ingvalson et al., 2014; Mishra et al., 2015; Roman et al., 2016), blinding in two studies (Hagr et al., 2016; Mishra et al., 2015), and power calculation in one study (Wu et al., 2007). In addition, lack of follow-ups post AT programme (Hagr et al., 2016; Ingvalson et al., 2014; Roman et al., 2016; Welch et al., 2015; Yucel et al., 2009), report of compliance (Hagr et al., 2016; Wu et al., 2007; Yucel et al., 2009), and training feedback (Good et al., 2017; Kronenberger et al., 2011; Roman et al., 2016; Yucel et al., 2009), which were evident across the studies, further reduced the overall quality score. Moreover, the lower scores of the quality of studies increased the risk of bias; such findings may degrade the confidence of clinicians when recommending AT to their patients.

2.4.3 Trained Skills and Outcomes of AT

Trained skills included working memory, speech perception, music, pitch and rhythm discrimination, and environmental sounds. Benefits of AT were clearly illustrated through the improvement of all trained tasks across all nine studies regardless of the duration of training which ranged from 4 weeks (Ingvalson et al., 2014) up to 2 years (Yucel et al., 2009), or type of training.

2.4.3.1 Working Memory with or without AT

Two studies used auditory and/or cognitive training materials; where AT focused on phonological-awareness skills (Ingvalson et al., 2014) and cognitive training focused on training working-memory skills (Ingvalson et al., 2014; Kronenberger et al., 2011). Kronenberger et al. (2011) used Cogmed Working-Memory Training (Klingberg et al., 2005) (CWMT; www.cogmed.com) to assess its effectiveness for improving memory and language skills in Children with CIs. Cogmed is a computer-based programme that exercises auditory and visuospatial memory or combined auditory–visuospatial short-term and working memory. The training led to an improved performance on most training exercises including verbal and nonverbal working-memory tasks. Even though improvement in working memory decreased after 1 month at follow-up, sentence repetition continued to show improvement up to 6 months. Such improvement that remained over a period of time post the AT intervention led the authors to suggest that working memory training might improve aspects of memory and language in children with CIs, but of course it is hard to tease apart the specific effects due to working memory and visual-spatial awareness.

Ingvalson *et al.* (2013) used Earobics ("Earobics: Auditory development and phonics programme [Computer software] ," 1997), which trains both phonological awareness and working-memory skills simultaneously through exercises for matching phonemes to graphemes; identifying target phonemes as initial, medial or final; recalling a sequence of drumbeats, identifying sound, phoneme, syllable and rhyme, and recognising speech perception in noise. The group of children who received the training showed significant gains on language measures post intervention whereas the control group did not. The authors suggested that phonological and working memory training in children with CIs may lead to improved language performance but it is hard to determine which aspects of the training were most influential.

2.4.3.2 Speech Stimuli and Environmental Sounds

Three of the nine studies used speech stimuli (in quiet and/or noise) to improve speech perception skills. Tasks were focused on detection, discrimination, and identification of speech sounds/words (Hagr et al., 2016), identification and discrimination of phonemes, vowels and constants (Wu et al., 2007), and recognition of digits in noise (Mishra et al., 2015).

Wu et al. (2007) investigated the impact of computer-assisted speech training on speech recognition performance of Mandarin-speaking children with hearing-impairment. Training stimuli included discriminating between phonemes and acoustic speech features in vowels (1st and 2nd formant frequencies and duration) and consonants (e.g. voice, manner and place of articulation). Children receiving the intervention showed significant improvements in vowel, consonant, and tone recognition. The authors suggested that moderate amounts of AT led to improvements in speech understanding in children with hearing loss.

Mishra et al. (2015) evaluated training speech in noise skills in children with CIs in which training used adaptive speech (mainly numbers) in a white/speech-shaped noise. The speech-in-noise recognition training used a customized version of “Angel Sounds” (Version 5.08.01, Emily, Shannon, Fu Foundation, Los Angeles, CA). Speech-in-noise performance improved in the group that received the intervention compared to the control group. The authors concluded that AT improves speech-in-noise performance in children with CIs.

In another group of children Hagr et al. (2016) assessed the effectiveness of "Rannan", an auditory-training programme developed for Arabic speaking children with CIs. The software provides computer-based exercises for sound detection and discrimination skills. Namely, sound-detection exercises use Ling sounds, environmental sounds, and phrases. In addition, supra-segmental discrimination exercises including stimuli that differ in intensity, duration, pitch, or intonation/stress/rhythm and rate, whereas segmental discrimination and association exercises including discrimination of words that differed in vowels, consonants, and number of syllables, and similar words were also available. The study showed that the group who received the Rannan computerized training intervention in addition to the basic aural rehabilitation programme, scored significantly higher on the Infant-Toddler Meaningful-Auditory-Integration Scale (IT-MAIS) parent questionnaire (Zimmerman-Phillips, Osberger, & Robbins, 2001) and Listening-Progress Profile (LiP) (Nikolopoulos, Wells, & Archbold, 2000) compared to control.

2.4.3.3 Music, pitch and rhythm discrimination, and environmental sounds

Four studies used non-speech stimuli such as environmental sounds, and music. Roman et al. (2016) assessed the impact of training on four main areas of auditory cognitive processing, namely identification, discrimination, auditory memory and auditory scene analysis (ASA) in children with CI using "sound in hand" apparatus (Rochette & Bigand, 2009). Sound in hand is a tool that looks like a mini keyboard but was specifically developed to assess different auditory cognitive skills. In the identification task, the subject listens to one sound and has to find the key that corresponds to it. In the discrimination task the subject listened to a continuous sound that can be modified by changing its pitch or duration and the subject determined if it is the same or different. In the auditory memory task, the subject is asked to imitate or recall a sequence of sound. In ASA task, the subject is familiarized with elements of the auditory scene, and then listened to a continuous auditory scene consisting of two or three different sources. Surreptitiously, removing one or two elements modifies the auditory scene and the subject has to identify the change that occurred. The authors reported a significant improvement in the identification, discrimination and auditory-memory tasks, but not in ASA task in the experimental group compared to the control group. In addition, improved performance was also transferred to phonetic discrimination skills.

Good et al. (2017) assessed the impact of music training (individual piano lessons) on various aspects of auditory processing in children with CIs. The study aimed to assess generalization of music training to other learning domains rather than assessing improvements of trained tasks. The children received individual piano lessons, which involved music theory and hands-on techniques such as playing musical scales, learning finger control, and hand position. In addition, subjects learnt a new song and rehearsed it. The authors reported improved scores on discrimination of melodic contour, rhythm, and memory for melodies in the experimental group compared to the control group. These improved skills were also transferred to improved emotional-speech-prosody perception.

In a slightly younger group of children, Yucel et al., (2009) trained pitch and rhythm perception and assessed the impact of training on speech perception. The musical training programme used electronic keyboards to improve pitch and rhythm discrimination and familiar melody recognition. By the end of the 2 years follow up, the experimental group had developed more rapidly than the control group in all aspects of musical skills assessed; a positive trend was noted for an improvement in open-set speech perception scores for the experimental group but the difference between the groups did not reach significance.

Finally, Welch et al. (2015) offered 20 weekly sessions of singing and vocal exploration training. Normal-hearing children and children with hearing impairment participated together in training exercises which aimed to teach them simple songs with actions, descending/ascending pitch glides, contrasting vocal timbres, explorations in visual imagery for sound, and mimicry of vocal patterns. The training had a positive impact on participants singing skills in terms of accuracy of singing simple songs as measured using the England National-Singing Scale (Welch, Saunders, Papageorgi, & Himonides, 2012). Overall, pitch perception also improved measurably over time for children, particularly for those with hearing loss. Findings imply that sustained age-appropriate musical activities can benefit all children, regardless of hearing status.

2.4.3.4 Retention of improved performance

Retention of benefits or sustaining of improvements is measured by comparing the performance of the subjects at baseline and after the training regimen has ceased on trained tasks and/or non-trained tasks. Mishra et al. (2015) investigated retention of improvements post training children with CI to recognize numbers in white noise and in speech-shaped noise, and subjects showed retained improvements up to three weeks post AT intervention. Kronenberger et al. (2011) also assessed the benefits of retention post working memory training in children with CI. Although language was not the focus of the training, retention of improvement in speech measures was retrained for up to 6 months whereas retention in working memory measure was retained for up to one month post training. Wu et al. (2007) trained discrimination of phonemes and acoustic speech features in vowels (1st and 2nd formant frequencies and duration) and consonants (e.g. voice, manner and place of articulation). The authors reported retention of improvement in all measures assessed (vowel, consonants and Chinese tone recognition) for up to 2 months post training.

2.4.3.5 Generalization and transfer of learning

Four of the six studies, which assessed generalization, reported transfer of learning to other skills. Good et al. (2017) demonstrated a transfer of learning from music training to emotional speech prosody perception. Accordingly, the authors concluded that music training can be an effective tool to be integrated in auditory-rehabilitation plan post cochlear implantation. Roman et al. (2016) showed a transfer of learning from identification and discrimination of non-speech stimuli such as environmental sounds and music to phonetic discrimination skills. Mishra et al. (2015) reported “near transfer” as learning effects were established and generalized to similar but untrained conditions. The trained tasks included number recognition in white noise and untrained task consisted of digit triplets in speech-shaped noise. Kronenberger et al. (2011) also observed generalization of learning from improved working-memory skills to improved language processing skills post working-memory training. The two studies that did not observe generalization of learning from music training to speech perception (Welch et al., 2015; Yucel et al., 2009) were both studies that were conducted over an extended period of time (longitudinal). Yucel et al. (2009) observed a transfer of learning in one of the speech measures but not in the other. Welch et al. (2015) did not report any transfer of learning but the authors acknowledged that resources were insufficient to allow focused singing training with children with hearing loss, and participants were a heterogeneous mix of CI users, HA users, and normal hearing children.

2.4.3.6 AT Approaches

2.4.3.6.1 Analytic (bottom-up) & synthetic (top-down)

When assessing the approaches of AT across the studies, we found that four studies used both analytic and synthetic approaches, and others used either one or the other. For instance, Mishra et al. (2015) used a combination of both analytic and synthetic approaches in their training programme. Detection and discrimination of acoustic differences between several speech tokens in noise reflects the analytic element of learning whereas the synthetic component involved listening to an accented speech that require more attention and higher level of language processing. Roman et al. (2016) also utilized both approaches training auditory memory, identification and discrimination of sound and ASA. Furthermore, Ingvalson et al. (2014) trained both phonological awareness skills and auditory working memory; phonological awareness exercises train mostly bottom-up skills whereas working memory exercises train top-down skills. Finally, Yucel et al. (2009) used both approaches training pitch discrimination, rhythm discrimination, and sequence repetition. All four studies reported improved skills on trained asks.

2.4.3.6.2 Synthetic (top-down) versus Analytic (bottom-up)

Five studies used just one approach, two studies used an analytic training approach and three a synthetic approach. Wu et al. (2007) trained discrimination using vowels and acoustic speech features such as formant frequencies and duration, in addition to discriminating between phonemes. Hagr et al. (2016) trained for detection of Ling and environmental sounds, discrimination between intensity, duration, pitch, or intonation stress, rhythm and rate, vowels, consonants, and number of syllables in words. Both studies reported improved skills on trained asks.

On the other hand, Good et al. (2017) utilized synthetic training in private piano lessons including musical theory, technical exercises eventually learning a song. Welch et al. (2015) also opted to use a synthetic-training approach where the training stimuli were singing exercises, vocal explorations, and explorations in visual imagery for sound. Finally, Kronenberger et al. (2011) trained working memory using Cogmed training software, which involved auditory, visuospatial, and combined short-term and working-memory skills. All approaches resulted in an overall improvement in performance and no advantage of either approach over another was evident.

2.4.3.7 Risk of Bias Across Studies

The level of evidence is generally considered to be low except for one study (Mishra et al., 2015), which reached moderate level of evidence. A low level of evidence is claimed to be indicative of unrepeatable results, and lower confidence in the research. Such an issue could increase bias when interpreting the evidence in favour of AT. For some of the articles the reported research outlined the proof of concept in a pilot study and stated that larger scale studies were intended (Kronenberger et al., 2011; Welch et al., 2015; Yucel et al., 2009). For many of the studies one of the main issues related to the small sample (average of 10.33 subjects for studies that used repeated measure design and 22.83 subjects for studies that included controls) size which potentially resulted in an underpowered study.

Table 2-1: Summary of the Extracted Data from the Nine Included Studies

Study		**Participants**		**Training**					**Findings**		
Authors	**Design**	**N**	**Age**	**Stimuli**	**Skills Trained**	**Frequency and Duration**	**Place of Training**	**Outcome Measures**	**Improved?**	**Retention**	**Generalization**
Good et al. (2017)	Non-RCT	9CIEG/ 9CI CG	6-15y	Piano training (musical theory and technical exercises; and learning a song)'	music theory & technical exercises scales (bilateral finger control, and hand positions; learning a song)	24 session; private half an hour lesson per week for 24 weeks for 6 months.	Lab	-Montreal Battery for Evaluation of Musical Abilities (MBEMA) (Peretz et al. 2013) -Perceived Emotional Prosody based on Diagnostic Analysis of Nonverbal Accuracy Scale (Nowicki and Duke 1994)	Not assessed (Purpose was to investigate generalization not trained task)	Not assessed	Yes
Hagr et al. (2016)	Non-RCT	13CI EG / 13 CI CG	3-7y	Detection of Ling sounds, environmental sounds, and phrases; discrimination between intensity, duration, pitch, or intonation stress, rhythm and rate; discrimination of vowels, consonants, and number of syllables in words	Sound detection and discrimination using Rannan software	1 hour of weakly speech therapy + extra 1 hour of AT using Rannan weekly (in a different day) for 12 months.	PC based in clinic	- Listening Progress Profile (Lip) (Nikolooulos et al ,2000); -The Infant-Toddler Meaningful Auditory Integration Scale. (IT MAIS) (Zimmerman-phillips et al, 2001)	Yes	Not assessed	Not assessed

Ingvalson, Young, and Wong (2014)	RCT	10 CI EG/ 9 CI CG	4-7y	Recalling and sequencing environmental and speech sounds in quiet and noise; matching phonemes to graphemes; identifying and discriminating between phonemes; recalling sequence of drumbeats, speech sounds, syllables, and phonemes; blending words, syllable	Phonological awareness skills and auditory working memory.	Interactive exercises, 75 min of training per week for four weeks.	PC based in school	- Expressive One Word Picture Vocabulary Test (EOWPVT) (Martin, Brownell, 2011). - Receptive One Word Picture Vocabulary Test, (ROWPVT), (Martin, Brownell, 2011). - Oral Written Language Scales (OWLS) (Carrow-Woolfolk, 2008)	Yes	Not assessed	Not Assessed
Kronenberger, Pisoni, Henning, Colson, and Hazzard (2011)	Repeated measure	9 CI	7-15y	Cogmed Working Memory involving auditory, visuospatial, or combined short-term and working memory skills.	Working memory	30-40 min per day for 5 days a week for 5 weeks	PC based in Home	- Digits forward and backward - Spatial span forward and backward; BRIEF: - Sentence repetition	Yes	yes (all working memory and Language for 1 month and language only up to 6 months)	Yes (working memory to language processing)
Mishra, Boddupally, and Rayapati (2015)	Non-RCT	13 CI EG/ CI 14 CG	5-12y	Adaptive speech (numbers) in noise recognition in a white/speech	Speech in noise	2 sessions 40 minutes per day for 6 days a week for 5 weeks	PC based in Home	- Numbers in white noise, - Number in speech-shaped noise (trained);	Yes	Yes for up to 3 weeks	near transfer but not far transfer

				noise. (Angel Sound)				- Digit triplets in speech shaped noise (untrained)			
Roman, Rochette, Triglia, Schön, and Bigand (2016)	RCT	10 CI EG / 9 CI CG	4-10y	Environmental sound, music, voices, and abstract	Auditory cognitive processing (identification, discrimination, Auditory scene Analysis (ASA) and auditory memory)	30 minutes per 1 session per week for 20 weeks,	Sound in Hand instrument; in clinic/ Lab	- Same as training stimuli but different sets used only as outcome measures	Yes in all except ASA	Yes	Yes (Phoneme Discrimination)
Welch et al. (2015)	Non-RCT	12 9CI/3HA)/17NH	5-7y	Singing exercises vocal explorations; tongue twisters; explorations in visual imagery for sound, sound imagery and metaphor	Singing and vocal exploration	once a week for 20 weeks	School	- Singing competency profile *Sing Up* (Welch et al, 2014); - Chord pitch discrimination test; - Speech perception in noise.	Yes but not speech in noise	Not assessed	Not assessed
Wu, Yang, Lin, and Fu (2007)	Repeated measure	7CI/3HA	5-11 yrs	Discrimination task, trained to identify final vowels. Discriminating between phonemes. For vowels, acoustic speech features included (F 1 and F 2) and duration;	Identification and discrimination of speech sound	30 min per 1 session 5 days a week for 10 weeks	PC based in home	- Vowel and consonants discrimination - Chinese tone recognition	Yes	yes for 2 months	Not assessed

Yucel, Sennaroglu, and Belgin (2009)	Non-RCT	9 CI EG /9 CI CG	6-96 m	Pitch discrimination task; rhythm discrimination, and sequence repetition	Child listening to different pairs of notes using electronic keyboard,	10 minutes daily for 2 years post CI activation	Key-board in home	- Music: developed questionnaire. - MAIS and MUSS - Phonetic discrimination, - Word identification, - Comprehension of simple auditory instructions, and - Sentence repetition	Yes	yes	No (No transfer to speech)

Table 2-2: Summary of the Main Findings

Study	Findings				
Authors	**Outcome measures**	**Improved trained skills**	**Retention**	**Generalization**	**Reporting Compliance**
Good et al. (2017)	- Montreal Battery for Evaluation of Musical Abilities (MBEMA) (Peretz et al. 2013) - Perceived Emotional Prosody based on Diagnostic Analysis of Nonverbal Accuracy Scale (Nowicki and Duke 1994)	The purpose of study was to investigate generalization not trained task	Not assessed	Yes	Not explicitly reported but can be deduced 1
Hagr et al. (2016)	- Listening Progress Profile (Lip) (Nikolooulos et al ,2000); - The Infant-Toddler Meaningful Auditory Integration Scale. (IT MAIS) (Zimmerman-phillips et al, 2001)	Yes	Not assessed	Not assessed	Not reported
Ingvalson et al. (2014)	- Expressive One Word Picture Vocabulary Test (EOWPVT) (Martin, Brownell, 2011) - Receptive One Word Picture Vocabulary Test, (ROWPVT), (Martin, Brownell, 2011) - Oral Written Language Scales (OWLS) (Carrow-Woolfolk, 2008)	Yes	Not assessed	Not Assessed	Not explicitly reported but can be deduced 1
Kronenberger et al. (2011)	- Digits forward and backward - Spatial span forward and backward - Working memory and sentences repetition - Sentence repetition	Yes	yes (all working memory and Language for 1 month and language only up to 6 months)	Yes (working memory to language processing)	Not explicitly reported but can be deduced 1

Mishra et al. (2015)	- Numbers in white noise, - Number in speech-shaped noise (trained); - Digit triplets in speech shaped noise (untrained)	Yes	Yes for up to 3 weeks	Near transfer but not far transfer	Explicitly reported
Roman et al. (2016)	- Same as training stimuli but different sets used only as outcome measures	Yes, in all except ASA	Not assessed	Yes (Phoneme Discrimination)	Not explicitly reported but can be deduced 1
Welch et al. (2015)	- Singing competency profile Sing Up (Welch et al, 2014); - Chord pitch discrimination; Speech perception in noise.	Yes, but not speech in noise	Not assessed	No transfer to speech	Not explicitly reported but can be deduced 1
Wu et al. (2007)	- Vowel and consonants discrimination - Chinese tone recognition	Yes	yes for 2 months	Not assessed	Not reported
Yucel et al. (2009)	- Music: developed questionnaire. - MAIS and MUSS - Phonetic discrimination, - Word identification, - Comprehension of simple auditory instructions, and - Sentence repetition	Yes (only trained)	Not assessed	No transfer to speech	Not reported

Table 2-3: Level of Evidence and Quality of Studies

Article	**Scientific study Validity criteria**					**Training-specific Study Validity Criteria**					**Study Quality Score**	**Leve of Evidence**
	Randomization	Control Group	Power Calculation	Blinding	Outcome measure reporting	Outcome Measure Selection	Training Feedback	Ecological Validity	Reporting of Compliance	Follow Up		
Good et al. (2017)	1	2	0	0	2	1	0	1	1	0	8	Low
Hagr et al. (2016)	0	1	0	1	2	1	2	1	0	0	8	Low
Ingvalson et al. (2014)	1	1	0	0	2	2	2	1	1	0	10	Low
Kronenberger et al. (2011)	0	0	0	0	2	2	0	2	1	2	9	Low
Mishra et al. (2015)	1	1	0	1	2	1	2	2	2	1	13	Moderate
Roman et al. (2016)	1	1	0	0	2	1	0	1	1	0	7	Low
Welch et al. (2015)	0	1	0	0	2	2	1	1	1	0	8	Low
Wu et al. (2007)	0	0	1	0	2	1	2	2	0	2	10	Low
Yucel et al. (2009)	0	1	0	0	2	2	0	2	0	0	7	Low

2.5 DISCUSSION

2.5.1 Summary and Recommendations

This systematic review assessed the literature on the benefits of AT with paediatric CI users. For two of the studies, the study group contained children with other hearing devices as well, however the focus was on CIs. Trained tasks included working memory phonological awareness, speech perception, music perception, singing, pitch and rhythm discrimination, and environmental sound identification. Benefits of AT were illustrated through improvement on trained tasks in all nine studies regardless of the duration or type of training. In addition, four out of six studies, which assessed generalization of training, demonstrated a transfer of improvement to other learning domains, such as working memory training that led to improved language processing skills along with improved working memory skills (Kronenberger et al. (2011), and music training that lead to improved emotional-speech-prosody perception (Good et al. 2017). Although these results are encouraging for clinicians when considering whether to incorporate AT in the rehabilitation pathway of paediatric CI users, clinicians have to bear in mind that the evidence supporting such claims are not solid. In fact, a recent meta-analysis (Melby-Lervag, Redick, & Hulme, 2016) demonstrated that working memory training does not improve other skills that are not working memory specific, including speech perception. However, there is no evidence either that such findings apply to CI users since the number of working memory training studies with CI is extremely limited.

The findings also suggest that the type of AT should be determined based on individual needs, since both analytic and synthetic approaches led to improvements with no definite benefits of one approach over another. Further work is required to understand if there are specific reasons to use different techniques or whether any AT approach will suffice.

Interestingly, it was observed that almost all studies that used synthetic training, independently or along with analytical exercises, assessed the benefits of generalization of learning to untrained tasks or other auditory perceptual domains. Namely, Good et al., Kronenberger et al., Mishra et al., and Roman reported benefits in untrained tasks (Good et al., 2017; Kronenberger et al., 2011; Mishra et al., 2015; Roman et al., 2016) whereas studies by Hagr et al. (2016) and Wu et al. (2007) used only analytic tasks and did not assess the benefit of generalization to untrained tasks, perhaps because training stimuli were targeting basic discernible skills that were not expected to influence untrained skills. Although there was no clear evidence for benefits of using one training approach over another, a trend emerged to suggest that adding synthetic training tasks to analytic training might be optimal because it combines higher language and/or cognitive processing with the more basic perceptual discrimination abilities. This trend supports the recommendation by Amitay et al., (2006) who also suggested combining the two approaches to achieve maximum benefit.

An essential measure when assessing the benefits of AT is retention of benefit and is measured in follow-up assessments after AT is completed. Such factors can influence the clinicians' decisions when offering AT in clinical settings; if the retention is low, the motivation for utilizing AT will be low, and vice versa. Hence, retention of improvement was assessed in this review. Surprisingly, only three studies (Kronenberger et al., 2011; Mishra et al., 2015; Wu et al., 2007) investigated retention post AT and revealed that improvements were sustained for a period ranging from two weeks and up to two months post AT intervention. Such great variation in retention periods could also be reflective of subjects' compliance to training programmes, yet another essential measure for the effectiveness of AT. Unfortunately, only one study (Mishra et al., 2015) assessed compliance to AT programmes, which illustrated its importance as a sign of children's and their families' interest in AT, and ultimately as an indicator of the intervention's success. Therefore, we recommend investigating these two AT specific measures to demonstrate the effectiveness of AT in future studies.

Another factor that was not investigated in the studies is quality of life. Quality of life is an essential outcome, which may also influence clinicians' and service providers' decisions to offer AT in their practice. The only study to include self or parent report questionnaires as an outcome measure was Yucel et al. (2009). Such tools are valuable when assessing the outcome of AT as it directly determines the attitude of the end-users to the intervention and highlights if they observed changes in speech perception and production, and how the training affects everyday life.

None of the studies that were included investigated the effects of specific methods that may maximize participants engagement in AT interventions. It would be beneficial to explore such methods that may encourage participants to be fully engaged in AT intervention, such as familiarisation, support groups, family involvement, or feedback and reinforcement. For example, incorporating feedback into AT interventions can maximize its benefits (Tye-Murray, 2019). Feedback, whether orthographic or auditory, was shown to be successful at embedding training effects in adult participants to improve the accuracy of identification and discrimination of stimuli (Burk & Humes, 2008). Another factor that has been shown to successfully improve outcomes of training in children is that of active parental engagement (Roberts & Kaiser, 2017).

The categorisation of the articles indicated that quality of the studies was low to moderate. This is in line with Henshaw and Ferguson (2013) who assessed the AT literature for adults with hearing loss and found that the level of evidence was very low to moderate. In other medical fields such as plastic surgery, there is an agreement that the grading system should not dismiss lower quality evidence when deciding on recommendations if the results are consistent (Burns, Rohrich, & Chung, 2011), a pattern that was observed here. When looking at the specific studies in this review, factors contributing to a lower overall quality scores are mainly lack of randomization, lack of a power calculation, and lack of blinding, which can all be practically difficult to achieve in studies dealing with populations such as children with CI because of the size of the population and constraints due to delivery approaches for the intervention, such as within a school, which can make randomisation very difficult. Future AT research with this population should attempt to overcome some of these limitations by using greater control in the participant recruitment and intervention delivery. The population size available now is far larger than previously had been the case for some of the earlier studies and there are many outcome measures that have published reliability values to be able to conduct power calculations, so some issues can readily be overcome. Future studies should be careful to report participant compliance, and appropriate outcome measures selected to reflect direct, generalised and real-life listening situations. For assessing generalisation, the use of outcome measures should be both specific and general and include periods without intervention to assess retention. Even though meta-analysis of the benefits of AT is not feasible due to the diversity of the outcome measures used across studies, generalization and retention of benefits can be the focus of future studies as primary AT outcomes regardless to the measures used in the studies, and eventually be investigated in a meta-analysis.

2.5.2 Limitations of this Review

There were three main limitations in this review. Firstly, CI and hearing aid users were followed in two of the studies (Welch et al., 2015; Wu et al., 2007), which could be considered as inconsistency in the targeted population in the analysis because the intention was to only explore studies using children with implants. Due to the small number of studies available investigating outcomes purely with children with CIs, it was decided to include them. Furthermore, as more present-day CI users have greater degrees of residual hearing the distinction between these two populations becomes less clear. The second limitation occurred because it was not possible to conduct a meta-analysis because of a lack of commonality amongst outcome measures. Finally, this analysis did not consider the impact of duration and frequency of the intervention on the outcome of AT, which could have a large impact on outcomes; this aspect is not clearly reported in the literature.

2.6 CONCLUSION

The literature on the benefits of AT in paediatric CI recipients was systematically reviewed. Benefits of AT were demonstrated through the improvement of all trained tasks in the studies analysed, regardless of the duration or type of training. Transfer of improvement to untrained tasks was measured in number of the studies (6 out of 9). Retention of benefits after a period without training, following the intervention, was evident in the cases where it was assessed (3 out of 9) but time periods for evaluation varied. None of the studies assessed changes in quality of life despite its value when assessing the effectiveness of interventions. In agreement with previous reviews, a higher quality of evidence for examining outcomes of AT in paediatric CI recipients is still required. The lack of higher quality studies should not be associated with the effectiveness of AT intervention. It is important not to draw the conclusion that the current level of evidence infers lack of benefit especially because the studies reviewed consistently reported benefit.

To ensure that future AT studies achieve a higher level of evidence when graded and to minimize the potential bias, general measures such as randomization, power calculation, blinding and control groups should be used. Other outcome measures such as quality of life, retention of benefit and compliance to AT programme should also be incorporated and be considered as key indicators to the success of any AT programme.

CHAPTER 3: THE DEVELOPMENT OF A PAEDIATRIC PHONEME DISCRIMINATION TEST FOR ARABIC PHONEMIC CONTRASTS

3.1 OVERVIEW

This chapter presents a new Arabic monosyllabic closed-set consonant-discrimination test for the use with Arabic-speaking children over the ages of five years. The main motivation for this research was the lack of materials that can be used with this population to assess their consonant perception skills and monitor changes over time or after an intervention. This chapter describes the steps for producing a translated version of the CAPT in Modern Standard Arabic for developing and evaluating the materials based on knowledge of the vocabulary and contrastive words. The chapter will also describe piloting the initial version of the materials to evaluate the familiarity of the target population to the words, develop final lists, and assess test–retest reliability.

3.2 INTRODUCTION

3.2.1 Background

The global prevalence of hearing impairment ≥35 dB HL among children between the age of 5–14 years was estimated to be 1.2%. Saudi Arabia has a higher prevalence of hearing loss in paediatric population, with an estimation of 13% for both conductive and sensorineural hearing impairments (Al-Shaikh et al., 2002). A large-scale epidemiological study was carried out between 1997 and 2000 and reported that the percentage of Saudi Arabian children with confirmed diagnosis of sensorineural hearing loss (SNHL) was 1.5% and children with confirmed diagnosis of severe to profound hearing loss was 0.7% (Al-Shaikh & Zakzouk, 2003; Al-Shaikh et al., 2002). Such a high prevalence has been associated with congenital conditions (Al Salloum, El Mouzan, Al Herbish, Al Omer, & Qurashi, 2015; Habib & Abdelgaffar, 2005), childhood onset loss (Al-Rowaily, AlFayez, AlJomiey, AlBadr, & Abolfotouh, 2012), and attributed to the common practice of consanguineous marriage, which concentrates genes known to cause hearing impairment (Al Salloum et al., 2015; Zakzouk, 2002).

Inevitably, the incidence rates of hearing loss are increasing over time (A Al-Abduljawad & Zakzouk, 2003) and the number of paediatric candidates for hearing devices is growing. Such a high incidence rate may have led Saudi Arabia to establish the largest centre for CIs in the Middle East and in to an extensive use of hearing devices (Hazaimeh, 2013). For appropriate monitoring of paediatric outcomes, well-validated measures of hearing ability are required. Currently, there is an extremely limited selection of tests that can be used to reliably assess speech perception in Saudi-Arabic speaking children. A common practice in Saudi Arabia is to use Arabic translations of English tests or Arabic tests that have been developed in other Arabian countries to assess speech perception in Saudi Arabian children. Clearly, tests translated directly from English to Arabic without proper validation with Saudi Arabic speaking children are unlikely to be balanced and likely to have poor reliability. Similarly, tests that were developed in other Arabic countries often include unfamiliar words to Saudi Arabian children (Alsari, 2015). In the current practice, the reliability of speech tests used with Saudi Arabian children is seldom known.

Speech perception tests are the primary outcome measures used to assess speech development for children with CIs (Schaefer et al., 2017) and are essential tools for the assessment and management of hearing loss. There is value in developing and validating reliable speech perception tests that can assess and monitor auditory perception since good auditory discrimination skills are essential for the development of speech and language in children (Kuhl et al., 2008; Sharma, Dorman, & Kral, 2005).

3.2.2 Selecting Appropriate Speech Materials in Arabic

Approximately 319 million people in the world speaks Arabic, inclusive to all dialects and forms, making it one of the five most spoken languages worldwide (Doochin, 2019; McCarthy, 2018). Native-Arabic speakers are spread all over the globe as Arab are amongst the fastest growing diaspora population in the world; they make up 4% of Berlin’s population in Germany, 4% of Belgium’s population, 2.5% of France’s population, nearly 1.5% of the United Kingdom’s population, and 1.1% of the United States’ population (Arab Institute Foundation, 2018; Doochin, 2019; McCarthy, 2018; Winter, 2019).

Similar to Modern Greek, Swiss German, and Haitian Creole, Arabic is a diglossic language where the same speaker uses different forms of the language in different settings (Ferguson, 1959). There is a coexistence of two varieties of the language throughout the community, one of which is the literary or formal dialect while the other is a colloquial dialect that is spoken on everyday basis. Colloquial dialects are the true mother-tongue of native speakers of Arabic; they vary slightly within regions and between different parts of the country, as well as between different Arabic-speaking countries where dialects may even be incomprehensible (Broselow, 2003). Nevertheless, the Modern Standard Arabic is the common form of Arabic that can be understood across this diverse Arabic-speaking population.

The diglossia and range of dialects add complexity when developing speech assessment materials. Ideally an Arabic language speech test should be appropriate for all Arabic speaking countries and appropriate for use in other countries with large populations of Arabic speaking individuals (e.g. Belgium, Germany, France, UK, US) migrated from different countries. This diversity and complexity should be considered when working with the Arabic language and could potentially be the reason why few validated Arabic speech assessment measures exist. Fortunately, a common practice across all Arabic-speaking countries is to use the Modern Standard Arabic in formal settings such as education and media. This across-language shared approach enables us to use Modern Standard Arabic when developing Arabic speech materials that are relevant to many countries. One example of the use of this approach is the work by Ashoor & Prochazka (1985) who developed the Saudi Speech Recognition Threshold (SRT) test using Modern Standard Arabic. Modern Standard Arabic is unfamiliar to some pre-school children because this form of language is not used in everyday conversations. However, it is formally studied in schools, used in the media, in children’s programmes and cartoons. Therefore, it is appropriate to use Modern Standard Arabic with children aged five and older, because they attend schools and have had years of exposure to media and children’s television.

3.2.3 Previous Arabic speech tests that can be used with Saudi children

There are a limited number of tests that can assess speech perception in Arabic-speaking children from Saudi Arabia. A test that was developed recently was the Arabic version of the Lexical Neighbourhood Test (LNT) (Kirk, Pisoni, & Osberger, 1995) for children using CIs in Saudi Arabia (Alsari, 2015). Alsari (2015) developed this open-set test to assesses speech recognition skills in children with hearing impairment. The test was based on two main principles; firstly, to ensure that the words of the LNT are familiar to young children with their limited vocabularies and secondly to construct the LNT based on the standards of the Neighbourhood Activation Model (NAM) (Luce & Pisoni, 1998). NAM recommends that words are organized into “similarity neighbourhoods” based on their frequency of occurrence and also the organization of the word in the mental lexicon which is based on “lexical density” i.e., acoustic–phonetic similarity of words within the lexical neighbourhood. The Arabic Lexical Neighbourhood Test (ALNT) (Alsari, 2015) consists of two lists (easy and hard) with 50 words each. The ALNT (Alsari, 2015) was developed in colloquial Najdi Arabic dialect and was validated with normal-hearing children. The test was shown to be reliable even when administrated repeatedly over time.

Cochlear Ltd in collaboration with the Centre of Competence HörTech published the Arabic Matrix Sentence test (Kollmeier, 2014). This test utilizes an adaptive staircase approach to determine the speech reception threshold (SRT) in noise and in quiet. All sentences have the same syntactic structure (e.g. verb, name, number, noun and adjective) presented in modern standard Arabic. Test lists are created by selecting random sentences from an inventory (matrix) of 50 words, i.e. ten words per category. Despite the random selection of items for use in the sentences, every sentence is syntactically correct. The materials used require a reasonably mature language development and the ability to read, therefore it has been recommended for use for adults and children over the age of 12 years, ruling it out as an assessment for younger children in primary schools.

Other Arabic language paediatric speech tests that are routinely used to assess speech perception in Saudi Arabian children include the SRT test in Saudi (Ashoor &

Prochazka, 1985) and Egyptian (Soliman & Fathallah, 1984) dialect, and an Arabic word intelligibility (recognition) by picture identification in Egyptian dialect (Soliman, Abd El-Hady, Saad, & Kolkaila, 1987). These test materials are usually delivered to the individual via live-voice since recorded materials are either not accessible or mainly available in Egyptian but not Saudi dialect.

Currently, there are only two published speech perception tests that were validated on Saudi Arabic speaking children (Alsari, 2015; Ashoor & Prochazka, 1985). Although the SRT test (Ashoor & Prochazka, 1985) was validated on Saudi children, its recordings are not digital or readily accessible. A-LNT (Alsari, 2015), on the other hand, was validated with Saudi speaking children with normal hearing and also with a group with CIs; its recordings are digitised and available. Nevertheless, neither of these tools provide frequency-specific information about the audibility and discrimination of speech cues from the pattern of phoneme confusions with known reliability, such a valuable feature that can be used in assessing and monitoring auditory perception in children. In addition, one measure (Arabic LNT) recorded in one dialect is not sufficient to assess speech perception outcomes in all Saudi Arabian children especially since dialect can significantly influence speech recognition performance.

3.2.4 Chear Auditory Perception Test

There are many published speech perception tests in English for children that assess speech recognition, auditory discrimination, and or monitor progress of speech and language skills. This Arabic auditory perception test was developed based on the English CAPT (Vickers et al., 2018), which was reported to provide valuable frequency-specific information about the audibility and discrimination of speech cues from the pattern of phoneme confusions produced after a child completed the test. The CAPT was shown to be sensitive to hearing aid gain settings (Marriage & Moore, 2003; Marriage et al., 2018) and was used along with the McCormick Toy test (McCormick, 1977) to derive UK CI candidacy criteria (Lovett, Vickers, & Summerfield, 2015). It is a phoneme discrimination test, which consists of monosyllabic words in form of consonant-vowel-consonant (CVC) or consonant-vowel-consonant-consonant (CVCC) that differ in either first or final phoneme. It is a closed-set test that uses four response alternatives on each trial. All the items in the test are real words that are familiar to children in the targeted age range.

The CAPT was validated and the critical difference for the measure was calculated. The critical difference is a measure that takes into account the reliability of the materials. It is a measure that can be used on an individual basis to compare performance in two listening conditions (e.g. with and without hearing devices) or different hearing devices fittings. The calculation of the critical difference provides a range of values for each individual's scores that indicates a "true" difference, only if an individual's score in the second condition falls outside the provided range would this be seen as a genuine change in scores. The critical differences for the original CAPT were calculated and were consistent with other speech tests, where the theoretical critical differences were somewhat smaller than the obtained critical differences for children, indicating that the children are less consistent across the test and retest sessions than predicted.

3.2.5 Rationale and Aim of this Research

There are a limited number of published tests that were validated with Arabic speaking children in Saudi Arabia (Khoja & Sheeshah, 2018; Khoja, 2019); a summary of these tests is listed in Table 3-1. The scarcity of tools to help assess consonant perception in Saudi-Arabic speaking children was the main motivation of this study but with the intention of providing a measure that is also relevant and usable for other Arabic-speaking children. With tools for the assessment of the discrimination of speech cues to provide frequency-specific information with known reliability, it would help clinicians to verify the benefits of hearing devices or assess effectiveness of habilitation interventions. Therefore, the primary aim of this study was to develop the Arabic CAPT (A-CAPT), a closed-set phoneme discrimination test in Modern Standard Arabic that was developed based on the British English CAPT (Vickers et al., 2018). A secondary aim of this research was to investigate whether a discrimination test in Modern Standard form of a language, in this case Arabic, can assess consonant perception in school-aged children.

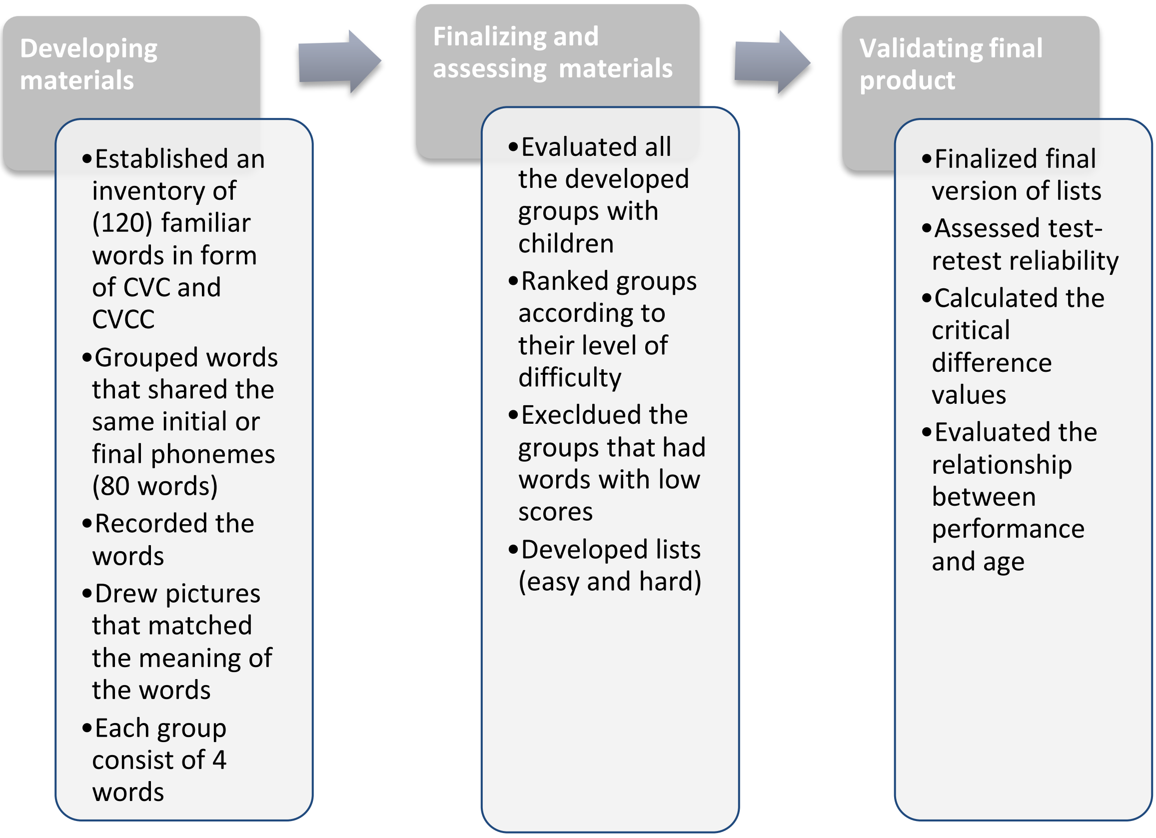
This work outlines a procedure for producing a carefully translated version of a speech test in another language. The stages were to: 1) develop the materials based on knowledge of the vocabulary and contrastive words, 2) evaluate the stimuli and the response pictures with an expert panel, 3) pilot the initial version of the materials in a group setting, using electronic response voting, to understand whether all words are understood by the target population and derive final lists, 4) run test retest reliability with target population using individual testing approach.

Table 3-1: Published speech tests that were validated with Saudi-Arabic children

Name	Authors	Type	Target age	Dialect
Arabic Lexical Neighborhood Test (LNT)	(Alsari, 2015)	Open-set word recognition test	5-13 year old children with CI	Colloquial Najdi Arabic
Arabic Matrix Sentence test	(Kollmeier, 2014)	Closed-set sentence recognition	12 year old children and above	Modern Standard Arabic
SRT test in Saudi	(Ashoor & Prochazka, 1985)	Open-set	5 year old and above	Modern Standard Arabic

3.3 MATERIALS AND METHODS

The process of the development and validation of this test was conducted in three main steps (Figure 3-1). The first step was the development of the materials; followed by the validation of the intelligibility of the selected words within materials, and finally the validation of the developed lists.



Developing materials

- Established an inventory of (120) familiar words in form of CVC and CVCC
- Grouped words that shared the same initial or final phonemes (80 words)
- Recorded the words
- Drew pictures that matched the meaning of the words
- Each group consist of 4 words

Finalizing and assessing materials

- Evaluated all the developed groups with children
- Ranked groups according to their level of difficulty
- Execldued the groups that had words with low scores
- Developed lists (easy and hard)

Validating final product

- Finalized final version of lists
- Assessed test-retest reliability
- Calculated the critical difference values
- Evaluated the relationship between performance and age

Figure 3-1: Summary of the process of development and validation of the test

3.3.1 MATERIALS

To develop the A-CAPT, we established an inventory of 120 monosyllabic/monophthong words; there are six monophthongs in Modern Standard Arabic: three long vowels: /a:/, /i:/ and /u:/ and three short vowels: /a/, /i/ and /u/. Selected words were found in Arabic children’s story books and commonly used in everyday life, and thus it was presumed to be familiar to Arabic-speaking children aged five years and older. Unfortunately, graded children’s book series similar to the graded reading series found in English were not available in Modern Standard Arabic, and thus we opted to use popular Arabic children’s ungraded story books.

We arranged the monophthongs in groups of four Arabic meaningful monosyllabic CVC or CVCC words that differ only in one phoneme. Words were grouped based on their consonant environments, where each selected word was in a group with three other confusable words. These confusion groups (CGs) containing the four similar words differed only in the first or final phoneme. The test therefore used a closed-set four-alternative-forced-choice test paradigm. Since not all the words in the inventory could fit into groups of four minimally contrastive words with similar phonemes, only 77 words were used and we produced 20 CGs consisting of 80 words (three words (/batˤ/, /tˤi:n/,/bar/), were presented in two CGs). Each CG contained four similar words that differed in either the initial (e.g. /jad/, /sad/, /yad/, /xad/) or final phoneme (e.g. /xatˤ/, /xas/, /xal/, /xad/). The groups were divided equally into two subgroups (ten CGs i.e. 40 words each), one of which assessed the first phoneme and the other assessed the third or final phoneme. The subgroups were also divided equally into two subgroups for vowel length (five CGs). In total, there were two groups of words, first phoneme contrast and final phoneme contrast. Both groups contained forty words divided into twenty long vowel words and twenty short vowel words.

Familiarity and intelligibility of words within CGs was assessed by a simple binary forced response survey that was used with the three native Arabic audiologists who volunteered to assess the appropriateness of the materials for the target group of children. In a group setting, the clinicians listened to each stimulus and matched it to the corresponding picture then decided whether it was appropriate or inappropriate. The group agreed that the word /ɣaːb/, which refers to a verb form of the word absent, was rather abstract and the illustration may cause confusion to the children. Accordingly, the CG that contained this word was marked for elimination in the final version of the word lists. To keep an even number of CGs for the purpose of creating equal word lists, we opted to eliminate the CG that was determined to be least familiar to children (/kaf/, /raf/, /daf/, /saf/) and this was used for a practice run. Otherwise, the expert panel agreed that the words selected were appropriate and matched the illustrations.

An attempt to create CGs in which three words were grouped based on their vowel environments, long vowels /a:/, /i:/, and /o:/ or short vowels /a/, /i/ , and /o/. However, most of the words in the inventory could not fit into groups of three minimally contrastive words. Furthermore, whenever a CG was formed, the meaning or familiarity of the words within was questionable. For example, the CG that included the words /tˤa:r/, /tˤi:r/ and /tˤo:r/, which are translated to the words flew, fly and name of a mountain, respectively. The word /tˤo:r/ is not a familiar and it is not expected to be recognised by children. Hence, it was decided to make the A-CAPT a consonant discrimination test.

3.3.2 Recording Methods and Handling Speech Materials

3.3.2.1 Recording Words

Three native Arabic-speaking adults volunteered to record the words in Modern Standard Arabic, two females and one male (age range 35-46 years old); each word was recorded twice. The female speakers were both originally from the central region of Saudi Arabia (Al-Qassim), the first speaker was a post-graduate student while the other speaker was an elementary-school teacher. The male speaker was from the western region of Saudi Arabia (Makkah) and was a university lecturer. The speakers were seated a meter away from the microphone. The stimuli were recorded in an Anechoic Chamber (AC) at University College London with a Bruel & Kjaer 2231 Sound Level Meter fitted with a type 4190 condenser microphone. The signal was digitised with a Focusrite 2i2 USB sound card at a sample rate of 44100 Hz. Six continuous wav files were recorded using ProRec 2.4 (Huckvale, 2018) and the recording software was developed at UCL. Automatic separation before and after utterances and labelling for each word was achieved through ProRec after filtering the .wav files with a high pass filter to reduce gross fluctuations (using a MatLab script). Finally, the RMS values for all words was equated.

Three Arabic native speakers critically listened to the recorded words and gave their feedback on pronunciation, clarity of the recordings, accuracy of utterances in the Modern Standard Arabic and their preferred speaker out of the three. Using google forms, the evaluators individually listened and rated each word as clear or not clear. One of the evaluators who completed the forms was a post-graduate student in linguistics and her Ph.D. project involved investigation of dialects in Saudi Arabia, the other two evaluators were highly educated clinical audiologists in Saudi Arabia. The three evaluators voted for the same female speaker, and thus her voice was selected for the A-CAPT.

3.3.2.2 **Test Materials and Illustrations**

The pictures were all drawn by a 14-year-old child to ensure that they were relevant for younger children. Although there is no evidence that children’s drawings are necessarily more relevant to other children than professional illustrators, we chose to do this because in the original implementation of the CAPT some of the figures had to be altered to make them more meaningful for children. For example, the word peg was originally a picture of a peg to be used on a washing line but had to be replaced with a clothes peg for hanging up coats. The pictures were then made into jpegs and a caption of the word written in Arabic was added to each figure (See Figure 3-2). After recording the words and matching them to corresponding pictures, the familiarity and appropriateness of the words and their corresponding pictures were assessed by three volunteers, Arabic speaking audiologists, who listened and evaluated all the words and their corresponding pictures.

Figure 3-2: Example of a CG (/duːr/, /nuːr/, /ħuːr/, and /suːr/) and their

3.3.3 Methods

3.3.4 Phase I: Assessment of Speech Materials and Development of Word Lists

In this phase, we evaluated whether or not the selected words in Standard Arabic language were appropriate and intelligible for the targeted age group of children.

3.3.4.1 Participants

Adverts were sent to the King Fahad Academy (KFA) in London, United Kingdom (UK) to recruit subjects for this experiment. The KFA is an independent school that follows the UK national curriculum and is funded by the Saudi Arabian Embassy in the UK. Twenty-six children aged between six and eleven years (mean age = 8.94 years) were randomly selected from families that responded to our adverts. All children were screened at 20 dBHL using pure-tone audiometry at frequencies 0.5, 1.0, 2.0 and 4.0 kHz. Transient evoked otoacoustic emission (TEOAE) was also performed on each child. All children passed the hearing screening and have no known learning disabilities

3.3.4.2 **Validation of Test Material.**

Test materials, 80 monosyllabic words, were delivered via a computer using Prezi presentation slides on a screen through an EB-X62 EPSON projector. The words were presented at a soft presentation level calibrated to be 50 dBA at the centre of where the children were sitting for the testing to avoid ceiling effects. Words were delivered via a wall speaker Model NV-WA40W-SP to a group of normal hearing children. The order of words was the same in test and retest sessions. Tests were conducted in a classroom where the average background noise level of the room ranged 40-45 dB SPL; the noise level was measured three times within each session, before conducting the test, during the test and at the end of the test session. The classroom windows were shut to minimize external background noise. The classroom was allocated for non-English speaking children who receive extra language sessions and was located in the administration area which was generally the quietest in the school. No incidents of sudden background noise were observed during testing. The dimensions of the classroom were: 800 cm long, 704 cm wide, and 250 cm high. The children sat on the carpet in the middle of the room with the first row two meters from the loudspeaker. There were five rows of children in total and the calibration was conducted at the midpoint of these rows.

The children were instructed to select one out of four pictures that visually and orthographically represented the presented word. Children had a practice run that consisted of 4 words to familiarize them with the process. Following Vickers et al., (2013) speech test procedure, each child was assigned a hand-held infrared transmitter to record her or his response to each trial. Turning point software and a USB receiver were used to capture the children’s responses. A rule from Vickers et al. (2013) was also followed, this was to exclude participants if they missed four or more items to rule out technical issues such as malfunction of transmitters.

3.3.5 **Phase II: Validation of the Developed Lists**

In this phase, data were collected to validate the developed lists from phase I, of which there were four. Data were analysed for individual lists and also by combining the two easier lists and also combining the two harder lists producing a total of two lists, an easy list and a hard list. Each list can be used repeatedly by merely changing the order of the words within the list because all words are represented on each run (See Table 3-3).

3.3.5.1 Participants

Children were recruited via advert at King Abdul-Aziz University in Jeddah, Saudi Arabia. Sixteen children aged between five and eleven years (mean age = 8.33 years) participated in the validation of this phase, nine males and seven females. All children were screened at 20 dBHL using pure-tone audiometry at frequencies 0.5, 1.0, 2.0 and 4.0 kHz. All children passed hearing screening and none were reported to have learning disabilities.

3.3.5.2 Technical Delivery

Experiments were conducted in a quiet room in King Abdul-Aziz University or at the participant’s home. The noise floor was measured using a sound level meter to be equal or less than 40 dBA. The test materials were delivered via a computer running a MatLab script to present stimuli, show response options and record responses. The participants were instructed to select one out of four pictures that visually and orthographically represent the presented word. Each participant was tested individually; the child listened to the stimuli over Sennheiser HD 650 headphones and selected the corresponding picture out of four choices shown on the computer's screen that visually and orthographically represented the presented word. The test was presented at four different levels 40, 50, 60, and 70 dB SPL or until the child reached the maximum score, to ensure that a range of performance was covered. Each word in the lists was tested and the choice of the four pictures for each CG was presented four times because every word was presented in each list.

3.4 RESULTS

3.4.1 Phase I: Assessment of Speech Materials and Development of Lists

In this phase, the speech materials were designed to assess two distinct skills, discrimination of initial consonant and final consonant of monosyllabic words. Therefore, the test and analysis were conducted in two parts that assessed each skill separately. Three measures were utilized to evaluate the appropriateness and intelligibility of the developed materials. The first measure was the scores of participants in each CG, where scoring was calculated by adding the number of correct words within a CG, then dividing it by the total number of words within a CG producing an average score for each CG (

Figure 3-3). Even though the test was presented at a soft level (50 dB SPL), participants' scores were overall high with an average of 3.4 points out of 4 for initial phonemes CGs and 2.8 points out of 4 for final phonemes CGs. Scores in the final phoneme component were consistently lower, with the lowest score was for the word /bar/. The word /bar/, which means land (particularly referring to desert in Saudi Arabic) is pronounced the same in the colloquial and Modern Standard Arabic and is a very familiar word to children in Saudi Arabia. It is worth noting that such a low score was not caused due to the use of Modern Standard Arabic language but potentially due to the difficulty of perception of the acoustic cues for /bar/ when presented at a low level.

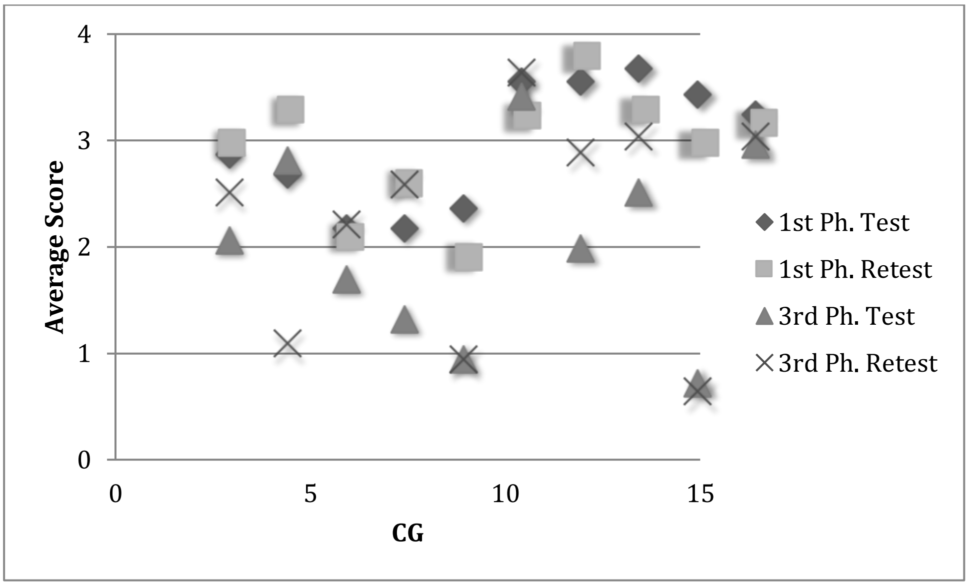


Figure 3-3: the scatter plot depicts Scores for each CG in the initial phoneme (♦ ■) and final phoneme (Δ ×) in the test and retest sessions.

The second measure was intraclass correlation coefficient (ICC) to assess the agreement between the participants' scores within each CG, where responses of all participants for each CG were assessed to evaluate the degree of agreement between participants. We used two-way mixed-effects ICC model with type consistency to assess agreement between subjects' averaged scores at each CG in the test and retest runs. The ICC showed excellent agreement of 0.94 between the participants in both the initial and final phoneme components, suggesting a significant correlation between participants' responses in each CGs.

The third measure was the comparison of the average scores of each CG. Two ANOVA were conducted, one for initial phoneme and one for final phoneme. For both, a repeated measures ANOVA with within-subjects factors of Test session (one and two) and Word group (1 to 10) were used. If the Mauchley's test of Sphericity was significant we used Greenhouse-Geisser corrections. For the initial phoneme, there was no significant effect of Test session, F (1, 22) = 0.04, p = 0.84, but a significant main effect of Word group, F (4.371, 96.168) = 14.589, p < 0.001. Results also showed significant interactions between Test session and Word group, F (5.056, 111.233) = 2.981, p = .014. For the final phoneme contrast there was no significant main effect of Test session F(1, 18) = 0.945, p = 0.344 but there was a significant main effect of Word group , F (9, 162) = 14.608 p < 0.001. Results also showed significant interactions between Test session and Word Group, F (9,96.966) = 7.580, p < .001.

A post-hoc multivariant analysis with Bonferroni adjusted alpha equals to 0.005 (0.05/10) was conducted to evaluate the interaction between Word group and Test session for the initial phoneme contrast and results revealed no significant change in scores within CGs on test and retest (Table 3-2). A post-hoc multivariant Analysis with Bonferroni adjusted alpha equals to 0.005 (0.05/10) was also conducted to evaluate the interaction between Word group and Test session for the final phoneme contrast and revealed a significant improvement in scores within CG4 on retest (Table 3-3). This significant change in performance in retest could be caused by external factors such as sudden background noise and technical errors, or could be reflective of genuine improvement. A conclusion cannot be drawn since such improvement only encountered in one CG (CG4).

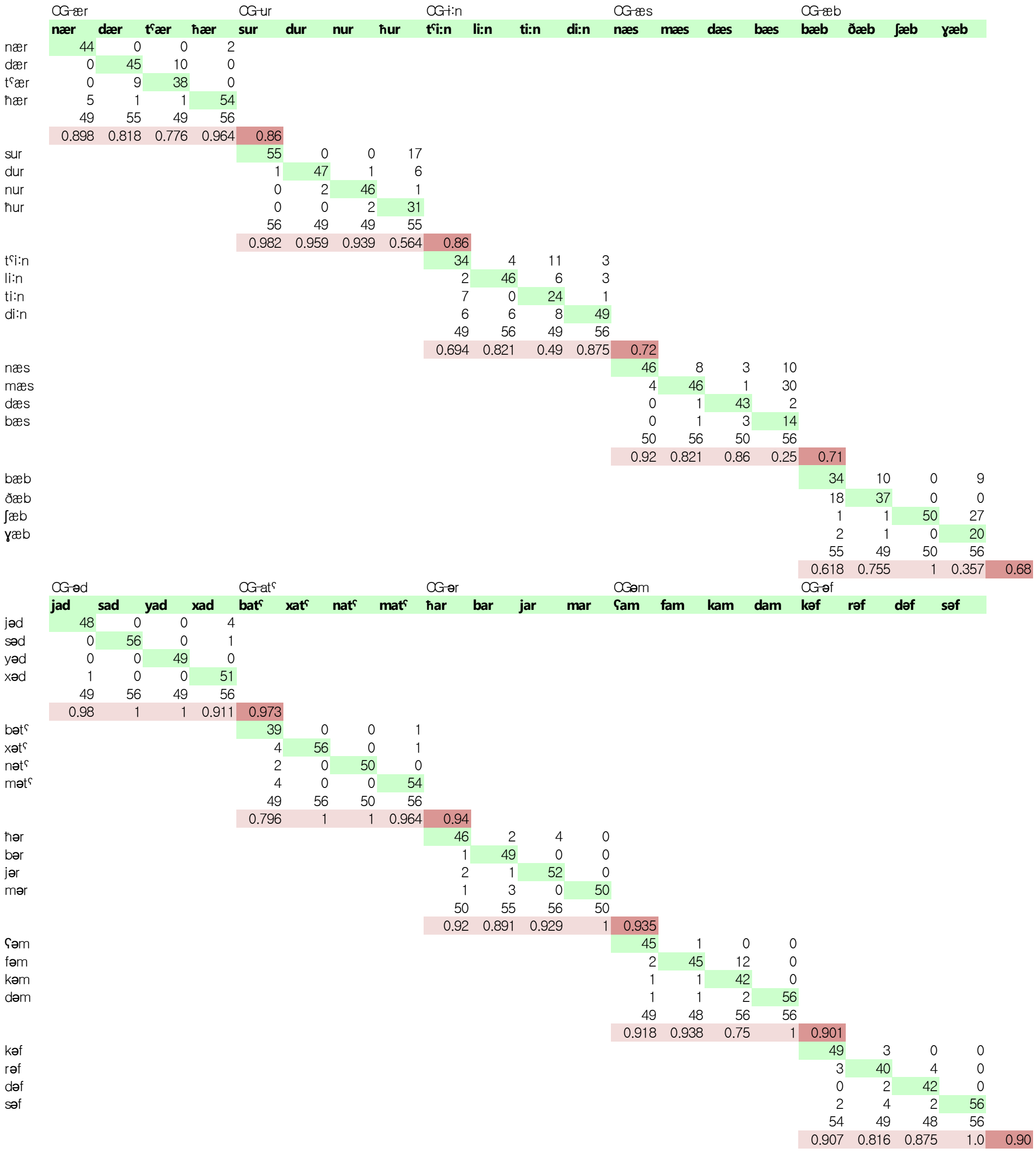
Table 3-2: Table shows the mean scores, standard deviation, F test and significance values for each CG for initial phoneme test and retest sessions.

Descriptive Statistics					**Between-Subjects Effects**		
CGs	**Tests**	**Mean**	**St. Deviation**	**N**	**df**	**F-test**	**Significance**
CG1	Test	3.33	0.7	24	1	0.13	0.72
	Retest	3.42	0.88	24			
CG2	Test	3.21	0.66	24	1	0.45	0.02
	Retest	3.63	0.58	24			
CG3	Test	2.88	0.8	24	1	0.02	0.88
	Retest	2.83	1.05	24			
CG4	Test	2.88	0.95	24	1	1.57	0.22
	Retest	3.17	0.64	24			
CG5	Test	3	0.72	24	1	2.24	0.14
	Retest	2.71	0.62	24			
CG6	Test	3.79	0.41	24	1	1.52	0.22
	Retest	3.58	0.72	24			
CG7	Test	3.79	0.51	24	1	2.22	0.14
	Retest	3.96	0.2	24			
CG8	Test	3.88	0.34	24	1	3.37	0.07
	Retest	3.63	0.58	24			
CG9	Test	3.71	0.55	24	1	2.80	0.10
	Retest	3.42	0.65	24			
CG10	Test	3.58	0.65	24	1	0.05	0.83
	Retest	3.54	0.66	24			

Table 3-3: Table shows the mean scores, standard deviation, F test and significance values for each CG for final phoneme test and retest sessions.

Descriptive Statistics					Between-Subjects Effects		
CGs	**Tests**	**Mean**	**Std. Deviation**	**N**	**df**	**F**	**Sig.**
CG1	test	2.80	0.77	20	1	1.27	0.27
	retest	3.10	0.91	20			
CG2	test	3.30	0.86	20	1	18.79	0.00
	retest	2.15	0.81	20			
CG3	test	2.55	0.60	20	1	1.74	0.20
	retest	2.90	1.02	20			
CG4	test	2.30	0.73	20	1	12.07	0.00
	retest	3.15	0.81	20			
CG5	test	2.05	0.89	20	1	0.00	1.00
	retest	2.05	0.94	20			
CG6	test	3.70	0.73	20	1	0.67	0.42
	retest	3.85	0.37	20			
CG7	test	2.75	0.72	20	1	5.63	0.02
	retest	3.35	0.88	20			
CG8	test	3.10	0.79	20	1	1.74	0.20
	retest	3.45	0.89	20			
CG9	test	1.90	0.72	20	1	0.03	0.86
	retest	1.85	0.99	20			
CG10	test	3.40	0.68	20	1	0.05	0.82
	retest	3.45	0.69	20			

To show the average score of CGs, confusion matrices (CM) for both the initial (Figure 3-4) and final phoneme components (Figure 3-5) were created to illustrate the overall performance of the children. These CMs were used to analyse the similarities and differences between CGs within each test to inform the development of the final word lists. After analysing the CGs within both matrices, the CGs that had lower average scores were considered difficult and were selected for a harder list. All CGs were sorted based on its level of difficulty from least confusing to most confusing, then four lists were developed, two of which were easy and two were hard.



CG-ær	nær	dær	tˤær	ħær	
nær	44	0	0	2	
dær	0	45	10	0	
tˤær	0	9	38	0	
ħær	5	1	1	54	
	49	55	49	56	
	0.898	0.818	0.776	0.964	0.86

CG-ur	sur	dur	nur	ħur	
sur	55	0	0	17	
dur	1	47	1	6	
nur	0	2	46	1	
ħur	0	0	2	31	
	56	49	49	55	
	0.982	0.959	0.939	0.564	0.86

CG-iːn	tˤiːn	liːn	tiːn	diːn	
tˤiːn	34	4	11	3	
liːn	2	46	6	3	
tiːn	7	0	24	1	
diːn	6	6	8	49	
	49	56	49	56	
	0.694	0.821	0.49	0.875	0.72

CG-æs	næs	mæs	dæs	bæs	
næs	46	8	3	10	
mæs	4	46	1	30	
dæs	0	1	43	2	
bæs	0	1	3	14	
	50	56	50	56	
	0.92	0.821	0.86	0.25	0.71

CG-æb	bæb	ðæb	ʃæb	ɣæb	
bæb	34	10	0	9	
ðæb	18	37	0	0	
ʃæb	1	1	50	27	
ɣæb	2	1	0	20	
	55	49	50	56	
	0.618	0.755	1	0.357	0.68

CG-əd	jad	sad	yad	xad	
jəd	48	0	0	4	
səd	0	56	0	1	
yəd	0	0	49	0	
xəd	1	0	0	51	
	49	56	49	56	
	0.98	1	1	0.911	0.973

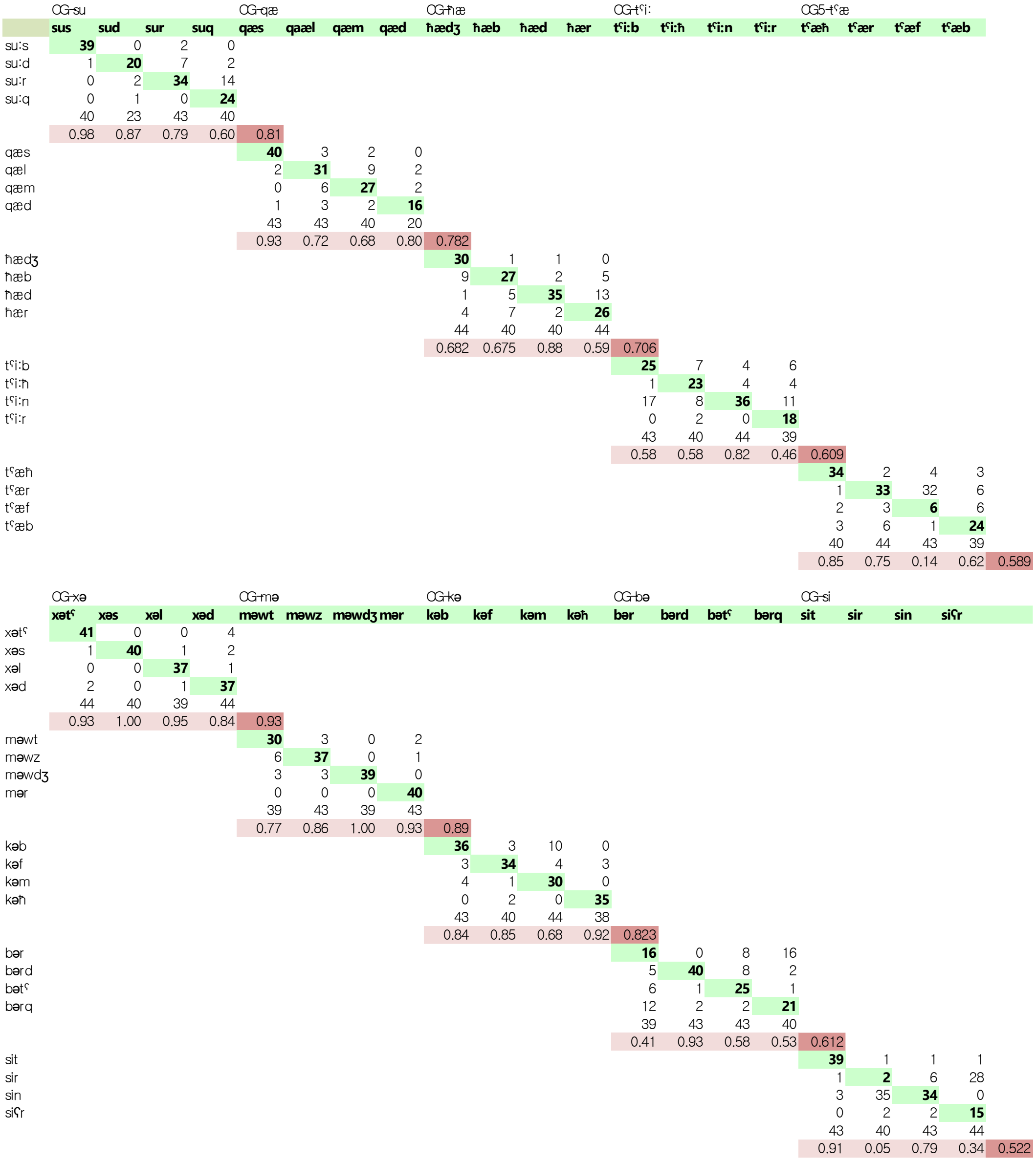
CG-atˤ	batˤ	xatˤ	natˤ	matˤ	
bətˤ	39	0	0	1	
xətˤ	4	56	0	1	
nətˤ	2	0	50	0	
mətˤ	4	0	0	54	
	49	56	50	56	
	0.796	1	1	0.964	0.94

CG-ər	ħar	bar	jar	mar	
ħər	46	2	4	0	
bər	1	49	0	0	
jər	2	1	52	0	
mər	1	3	0	50	
	50	55	56	50	
	0.92	0.891	0.929	1	0.935

CGəm	ʕam	fam	kam	dam	
ʕəm	45	1	0	0	
fəm	2	45	12	0	
kəm	1	1	42	0	
dəm	1	1	2	56	
	49	48	56	56	
	0.918	0.938	0.75	1	0.901

CG-əf	kəf	rəf	dəf	səf	
kəf	49	3	0	0	
rəf	3	40	4	0	
dəf	0	2	42	0	
səf	2	4	2	56	
	54	49	48	56	
	0.907	0.816	0.875	1.0	0.90

Figure 3-4: Confusion matrix for initial phoneme test. The words listed horizontally are the words presented to the subjects, while the words listed vertically represents the responses of the subjects. Diagonally, all groups are ordered based on their average scores from highest to lowest (highlighted in red). The top panel shows the groups with long vowels whereas the lower panel shows the groups with short vowels.



	OG-su				OG-qæ				OG-ħæ				OG-tˤi:				OG5-tˤæ				
	sus	**sud**	**sur**	**suq**	**qæs**	**qaæl**	**qæm**	**qæd**	**ħædʒ**	**ħæb**	**ħæd**	**ħær**	**tˤi:b**	**tˤi:ħ**	**tˤi:n**	**tˤi:r**	**tˤæħ**	**tˤær**	**tˤæf**	**tˤæb**	
su:s	**39**	0	2	0																	
su:d	1	**20**	7	2																	
su:r	0	2	**34**	14																	
su:q	0	1	0	**24**																	
	40	23	43	40																	
	0.98	0.87	0.79	0.60	0.81																
qæs					**40**	3	2	0													
qæl					2	**31**	9	2													
qæm					0	6	**27**	2													
qæd					1	3	2	**16**													
					43	43	40	20													
					0.93	0.72	0.68	0.80	0.782												
ħædʒ									**30**	1	1	0									
ħæb									9	**27**	2	5									
ħæd									1	5	**35**	13									
ħær									4	7	2	**26**									
									44	40	40	44									
									0.682	0.675	0.88	0.59	0.706								
tˤi:b													**25**	7	4	6					
tˤi:ħ													1	**23**	4	4					
tˤi:n													17	8	**36**	11					
tˤi:r													0	2	0	**18**					
													43	40	44	39					
													0.58	0.58	0.82	0.46	0.609				
tˤæħ																	**34**	2	4	3	
tˤær																	1	**33**	32	6	
tˤæf																	2	3	**6**	6	
tˤæb																	3	6	1	**24**	
																	40	44	43	39	
																	0.85	0.75	0.14	0.62	0.589

	OG-xə				OG-mə				OG-kə				OG-bə				OG-si				
	xətˤ	**xəs**	**xəl**	**xəd**	**məwt**	**məwz**	**məwdʒ**	**mər**	**kəb**	**kəf**	**kəm**	**kəħ**	**bər**	**bərd**	**bətˤ**	**bərq**	**sit**	**sir**	**sin**	**siʕr**	
xətˤ	**41**	0	0	4																	
xəs	1	**40**	1	2																	
xəl	0	0	**37**	1																	
xəd	2	0	1	**37**																	
	44	40	39	44																	
	0.93	1.00	0.95	0.84	0.93																
məwt					**30**	3	0	2													
məwz					6	**37**	0	1													
məwdʒ					3	3	**39**	0													
mər					0	0	0	**40**													
					39	43	39	43													
					0.77	0.86	1.00	0.93	0.89												
kəb									**36**	3	10	0									
kəf									3	**34**	4	3									
kəm									4	1	**30**	0									
kəħ									0	2	0	**35**									
									43	40	44	38									
									0.84	0.85	0.68	0.92	0.823								
bər													**16**	0	8	16					
bərd													5	**40**	8	2					
bətˤ													6	1	**25**	1					
bərq													12	2	2	**21**					
													39	43	43	40					
													0.41	0.93	0.58	0.53	0.612				
sit																	**39**	1	1	1	
sir																	1	**2**	6	28	
sin																	3	35	**34**	0	
siʕr																	0	2	2	**15**	
																	43	40	43	44	
																	0.91	0.05	0.79	0.34	0.522

Figure 3-5: Confusion matrix for final phoneme test. The words listed horizontally are the words presented to the subjects, while the words listed vertically represents the responses of the subjects. Diagonally all groups are ordered based on their average scores from highest to lowest (highlighted in red). The top panel shows the groups with long vowels whereas the lower panel shows the groups with short vowels.

3.4.2 Phase II: Validation of the Developed Lists

3.4.2.1 Lists Equivalency Analysis

Using the lists (two easy and two hard) that were developed in phase I, an experiment was conducted to evaluate the difficulty and equivalency across lists. The collected data at 40- and 50- dB SPL was used to evaluate the list equivalency and produce the final form of the lists since the difference between the lists was most apparent at lower presentation levels and ceiling effect was observed in the higher presentation levels (Table 3-4). A repeated-measure ANOVA was performed with a within-subject factor Test Sessions (test and retest) and Lists (four lists) and Sphericity was assumed.

Table 3-4: The participants' scores in ratios at 40, 50, 60, and 70 dB SPL are shown for the four lists.

dB SPL /Lists	L1	L2	L3	L4
40	0.74	0.71	0.66	0.56
50	0.93	0.90	0.89	0.86
60	0.94	0.93	0.95	0.83
70	0.92	0.95	0.92	0.89

The results indicated a significant main effect of List, F (3, 13) = 12.1, $p < 0.001$ and a significant main effect of Test Sessions, F (1, 15) = 5.54, $p = 0.03$, suggesting the existence of different level of difficulty within lists and improvements in performance on retest sessions. The results showed no significant interaction between the effects of Lists and Test Sessions on scores, F (3, 13) = 0.96, $p = 0.44$. Post-hoc comparisons using the Tukey HSD test indicated that the mean scores for

List D was significantly lower than the mean scores of List A ($p < 0.001$) and List B ($p < 0.001$). In addition, the mean score of List C was significantly lower than the mean scores of List B ($p = 0.03$) and List A ($p = 0.05$) (Figure 3-6). Results indicated that List A and List B were easier than List C List D. To maximize the reliability of the test by increasing the number of items within lists, it was decided to combine the two easy lists into one list of 32 items and the two hard lists into one list of 32 items (See Table 3-5). In addition, conducting the test multiple of times in different orders or using practice runs could minimize learning effects and improve reliability.

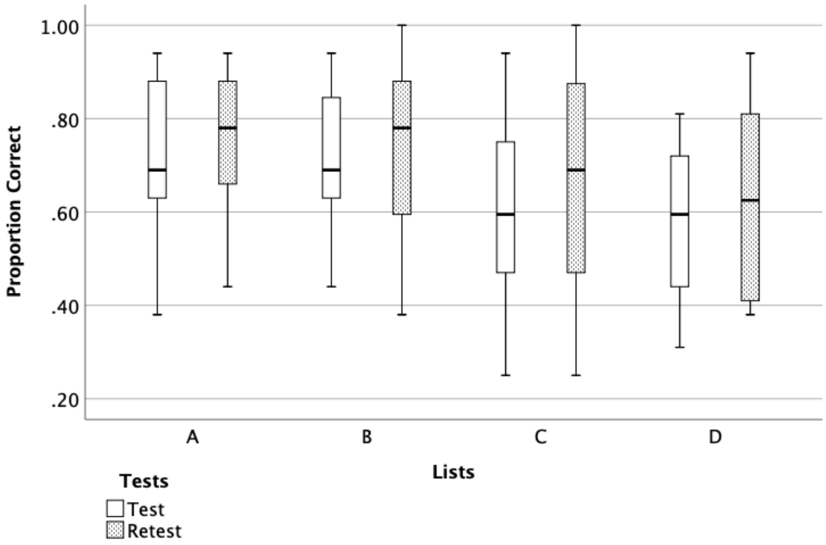


Figure 3-6: In the box plots, the Y axis represents the proportion correct score and the X axis represents the developed four lists. The boundary of the box closest to zero indicates the 25th percentile, a black line within the box marks the median, and the boundary of the box farthest from zero indicates the 75th percentile. Whiskers above and below the box indicate the 10th and 90th percentiles.

Table 3-5: Two lists were developed, an easy List and a hard List

A-CAPT Lists			
Easy List		**Hard List**	
/suːr/	/naːr/	/naːs/	/tˤiːn/
/duːr/	/daːr/	/maːs/	/liːn/
/nuːr/	/tˤaːr/	/daːs/	/tiːn/
/ħuːr/	/ħaːr/	/baːs/	/diːn/
/jad/	/batˤ/	/ʕam/	/ħar/
/sad/	/xatˤ/	/fam/	/bar/
/yad/	/natˤ/	/kam/	/jar/
/xad/	/matˤ/	/dam/	/mar/
/xatˤ/	/suːs/	/ħaːdʒ/	/tˤiːb/
/xas/	/suːd/	/ħaːb/	/tˤiːħ/
/xal/	/suːr/	/ħaːd/	/tˤiːn/
/xad/	/suːq/	/ħaːr/	/tˤiːr/
/qaːs/	/mawt/	/kab/	/bar/
/qaːl/	/mawz/	/kaf/	/bard/
/qaːm/	/mawdʒ/	/kam/	/batˤ/
/qaːd/	/mar/	/kaħ/	/barq/

3.4.2.2 Critical difference and Correlation Analysis

3.4.2.3 Critical Difference

A within-subject sω (Bland & Altman, 1996) was calculated to derive the 95% confidence interval of the score for an individual. The quantity sω is the square root of the mean group variance (mean across individuals of the variance calculated for each individual). An individual's observed score is expected to lie within $\pm 1.96\ s\omega$ of their true score (for 95% of observations; the CI). The critical difference is calculated as $\sqrt{2}*1.96s\omega$. If scores obtained on two different occasions differ by $\sqrt{2}*1.96\ s\omega$ or more, then they differ significantly at $p < 0.05$.

The critical difference was calculated for both levels 40- and 50-dB SPL. The mean critical difference at 40 dB SPL for the easy list scores was 18% and for the hard list scores was 28%. The mean critical difference at 50dB SPL for the easy list was 18% and the hard list was 12%. Since 40 dB SPL is considered soft speech and could be affected by variation of hearing thresholds and noise floor as observed by the participants' performance (see Figure 3-7), we decided to present the critical difference at 50 dB SPL.

Table 3-6 shows how the critical difference varies across the performance range at 50 dB SPL and what the critical difference is for an individual score out of 32. For example, a child scored 88% on the easy list one occasion and 77% in the second. This would not be considered as a significant change in performance since the score falls between 69% and 100%. However, if the child scored 66% on the second occasion, this would be viewed as a significant decrease in performance.

Table 3-6: Critical Differences (CD) for easy and hard lists expressed in percentages (95% confidence interval)

Easy List at 50dB SPL	**CD -+18 %**		**Hard List at 50dB SPL**	**CD -+12%**	
Scores (out of 32, %)	**Lower Boundary**	**Upper boundary**	**Scores (out of 32, %)**	**Lower boundary**	**Upper boundary**
100	82	100	100	88	100
97	79	100	97	85	100
94	76	100	94	82	100
91	72	100	91	79	100
88	69	100	88	76	100
84	66	100	84	73	100
81	63	99	81	69	93
78	60	96	78	66	90
75	57	93	75	63	87
72	54	90	72	60	84
69	51	87	69	57	81
69	51	87	69	57	81
66	47	84	66	54	77
63	44	81	63	51	74
59	41	78	59	48	71
56	38	74	56	44	68
53	35	71	53	41	65
50	32	68	50	38	62
47	29	65	47	35	59
44	26	62	44	32	56
41	22	59	41	29	52
38	19	56	38	26	49
34	16	53	34	23	46
31	13	49	31	19	43
28	10	46	28	16	40

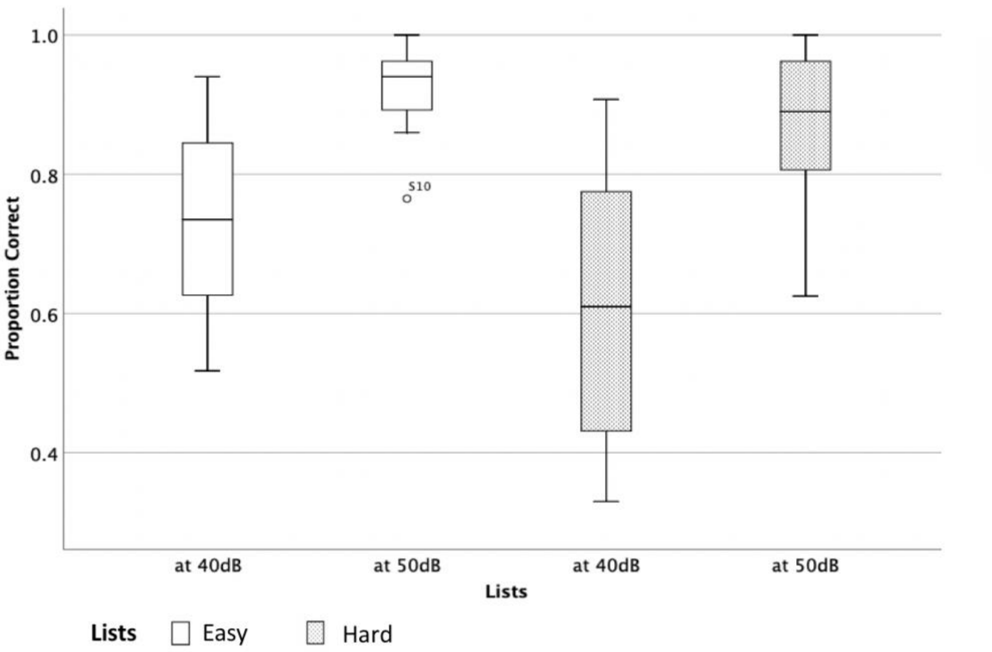
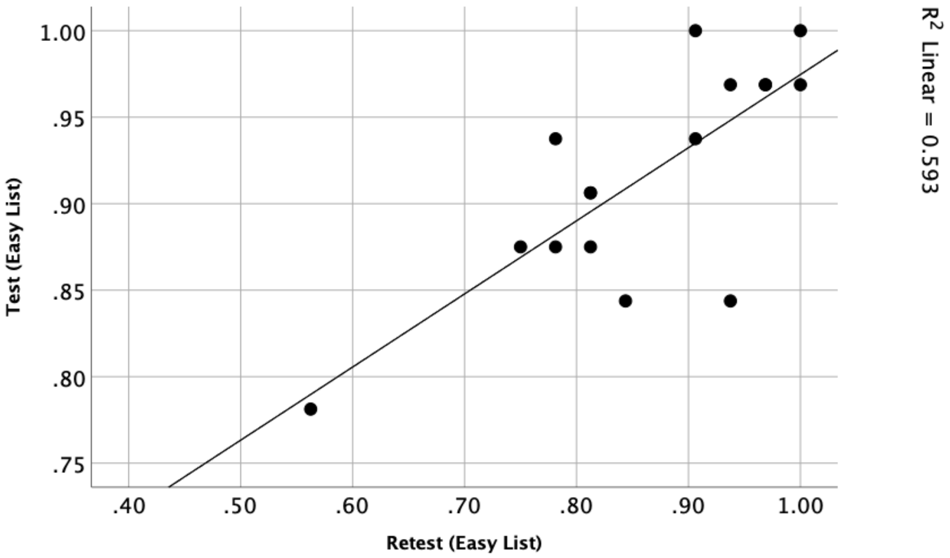


Figure 3-7: Proportion correct scores for the easy and hard lists at 40 and 50 dB SPL

3.4.2.4 Test-retest Reliability and Age Effect

Pearson correlation was conducted to assess test-retest reliability and showed significant correlations at 50-dB SPL for easy ($r = 0.77$, $p < 0.001$) and hard ($r = 0.79$, $p < 0.001$) lists (Figure 3-8). Pearson correlations was also conducted to evaluate the relationship between participants' performance and age at 50 dB SPL ($n = 16$) (Figure 3-9). The findings revealed significant correlations between age and subjects' scores at easy list ($r = 0.63$, $p = 0.01$) and hard list ($r = 0.62$ $p = 0.01$).



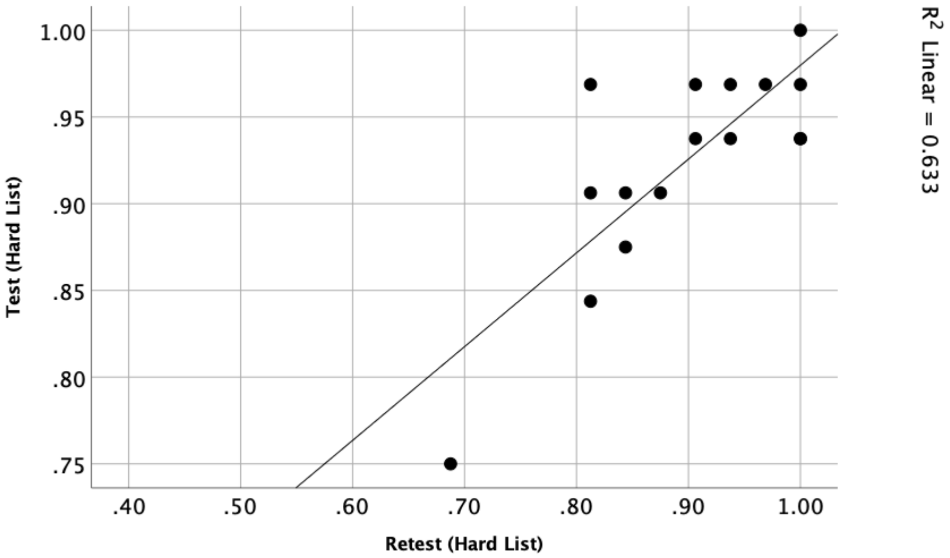
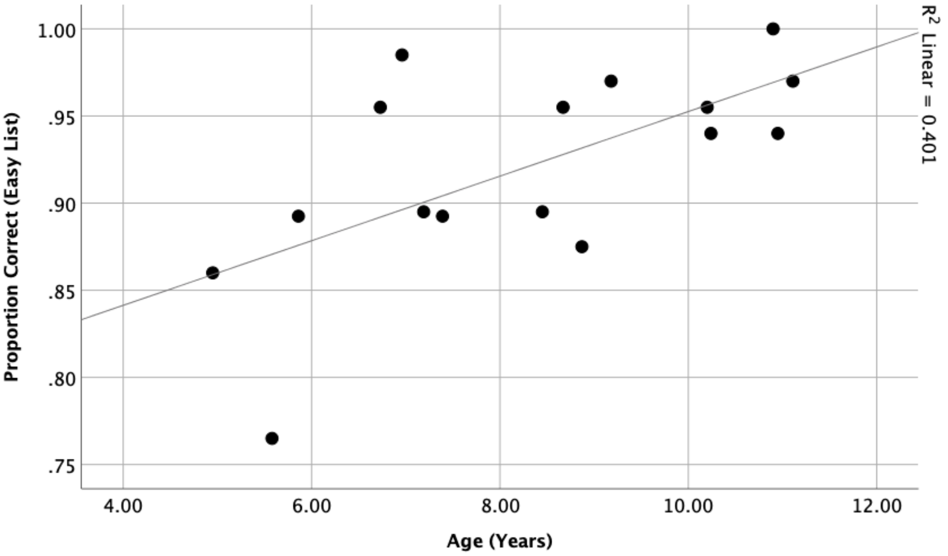


Figure 3-8: The scatter plots depict the relationship between test and retest 50 dB SPL at easy (top) and hard list (bottom)



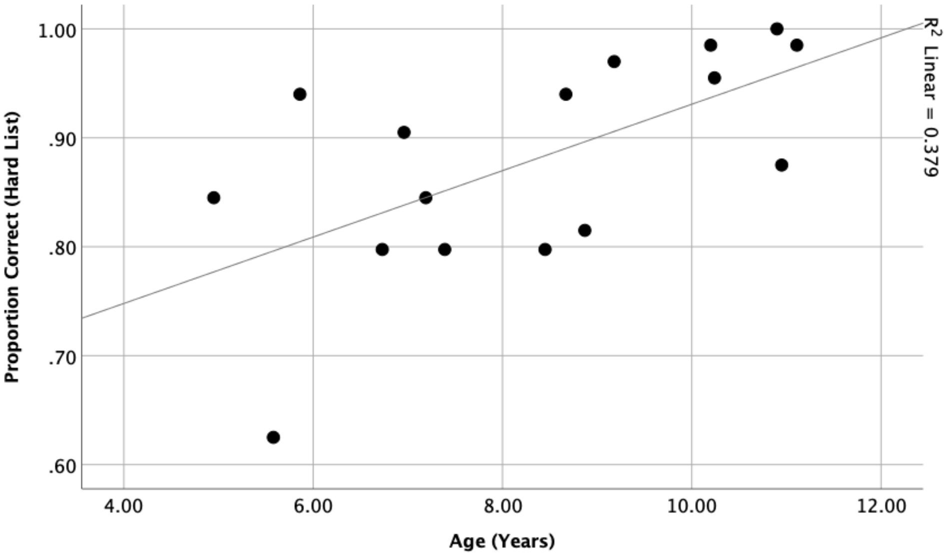


Figure 3-9: The scatter plots depict the relationship between age and proportion correct scores at easy (top) and hard (bottom) lists at 50 dB SPL

3.5 DISCUSSION

This research developed and validated a new Arabic monosyllabic closed-set consonant-discrimination test, for use with Arabic speaking children in Saudi Arabia between the ages of five and eleven years. The main motivation for this research was the lack of validated materials that can be used with this population to assess their consonant perception skills and monitor changes over time or after an intervention. The new test is an Arabic version of the CAPT (Vickers et al., 2018).

Selecting the format of the speech test is key to ensure that the measurement is assessing the desired function in a reliable way (Bergeson, Pisoni, & Davis, 2005; Clopper, Pisoni, & Tierney, 2006). We therefore decided to use a closed-set format because this makes the task a discrimination task, which is easier for younger children, not reliant upon a child’s ability to produce sounds, and shows how different phonemes are confused. This analytical information can be used to guide fitting to optimise delivery of speech information. The mode of delivery of this test was auditory-only in a quiet environment following the standard approach used for the CAPT (Vickers et al., 2018). We chose to adapt the CAPT because it has been shown to be a reliable outcome measure for assessing speech perception in young children and is sensitive to hearing aid gain settings (Marriage & Moore 2003) and differences in the audiogram (Lovett et al., 2015). It also can be used for monitoring performance of children overtime. Measuring auditory perception in children can be dependent on their knowledge of the vocabulary used in the CAPT. However, when assessing children’s performance over time, their auditory perception is less dependent on their knowledge of the vocabulary because of the comparison is across multiple testing sessions.

To develop this word discrimination test, the word familiarity and language level was determined to be aligned with recommendations from other authors (Bergeson, Pisoni, & Davis, 2005; Clopper, Pisoni, & Tierney, 2006). This was done by presenting the selected Arabic words in Modern Standard Arabic to a group of primary-school Saudi Arabian children to assess the familiarity and appropriateness of the words and the clarity of the corresponding pictures. The scores of the children in the development stage were generally very high suggesting that the pictures and words were intelligible for the majority of normal hearing participants. This part of the study was conducted in the UK at the KFA, a Saudi-funded primary school. The children at this school who partook in this experiment were from Saudi Arabia and were exposed to the same type of Modern Standard Arabic as those children who participated in the final phase of the study that was conducted in Saudi Arabia. The only difference between these two groups was the fact that most of the children who participated in the study that was conducted in the UK were bilingual while most of the children who participated in the study conducted in Saudi Arabia were monolingual. Such a difference is not expected to be an issue since all the children were native Arabic speakers who were equally exposed to Modern Standard Arabic.

It is important to note that this scenario illustrates the benefits of using the formal form of the language with school-aged children when dealing with diglossic languages such as Arabic, Modern Greek, Swiss German, and Haitian Creole. This study showed that despite the existence of various number of dialects within the Saudi-Arabic language, children with different backgrounds were familiar with the materials that were presented in a closed-set format in the formal form of the language and achieved high scores in this test. This may also indicate that the test can be used with Arabic speaking children in non-Arabic speaking countries. For example, it has been reported that Arabic is the fastest growing language in the United States (Brown, 2016). In addition, providing such tests can minimize the reported lack of materials for assessing speech in children who lives in the UK but speaks languages other than English (Cattani et al., 2014). In large cities, it has been reported that the proportion of children who have English as not their first language can be higher than those who have English as the family language (Mehta et al., 2017).

The final version of the test consisted of two lists, one was considered ‘easy’ and one ‘hard’. For each of these lists the measures were shown to have strong agreement between the test and retest sessions with Pearson correlation values of 0.77 and 0.79 for the easy and hard lists, respectively; these values were similar to the reported value (0.83) in the original CAPT (Vickers et al., 2018).

The effect of age is a critical element to evaluate when validating a speech perception measure (Bergeson et al., 2005; Clopper et al., 2006). There was a significant correlation between the scores on the test and the age of the participants, with older children performing better than younger children. Such trend was expected as older children with normal hearing are reported to have better speech discrimination skills and advanced spectral resolution maturity compared to younger children (Horn et al., 2017; Rayes, Sheft, & Shafiro, 2014). Age was accounted for approximately 40% of the variance for both the easy and hard lists. The relationship did not reach a stronger level possibly due to the small number of sample (n = 16) that was used in this experiment. However, this relationship was stronger than the reported in the original CAPT (Vickers et al., 2018), which only accounted for 15% of the variance and was explained by the limited spread of ages of participants in the CAPT (Vickers et al., 2018).

The critical differences for the two lists can be used to determine whether changes in performance of an individual child are significant. The critical difference values reported by Vickers et. al (2018) for the CAPT was 13.7%, which is similar to the critical values that we calculated for the hard list (12%). For the easy list, the average critical difference was larger (18%). These critical difference values indicate that the harder list is a better discriminator when comparing an individual child's performance in two different conditions

Finally, limitations of the test and study should be noted. First, the critical differences are larger than would be desired but in a similar region to the CAPT (Vickers et al., 2018). This is typical for measures used with young children. It does mean that in an ideal clinical situation that two lists would be conducted to improve the confidence in the scores or at least a short practice list is needed prior to running the actual test. As with all speech measures for children, it means that on the individual level small changes in performance will not be detected. The critical differences were larger at the lower level (40 dB SPL compared to 50 dB SPL) and this is again as expected because the children were being tested at a lower point on the psychometric function closer to their hearing threshold, where greater variability is typically observed. Third, this test is conducted in Modern Standard Arabic, which is not the everyday mother tongue spoken language by Saudi Arabic speaking children, but rather a formal form used in media and at school. It was selected because the variation in dialects for Saudi Arabic is vast and the Modern Standard Arabic can be considered as a common ground that everyone is exposed to on daily basis, including the selected age group children. However, we believe that further work is needed to develop speech materials for younger children using regional dialects. In addition, a version of the A-CAPT should be validated with the words presented in background noise.

3.6 Conclusion

This A-CAPT has been developed to assess consonant perception in Saudi Arabic speaking children aged 5 years and older. The test consists of an easy list and a hard list, which were validated with normal-hearing children (aged 5 to 11 years). Test–retest reliability was good for both the easy and hard lists. Overall, children's performance improved with increasing age. Just like the CAPT (Vickers et al., 2018) from which it was adapted, the A-CAPT uses a wide range of phonemes in a speech discrimination task that will be helpful when programming hearing devices or planning an intervention.

CHAPTER 4: IMPACT OF AUDITORY TRAINING ON SPEECH PERCEPTION ABILITIES OF CHILDREN WITH COCHLEAR IMPLANTS

4.1 OVERVIEW

Although hearing devices may help children with hearing loss to access sound, the technology still not equivalent to normal hearing and sometimes fail to discriminate between similar auditory input especially in challenging auditory environments. To maximize the benefits of hearing devices, AT is often recommended. This chapter describe an auditory intervention, HIBA, for use with school-aged Arabic-speaking children. A multi-modal parent-delivered AT intervention was evaluated and the benefits of speech and pitch perception outcomes in children with CIs were assessed through a RCT.

4.2 INTRODUCTION

Although CIs may restore hearing to children with severe-to-profound hearing loss and help them to access sound, acquiring speech, developing language, and attaining effective communication requires more than just accessing sound. Even with quite similar auditory input via their CIs, processing of the incoming signal may vary between listeners. Variations of the ability to process the auditory input is reflected in substantial variation of outcomes in children with CIs. There are various factors that can influence the outcomes of auditory and speech perception in children with CIs.

The main factors predicting CIs outcomes in children including onset of deafness or language acquisition status prior to cochlear implantation (Kane et al., 2004), age of implantation, the level of residual hearing before implantation, quality of parent-child interactions, socioeconomic status and maternal education level (Baker & Hazan, 2011; Connor et al., 2006; Miyamoto et al., 2017; Niparko et al., 2010; Yoshinaga-Itano et al., 2018). In other words, differences between abilities of children with CI in decoding auditory inputs can be related to the integrity of auditory system, brain development status, previous exposure to auditory input, cognition, and family support.

In an effort to enhance the outcomes of children with CIs who do not do well and to narrow the performance gap between children, AT programmes have been utilized to accelerate the development of auditory and speech perception. Evidence of neuroplasticity has been established in children after participating in an AT intervention (Hayes, Warrier, Nicol, Zecker, & Kraus, 2003; Russo, Nicol, Zecker, Hayes, & Kraus, 2005). AT programmes have the potential to maximize benefit for people using hearing devices if appropriately designed and implemented and have been shown to promote the development of auditory abilities that are the basis for oral language acquisition and development (Kral & Sharma, 2012; Rayes et al., 2019) helping children to attain language development on a par with hearing peers (Tye-Murray, 2019).

The purpose of this research was to develop an evidenced-based parent-led AT intervention and determine whether it can lead to improvements in speech and pitch perception in children with CIs.

4.2.1 Needs for AT Intervention

Children with hearing loss show delay in both pre-literacy skills and spoken language; they perform poorly compared to children with typical hearing on measures of oral language, phonological memory, and conceptual print knowledge (Werfel, 2017). By the time they start high school, their reading proficiency level is delayed by up to five years compared to their normal hearing peers, and by the end of high school, half of hard-of-hearing students demonstrate reading levels below that of fourth grade (8-9 years old) (Qi & Mitchell, 2012). Reading comprehension in 106 deaf adults was also assessed and on average showed to be 6.2 grade reading level (Zazove, Meador, Reed, & Gorenflo, 2013). The use of CIs has decreased the observed delay in reading development and children with CIs frequently have more advanced reading skills than their deaf peers who use hearing aids, nonetheless they are still delayed when compared to their age-matched normal hearing peers (Marschark, Rhoten, & Fabich, 2007; Werfel, 2017). With the intention of reducing the observed delays with CIs, rigorous habilitation programmes needs to be accessible, readily available to children with CI, and individualized when necessary to meet their needs (Tye-Murray, 2019, p. 401).

In the majority of countries, millions of dollars (or the equivalent) have been invested in cochlear implantation but very little funding has been provided for post-implantation rehabilitation. This is also the case in Saudi Arabia, where there are limited tools available for effective AT and research for the development of such tools is scarce. The impoverished access to AT programmes may negatively influence outcomes for children with CIs. Therefore, the development of appropriate (re)habilitation tools is needed to fill the gap in knowledge and provide effective resources and protocols that can be used clinically.

4.2.2 Factors Considered when Developing AT Intervention

4.2.2.1 Range of AT Approaches

There are two overall approaches used, namely, bottom-up (analytic) and top-down (synthetic). For speech training, the bottom-up approach uses context-free acoustic phonetic signals to train the listener to decode the speech signal without context. In contrast, the top-down approach relies on the listeners' linguistic knowledge to be able to process the perceived cues and fill in the gaps. Both approaches can be beneficial for AT programmes whether utilised separately or in combination; a trend toward combining both approaches has been suggested to enhance AT outcomes (Rayes et. al, 2019, Amitay et al., 2006).

4.2.2.2 Type of AT Tasks

Various studies have shown that AT can improve auditory and speech perception in children but there are no clear guidelines of the parameters, including stimulus type and difficulty of the tasks, needed to develop an effective AT intervention. A review by Moore & Amitay, (2007) presented methods to optimize AT for children by varying stimuli type and difficulty of the tasks.

Small variation in the type stimuli in pitch discrimination training yielded to reduced learning for all listeners while large variation in the stimuli produced a different pattern according to the listeners' initial performance. Children who initially performed better at the baseline, large variations in type of stimuli was as effective as no variation training, while children who performed poorly in the initial assessments, performed badly at small and large variations in stimuli type. This may emphasize the need for individualized AT as some listeners may benefit from limited variation in training stimuli while other would gain benefits even with large variation in the stimuli.

In addition, the authors demonstrated that considerably easy tasks may not lead to significant improvements in learning, however challenging tasks have shown to produce robust learning, highlighting the significance of engaging in active tasks. Furthermore, AT may include non-auditory tasks as one of the experiments demonstrated that playing a visual-spatial task could also lead to improved performance on the auditory domain, suggesting that arousal and maintained attention on their own may improve listening skills.

Stacey & Summerfield (2008) assessed the effectiveness of different strategies for AT that could improve speech perception in adult with CIs. Normal hearing adults participated in the training which utilized vocoded speech (phonemes, words, and sentences) to simulate the input provided by CIs. The study found that word- and sentence-based training were equally effective and led to significant improvements in recognition of words in sentences, phoneme-based training were not as effective. No significant improvements in discrimination of phonemes (consonant or vowel) were observed post training. Findings of this study suggested that word- and sentence-based training were more effective than the phoneme-based training in improving speech perception.

In addition, aspects of speech perception could be improved by training general task such as music or pitch perception. Deficits in reading abilities was linked to poor phonological representation, which was associated with poor pitch perception (Anvari et al, 2002). Evidence of enhancing sound perception (Schlaug et al, 2005) and phonological processing (Verney, 2013) via music training since similar cortical mechanisms for processing sound in both speech and music domains are simultaneously activated in humans' brains (Patel, 2003; Slater et al., 2015; Strait & Kraus, 2011).

Since the studies mentioned above did not include particularly children with CI, the recommendations need to be taken with caution. The development of the auditory system has shown to differ between typically developing children and children with sensory loss, but there is no evidence to dismiss the above proposed rules when developing AT intervention. It is also practical to follow previous AT programs that were made for children with CI and use the tasks that were included in the training programmes and shown to produce benefits. Working memory, speech perception (e.g., phoneme or words identification or discrimination, speech-in-noise perception, words- or sentence- based training), music, pitch and rhythm discrimination, and environmental sounds were amongst the tasks that were trained in children with CI and improvements in all trained tasks were reported across all studies regardless of the approach of training.

To summarize, tasks in an effective AT should be engaging, challenging, individualized as needed and not necessarily targeting specific skill as generalisation of learning has been widely observed.

4.2.2.3 AT Doses Duration

There is flexibility when scheduling AT sessions (Tye-Murray, 2019). Humes, Kinney, Brown, Kiener, & Quigley, (2014) explored a range of training dosages and durations for a word-based training task for adults with hearing loss and revealed that the groups that received the AT session twice or three times per week performed significantly better than the group who did not receive any training. In addition, there was no significant difference between the participants in the two training groups indicating that the difference in dispensing the dosage and duration of the intervention did not affect its outcomes as long as the participants completed the total hours of the training. The study concluded that conducting AT sessions twice per week or three times per week for five to fifteen weeks can be sufficient to lead to measurable improvements. Furthermore, studies that assessed AT intervention in children with CIs reported benefit in all training tasks regardless of the duration of training (Rayes et al., 2019), which ranged from 4 weeks (Ingvalson et al., 2014) up to 2 years (Yucel et al., 2009)

4.2.2.4 Reinforcement and Feedback

Incorporating feedback into AT interventions can maximize its benefits (Tye-Murray, 2019). Feedback, whether orthographic and auditory, showed to be successful method to encourage participants to identify and discriminate stimuli in the AT programmes. Burk & Humes in 2008 investigated the effect of training words in noise on understanding of both trained and untrained words in noise in adults with hearing impairment. Training materials were presented in a closed-set condition with both orthographic and auditory feedback. Improvement in both open- and closed-set recognition was measured post training. Improvements were generalized to unfamiliar talkers but did not transfer to untrained words. Similar gains were observed when whether the feedback was orthographic or a mix of orthographic and auditory, but not when the feedback was absent.

4.2.2.5 Multi-modal Training

When working with infants or toddlers who have no or minimal language experience, AT tends to be more effective if there is a hierarchical structure starting by training sound awareness, then moving onto sound discrimination, sound identification, and finally comprehension (Tye-Murray, 2019, p. 412). This hierarchical method is helpful with young children because it starts with tasks that target fundamental skills such as sound awareness that are needed before mastering more complicated skills such as phoneme discrimination. It's important to know what the sound actually represents before working with those different sounds. An example of hierarchical learning is the acquisition of spoken language, children cannot effectively make sentences if they do not have the appropriate vocabulary building blocks in place. Although this hierarchal training approach has been shown to be effective, linguists may argue that acquisition of spoken language is attained by exposure to continuous stream of speech with minimal pauses between words. Infants have shown to successfully parse the speech stream into meaningful units by 7 months of age using their abilities to detect consistent patterns of sounds through statistical learning. For example, the syllables that are part of the same word tend to follow one another predictably, unlike syllables that span word boundaries. (Saffran, Senghas, & Trueswell, 2001)

In addition, for school-age children who have developed sound awareness skills and have been exposed to oral language, AT does not necessarily have to follow the hierarchical learning approach. When training older children and even adults, multi-training strategies, e.g. phoneme-based training, word-based-training, sentence-based training, and cognitive skill-based training, are often combined into a single training programme to be conducted together at each session (Tye-Murray, 2019, pp. 123–124). For older children variation within a training session is helpful to maintain children's attention and interest. Phoneme-based training relies mainly on bottom-up processing based on accessing acoustic cues requires minimal cognitive processing in terms of expectation and prior knowledge. Word-, sentence- and cognitive- based training utilize top-down processing as it is influenced by expectation, prior knowledge, thinking and problem solving.

Music training also uses top-down processing and many children find it interesting and engaging. Musical games can be designed to carefully adjust sound parameters based on pitch, melody, rhythm or timbre to find an engaging way to train contrasts. Galvin, Fu, & Nogaki, (2007) conducted a musical feature training study with 6 adults with CIs, and trained the identification of melodic contours and showed significant improvements post intervention. In addition, improved melodic contour identification was shown to generalize to improved vowel recognition performance. Welch et al. (2015) utilized the whole song training approach to assess benefits of singing activities on children's hearing acuity and pitch perception, and the intervention led to significant improvements in the perception of pitch changes in complex synthesized piano chords.

4.2.2.6 Parents Involvement

Parents play a prominent role in their children's language development (Hart & Risley, 1995; Smith, Landry, & Swank, 2000; Tamis-LeMonda, Bornstein, & Baumwell, 2001) and are acknowledged as children's first teachers. They have many more opportunities to interact with their children in meaningful everyday situations than the teachers or therapists do. Such interaction promotes learning as children learn to communicate during everyday activities. Huttenlocher, Haight, Bryk, Seltzer, and Lyons (1991) investigated vocabulary growth in typically developing children and revealed a significant positive relationship between the quantity of maternal linguistic input and children's vocabulary growth. Hart and Risley (1995) also observed a positive relationship between the amount of parent talk and the children's vocabulary size. A similar trend was reported by Rowe in 2008 between the extent of child-directed speech and receptive vocabulary skills.

Parents of children who receives speech interventions also play a critical role in their children's speech and language development. Having active parents who positively participate in their children's rehabilitation journey could significantly improve the rehabilitation outcomes. A meta-analysis (Roberts & Kaiser, 2017) that investigated the efficacy of parent-implemented language interventions on children's language skills revealed a significant positive improvement for receptive and expressive language skills of children. In addition, parents' ability to learn strategies and lead parent-implemented intervention is extremely valuable as it makes everyday interaction extemporaneous learning experience. The review by Roberts & Kaiser (2017) reported that parents who were coached, successfully learned communication strategies and used them when interacting with their children and it consequently had a positive effect on their children's communication development. Parents' use of communication strategies led to improvements in their child's verbal and nonverbal expressive skills, understanding, vocabulary, grammar, and the frequency with which their children communicated. The review also revealed that parents were as effective at helping their children's communication development as therapists were. In fact, parents were more effective than therapists when working on improving the children's understanding of language and grammar.

4.2.2.7 Experimental Design of AT Studies

The level of evidence of AT studies are generally low (Rayes et al., 2019) due to the lack of randomization, power calculation and/or blinding, and failing to report performance at follow-ups post AT programme, compliance of children and parents, and methods of reinforcement. There is often a trade-off between the ideal study design and the practical implementation. An ideal AT intervention study should be an RCT, and the control can be active participants engaged in a different activity for a similar amount of time as in Mason (2017) who used art activities as a control condition in their evaluation of executive function training for deaf children. An inactive/passive control group essentially means that participants do nothing new in their daily routine.

To achieve the desirable effect size, it is important to estimate the sample size by performing apriori power calculation; and to reduce bias, blinding should be employed. Furthermore, reporting outcome measures and the approaches of training used can improve the quality of the study as it indicates coherence and relevance, while providing reinforcement and feedback to children and their families and assessing their compliance with training protocols enhance the outcomes of the intervention and improve overall quality of the study. Also, assessment of retention of improvements post intervention is important to be investigated as the evidence of maintained benefits post AT intervention or its absence is an indicator of the effectiveness of the intervention. Last but not least is the location where AT is conducted could influence the ecological validity of the study as home-based AT could be more effective than laboratory-based AT since it reflects everyday listening environment. (Henshaw & Ferguson, 2013; Rayes et al., 2019).

4.2.3 Rationale and Aims

There is a lack of higher quality research to assess the effectiveness of AT programmes for deaf children with CIs. Although the level of evidence is generally low it should not be interpreted as a lack of benefit of AT programmes. Many studies have found improvements in trained tasks (Rayes et al., 2019) but due to the study design it makes it difficult to interpret the findings. This research aimed to implement a high quality RCT to evaluate a multi-modal AT programme against an active, art control. Hence, the aim of this research was to develop an evidenced-based parent-led AT intervention programme and determine whether it can lead to improvements in speech and pitch perception in children with CIs. This aim can be broken down into the following objectives:

1. Develop a multi-modal training approach for school-age children designed to:
    a. improve pitch perception (trained task)
    b. improve phoneme discrimination (untrained task)
    c. improve speech-in-noise perception (untrained task)
2. Develop the training programme such that it can be readily implemented by parents and caregivers at home, to ensure this has been achieved monitor parents' compliance and engagement.
3. Follow the guidelines for assessing the efficacy of the multi-modal intervention to achieve a high quality of evidence
    a. Measure untrained skills to determine generalisation of effects (phoneme discrimination and speech-in-noise perception)
    b. Have two baseline sessions to evaluate learning effects of the materials
    c. Use a randomised control trial
    d. Have an active control (art-based training)
    e. Use a power calculation to determine sample size
    f. Measure retention of improved abilities (if improvement occurs)

We hypothesized that use of a multi-modal parent-led AT intervention used over a four-weeks period, would improve speech and pitch perception in children with CI compared to a control group conducting art activities.

4.3 METHODS

4.3.1 Estimation of Sample Size

An apriori sample size calculation was conducted for an ANOVA repeated measures analysis with a between-subject factor of group (multi-modal training and art-training) using G-Power software (Erdfelder, Faul, & Buchner, 1996). The test retest, standard deviation and effect sizes were taken from our previous study on the validation of the outcome measure, the A-CAPT. The hard A-CAPT list, was previously found to be a more discriminatory subset of the test, and so this was used to estimate the required sample size for this intervention study. The test and retest data were collected from 23 children (16 children with normal hearing and 7 CI users) and data were significantly correlated (0.8). Using the measure of effect size (Partial Eta squared (η2) was equal to 0.2) of the test and retest session for the hard list of the A-CAPT, the sample size was determined to be 16 subjects per group at significance (alpha) of .05 and 80% power.

4.3.2 Participants

Participants were recruited from multiple venues including the Jeddah Institute for Speech and Hearing (JISH), Language and Listening Stimulation Centre (LLSC), King Abdulaziz University Hospital (KAUH) and through social media channels. Unfortunately, the target sample size (32 subjects) was not met because recruitment was halted due to lockdown restrictions during the Coronavirus disease 19 (COVID-

19) outbreak. In total fourteen children with CIs participated whose caregivers consented to enrol and participate in the programme. No other co-morbidities affecting development were reported. Children were randomly assigned to either the multi-modal training or a control group. After random assignment, the two groups did not significantly differ on age and duration of CI use. This limitation in recruitment numbers means that only large effect sizes (η2 equal to 0.14 or more (Watson, 2019)) can be detected.

All participants were congenitally deaf, aged between 5 and 13 years, and had been using their implants for at least one year. Participant summary demographics can be seen in Table 4-1 and individual demographics can be seen in Appendix V. On average, the children assigned to the control group were slightly younger than the multi-modal training group (M = 8.29 years vs. M = 10.13 years) but these differences were not significant. There was no difference in implant experience in control and multi-modal training group (M = 5.40 vs. M = 5.69). All children attended mainstream schools, however two children within each group attended classes for children with special needs that were offered and managed within the mainstream schools. All participants were compensated for their participation in this study to cover transportation expenses. The test and training procedures were reviewed and approved by the University College London (UCL) ethical committee review board (11265/002).

Table 4-1: Demographics for participants (Ages and duration of CI use is reported in years)

	Age at Testing	Age at Implant	Duration of CI use	Number Bilateral	Number of Females

Intervention (N=7)	**Mean**	10.13	4.44	5.69	1	3
	SD	2.39	0.96	1.86		
Control (N=7)	**Mean**	8.3	3.04	5.40	1	5
	SD	3.22	1.11	2.57		

4.3.3 Assessments

4.3.3.1 Outcome Measures

There were three main measures that were used to assess outcome, a consonant discrimination test in quiet (A-CAPT), a word recognition in noise (Arabic LNT), and a complex piano tone pitch discrimination test. The A-CAPT and Arabic LNT were selected to assess generalization of learning from complex speech training tasks that mainly rely on top-down processing to distinct tasks namely consonant discrimination and speech-in-noise recognition. The complex piano tone pitch discrimination test was selected to assess trained task which was pitch discrimination.

4.3.3.2 Arabic Chear Auditory Perception Test (A-CAPT)

A-CAPT is a computer-based test that consists of two lists at two levels of difficulty, easy and hard. The A-CAPT was developed in Modern Standard Arabic language and was validated on children with normal hearing. The A-CAPT is an appropriate speech perception test for children as young as 5 years old. This test can reliably assess consonant discrimination ability and monitor changes over time or after an intervention. It is a phoneme discrimination test, which consists of monosyllabic words in form of CVC (consonant-vowel-consonant) or CVCC (consonant-vowel-consonant-consonant) that differ in either first or final phoneme. A-CAPT is a closed-set test that uses four response alternatives on each trial. This Arabic auditory perception test was developed based on the British English CAPT (Vickers et al., 2018), which was reported to provide valuable frequency-specific information about the audibility and discrimination of speech cues from the pattern of phoneme confusions produced after a child completed the test. The A-CAPT was delivered to the children in a form of a computer game and stimuli were presented over Sennheiser HD 650 headphones at 70 dB SPL. Scoring was calculated based on the number of correct responses out of total number of presentations which was 32.

4.3.3.3 Arabic Lexical Neighbourhood Test (LNT)

Arabic LNT (Alsari, 2015) is an open-set test, which assesses speech recognition skills in children with hearing impairment. The test is based on two main principles; first was to ensure that the words of the LNT are familiar to young children with their limited vocabularies and the second was to construct the LNT test based on the standards of the Neighbourhood Activation Model (NAM) (Luce & Pisoni, 1998). NAM recommends that words are organized into “similarity neighbourhoods” based on their frequency of occurrence and also the organization of the word in the mental lexicon which is based on “lexical density” i.e., acoustic–phonetic similarity of words within the lexical neighbourhood. The Arabic LNT (ALNT) (Alsari, 2015) consists of two lists (easy and hard) with 50 words each. The ALNT (Alsari, 2015) was developed in colloquial Najdi Arabic dialect and was validated on children with normal hearing children. The test was shown to be reliable even when administrated repeatedly over time. This test was presented in speech-shaped noise which was adaptively altered to determine the 50% speech reception threshold (SRT) as a speech-to-noise ratio (SNR). The SNR is a measure of the noise level in decibels relative to the speech. Stimuli were presented over Sennheiser HD 650 headphones at 70 dB SPL.

4.3.3.4 Complex Piano Tone Pitch Discrimination Test

This test was developed by Griffin (2016) to examine pitch perception in musical contexts in CI users and then it was used to assess the potential benefits of singing activities on children's hearing acuity and pitch perception (Welch et al., 2015). The test was a three-interval, three-alternate forced choice task, where one out of three stimuli was different. The stimuli were synthesised piano-tones comprised of three note chords and the target stimulus was different by one semitone. Six chord contrasts were assessed, three for a base note of C4 and three for a base note of G4. Stimuli were delivered via a computer programme and responses recorded on a laptop; the sounds were presented over a loudspeaker at a loud but comfortable level. A pass or fail score was calculated for each contrast, and the total number of contrasts was 30.

4.3.3.5 Parent and children questionnaire

All children and parents who took part in the multi-model training and control groups received a questionnaire to evaluate the training approaches and any perceived benefits. This was conducted after the trial had been completed to better understand the findings. The parents' questionnaire contained 6 closed-set questions and an open-ended question for free comments on the parents' experiences and views of the programme. The closed-set questions assessed parents' perception toward the training approach (play-based, parental-led, and dosage) and any perceived benefits (training materials) (Appendix VI). Scores for each question were out of 2 for the closed-set questions creating a maximum score of 10 for the entire questionnaire.

A simplified version of the questionnaire was given to the children to assess the training approach and engagement by asking them how enjoyable the activities were and whether they would participate again in a similar programme. They were also asked to select which of the three activities they found most entertaining (Appendix VII). Scoring this questionnaire was based on a binary response of whether they enjoyed the programme or not given a pass or fail score for each child.

4.3.4 Training Tasks and Procedure

As stated earlier the multi-modal training programme was developed based on recommendations from a systematic review on the effectiveness of AT programmes for children with CIs (Rayes et al., 2019). One of the recommendations of the review was to combine both top-down and bottom-up approaches to maximize the benefits of the intervention. The top-down training tasks that were selected for this training battery were language-enriched exercises (Alefbata.com) and communication and speech comprehension activities using a semi-structured dialogue tool (Diapix), while the bottom-up tasks involved discrimination of pitch and rhythm contrasts using musical keyboards (Figure 4-1). Tasks were organized in home-based series of short 10-minute interactive exercises.

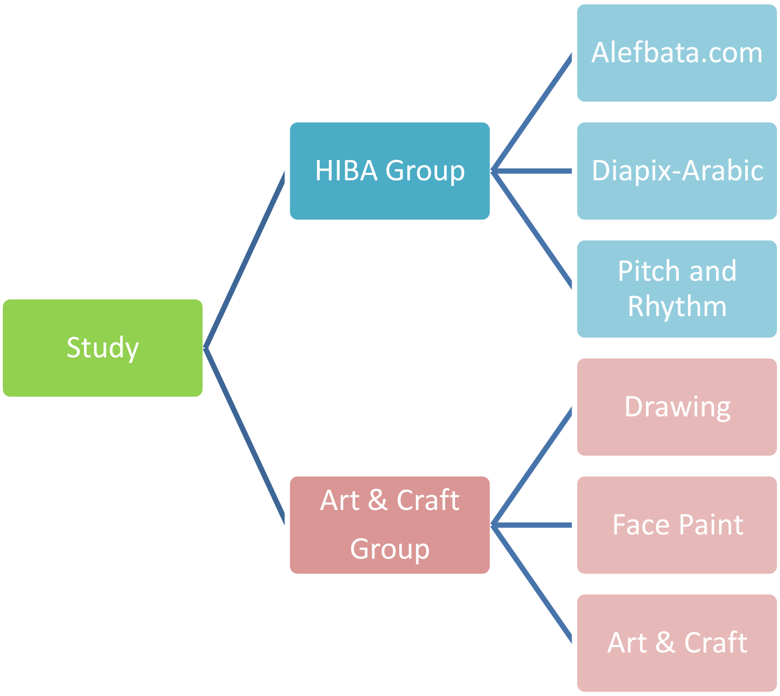


Figure 4-1: Diagram of the study design outlining the main activity areas for each group

4.3.4.1 Alfebata

The Alfebata ("Teaching Arabic Language to Children ," 2016) is an individualized language training application that relies mostly on a top-down approach. This language enriched training task was included in this programme because it promotes learning the Modern Standard Arabic language, which children are learning and exposed to in school and media. Learning the standard form of the Arabic language is important because it contribute to children's language competency. Three levels of language competency were established for this task to construct the individualized training scheme, such individualization was based on the children's language experience and age.

Level one, which was offered to 2 participants (5 and 7 years old), was consisted of exercises with low level of difficulty focusing on building up vocabulary through matching pictures to words, finding antonyms and reading and listening to very short stories and answer relevant questions. Level two, which was offered to the

10 year old participant, and consisted of exercises with a medium level of difficulty that involved building up vocabulary and phonemic awareness in addition to comprehension through participating in cross-words puzzle, constructing words from phoneme, matching pictures to words with similar (phonemes) e.g. sheep vs sleep, eliminating incorrect phenome from a word or eliminating incorrect words within a sentence in addition to reading and listening to medium-length stories and answer relevant questions. Level three, which was offered to 4 participants aged from 10-13 years, provided exercises of greater complexity aimed to improve phonemic awareness and syntax in addition to comprehension through different games that involved manipulation and construction of words and sentences and reading and listening to the highest level of stories that were available within the selected application.

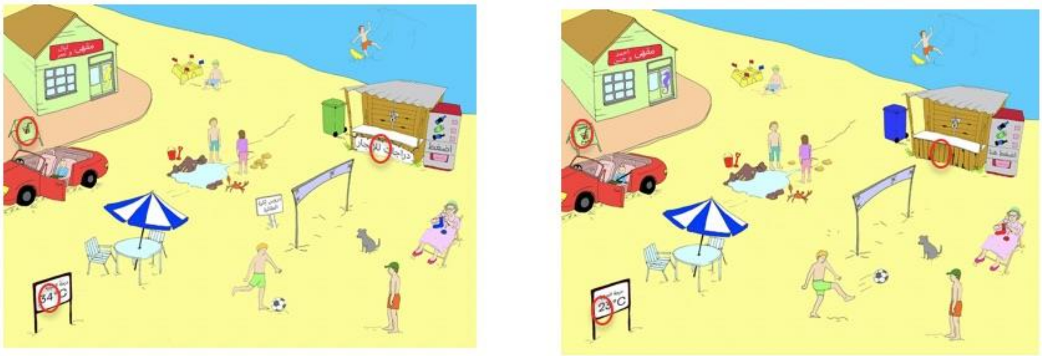
To determine children’s language experience, school grades and ages of participants and their duration of CI use were considered when preparing training materials. These were relatively good measures since children who were older and used their CI for longer duration and were exposed to aural language for longer period of time. This assumption led us to place one 10-year-old participant at a slightly lower level (level 2) than her peers as she was one academic year behind. This assumption was later confirmed when the participant scored lower than her peers in all the outcome measures that were collected at baseline.

Alefbata.com an online application that was selected to offer participants with enriched language exercises in Modern Standard Arabic. It offered interactive exercises in Modern Standard Arabic could improve children’s vocabulary and manipulation of the standard form of the language. The exercises, which were presented in the context of colourful computer games and children received feedback on each trial, consisted of matching pictures to words and finding antonyms as means to help learning new vocabulary, completing the missing letters in the beginning, middle, and last of the words, words puzzles, and restructuring words and sentences to help improving phonemic awareness and syntax, engaging in stories to improve reading and listening comprehension depending on the approach that was taken as both were available. Each child was provided with unique sign-in information and activities were organized and accessible in form of weekly assignments that they systematically worked through following the regime established by the test developers. Children's responses were recorded online within the application for the researcher to review and assess the child's engagement. Training was completed at the participants home with parental supervision using either computers or tablets with speakers recommended to be placed at approximately 0 degrees azimuth. Children seated themselves at a comfortable distance from the device and stimuli were presented at a comfortable loudness level.

4.3.4.2 Diapix, Arabic

This task was a communication game that utilizes a top-down training approach. It was based on the Diapix, UK (Baker and Hazan, 2011), which is a spot-the-difference picture game that is used for stimulating spontaneous speech interactions between two individuals. This task was selected because it can stimulate conversation in children's mother-tongue dialect that may help them learn new vocabulary, maintain their attention and enhance their speech comprehension through engaging in an interactive and game-based dialogue. In this task, participants are instructed to find the differences in the pictures without looking at the partner's copy. Although the pictures are designed to elicit certain words, it is not dialect dependent, which makes the Diapix an appropriate tool for practicing conversation in any dialect.

Controlling for dialect is generally difficult for many languages, however it is especially complex when it comes to the Arabic language due to the fact that Arabic is a diglossic language with a wide range of dialects that could differ widely across regions. To use the Diapix in HIBA, an Arabic version was developed, which used exactly the same pictures in the Diapix, UK with English writing translated into Arabic and few scenes were slightly modified to fit the Arabic culture. There were three main scenes, beach, farm, and street, and each of these scenes were modified to create four different versions, producing a total of 12 scenes, in addition to a park scene that was used only for practice. Figure 4-2 shows one copy of each scene in the Diapix, Arabic.



Beach 1A

Beach 1B

Farm 1A

Farm 1B

Street 1A

Street 1B

Figure 4-2: The graph shows version number 1 of the three different scenes of the Diapix, Arabic, beach, farm and street. Scenes A are different than scenes B. Participants were instructed to find and circle the 12 differences on the sheets. Three differences were shown by red circle in each scene.

4.3.4.3 Pitch and Rhythm Training

The pitch and rhythm music training utilized a bottom-up training approach. Children were provided with take-home electric keyboards, which were used for listening to different pairs of notes. They were expected to train their listening skills by learning to discriminate between pair of notes that were presented by their parents. For this training, only three octaves and one extra note at the high end of the keyboard were used. Parents were provided with list of pairs with increasing level of difficulty and were instructed to record their child's performance in the provided diary and note what they have been able achieve and any observation that they might have found interesting. Parents were also instructed that if the child could not manage to discriminate a pair of notes, they should record it in their diary and try it the following day, and to always try to end the training session with a task that the child can perform successfully. Task record sheets were also provided and were used together with the diary for evaluating overall engagement. This training protocol was provided by Advanced Bionics and had been previously developed for a paediatric music study, it was used previously in an AT study which reported positive outcomes (Yucel, Sennaroglu, & Belgin, 2009).

4.3.4.4 Multi-modal Training Folder

Each participant in the multi-modal training group was provided with a folder that organized the required tasks on weekly basis. Each week started with a task sheet which was planned for three days a week for four weeks. The tasks sheet listed the activities that the children needed to complete with a tick box to be ticked by parents when each activity was completed. Detailed instructions were attached following the tasks sheets; either the actual tasks as for the Diapix, Arabic and pitch discrimination or instruction on how to access the task as for the online exercises. For example, the first task sheet outlined the tasks for the first day in the first week of the training programme, participants needed to spend 10 minutes on the online application practicing the assigned language exercises,10 minutes on the daipix scene number 1from the beach collection, and spend 10 minutes practicing pitch discrimination using the provided list of pairs and musical keyboard.

4.3.5 Control Tasks and Procedure

Three main tasks were designed for participants in the control group. The first task was a drawing activity (Soloff Levy, 2016) the second was a face-paint activity (Multier & Ronzon, 2019), and third task was an art and craft activity (Tony, 2012). All three activities provided step-by-step instructions for parents and children.

The children who participated in the control group were also provided with folders that outlined their tasks. These tasks were organized in weekly tasks sheets, planned for three days a week for four weeks. The tasks sheet listed the activities that the children needed to complete with a tick box to be ticked by parents when each activity was completed. The art work was collected by the research after tasks were completed, and used along with the tasks sheets to assess the level of engagement.

4.3.6 Assessment Procedure

Children were assessed with the A-CAPT, Arabic LNT, and complex piano tone pitch discrimination test twice prior undergoing the training and once post training. The first assessments were to measure the baseline level for each child while the second measurements, which were conducted two weeks later, were performed to avoid learning effect that could occur from the second exposure to the test materials. The third assessment was conducted post intervention to evaluate children's performance after participating in the multi-modal training programme and to assess its efficacy on improving speech outcomes. Post training assessment was administered within a week following the completion of HIBA for the multi-modal training group or approximately four weeks following the completion of the pre-assessments for the control group (Figure 4-3).

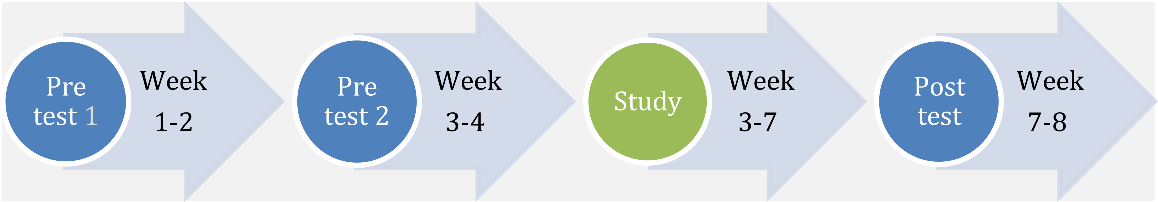


Figure 4-3: Illustration of the process of data collection at three different points of time.

Assessments were conducted in a quiet room at JISH, LLSC, or KAU. When transportation was reported to be a hurdle by families, assessments were given at the homes of children (3 participants in the multi-modal training group and 2 participants in the control groups). Following the pre-training assessments, those children assigned to the training group completed four weeks of HIBA while the children assigned to the control group completed a four weeks programme involved of art and crafts activities. Training was administered at participants' homes and the parents or caretakers were the leaders in this AT programme.

Prior to taking part in the study, all parents attended a session to learn how to conduct the assigned tasks and participated in a trial session to practice the steps and techniques needed to successfully complete each task. For example, when preparing for training with the DiaPix, familiarisation of the words within a given environment such as the beach was recommended. Another technique that was recommended to parents when training for pitch discrimination involved allowing the children to play with the keyboard and listen to the sound that it produced without following any instructions or guidance from the parent. In addition, continuous support whether in-person or online was offered by the researcher to answer any questions that parents may have during the study period. The children in the multi-modal training group completed 90 min of training per week for four weeks; this training dosage is similar to other AT training given to this age range (Ingvalson, Young, & Wong, 2014). The children in the control group were also requested to spend at least 90 minutes a week with their parents on the assigned art activities. Although ninety minutes of activities were scheduled for three days per week, allowing 30 minutes of activities per day, flexibility in the arrangement of training minutes was permitted to accommodate family daily routine, schedule or plans. Therefore, participants were allowed to choose which activity to perform first and to spread the recommend activates over the week if three days is too restrictive or challenging.

All assessments were conducted by the same researcher who was blind to the allocation groups at the first pre-training assessment but not the second. The participants were blind as to whether they were in the experimental or control arm of the study in the first and second pre-training assessment but not for the whole period of the study. In the post training sessions, children completed the A-CAPT, Arabic LNT, and the pitch perception test in a single session, just like pre-training assessments. We were keen to provide the trained and control children with equal exposure to the assessments and engagements in activities, and thus assessments and activates in both groups were planned to require the same amount of time and engagement.

Retention of benefits post intervention was planned to be conducted 4-week post training assessment. However, the study had to be terminated prior reaching this stage due to the COVID-19 outbreak and following lockdown.

4.3.7 Data Analysis

For statistical analysis, series of mixed ANOVA and t-tests were conducted to assess the effectiveness of HIBA on speech and pitch perception in children with CI. Alpha (∝) was set to be 0.05 significance and was reported based on two-tailed values

Mixed Analysis of Variance (ANOVA) models were conducted for each outcome measure with sessions (pre-training test 2, post-training test) as within subjects' factor and groups (multi-modal training and control) as between subjects' factor.

In addition, series of independent and paired t-tests were conducted to assess the effectiveness of HIBA on speech outcomes in children with CI. Independent t-tests compared the performance of children in the intervention and control groups while paired t-tests compared the performance of the children in the intervention prior and post intervention.

4.4 RESULTS

4.4.1 Comparing the Two Pre-intervention Sessions

There was a trend for a slight but non-significant (t-tests at the $p<0.05$ criterion level) increase in scores on all outcome measures from pre-training 1 to pre-training 2 (Figure 4-4). Even though the difference was not significant between the two runs in any of the measures, it was decided that it was more appropriate to use the 2nd pre-training test for comparison with the post-training session. There were outliers in three out of four assessments. In this case of test-retest runs, where the null hypothesis of no observed difference is the desired outcome, outliers can cause a significant difference and could be interpreted as a learning effect. Although outliers were present, their existence did not result in a significant difference. Accordingly, and due to the small sample size, outliers were not excluded. Table 4-2 contains the values and statistical findings for the analyses.

Table 4-2: Mean scores and SD for all outcome measures shown for the multi-modal training and the control groups in pre-training 1 and pre-training 2 sessions.

Outcome measure	Subset	Time	Multi-modal training (Mean (SD))	Control (Mean (SD))
A-CAPT (Scores out of 32)	Easy	Pre-Training 1	18.43(6.80)	18.86(7.03)
		Pre-Training 2	20.29(6.45	21(7.02)
	Hard	Pre-Training 1	13.43(13.43)	17.29(7.48)
		Pre-Training 2	16.29(16.29)	16.14(7.82)
ALNT (Scores as SRT)		Pre-Training 1	10.85(5.07)	6.55(4.97)
		Pre-Training 2	9.55 (5.70)	6.01(3.37)
Complex Piano Tone Pitch Discrimination Test (Scores out of 30)		Pre-Training 1	22.43(5.32)	17.00(3.42)
		Pre-Training 2	19.71(6.02)	16.71(4.03)

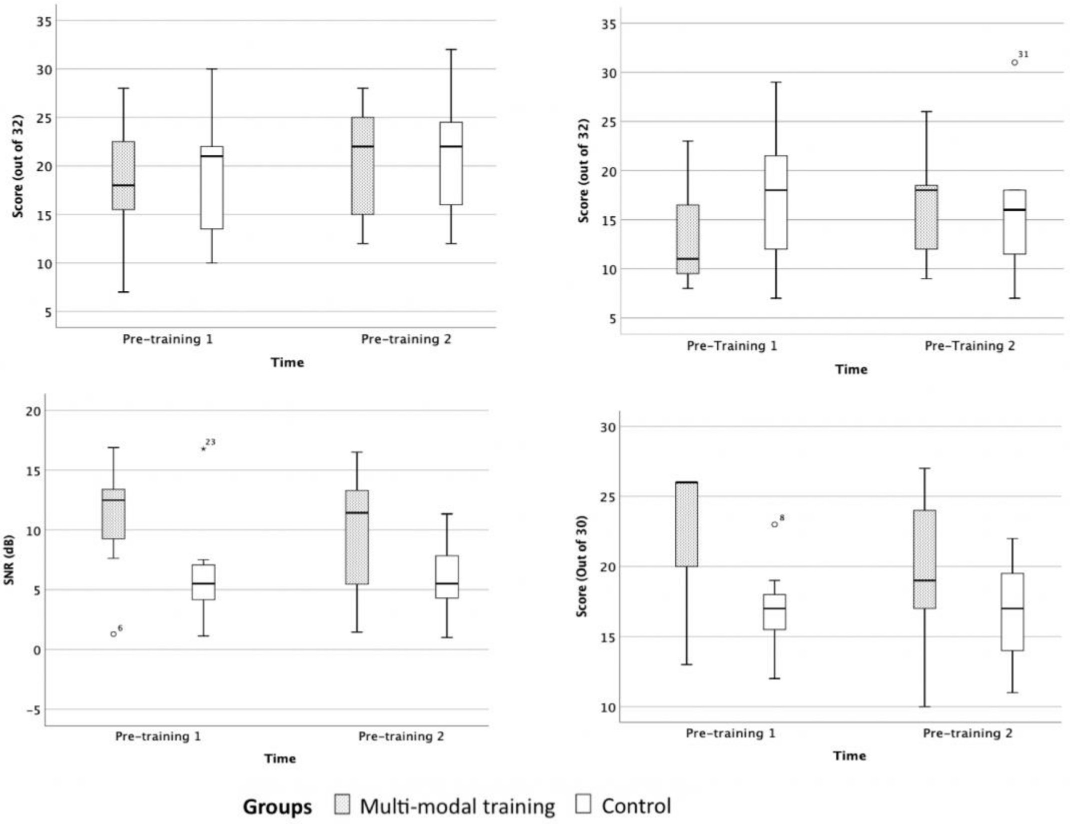


Figure 4-4: The shaded boxes in the box plots represent the multi-modal training group and the white boxes represent the control group. The x axis shows the pre-training time points. The y axis for the top two panels, shows the raw score (out of 32) for the A-CAPT. Easy lists were top left and hard lists top right. In the bottom left panel, the y axis represents the SNR at SRT for the ALNT test and the y axis in the bottom right panel represents the raw score (out of 30) of the pitch perception test. The box represents the interquartile range (25th to 75th percentiles), black horizontal line the median, Whiskers above and below the box indicate the 10th and 90th percentiles. Although only three out of four runs had at least one outlier (outliers shown in small circles and extreme values are marked with a star), their occurrence did not result in a significant difference between runs.

4.4.2 Comparison of Assessing Effectiveness of Intervention

4.4.2.1 Speech Discrimination Test (A-CAPT)

A 3-way mixed ANOVA was conducted with the factors group (multi-modal training, control), session (pre-training, post-training) and list (easy, hard) with consonant discrimination score (out of 32) as the dependent variable. There was no main effect of group, F $_{(1,12)}$ = 0.31 p = 0.59, η2 = 0.25, ß = 0.08, nor main effect of session, F $_{(1, 12)}$ = 2.23, p = 0.16, η2 = 0.16, ß = 0.28. However, there was a main effect of list, F $_{(1,12)}$ = 28.90 p < 0.01, η2 = 0.70, ß = 0.99, as participants' scores were higher in the easy list compared to the hard list. There was also a significant interaction bwtween group and session, F $_{(1,12)}$ = 15.68, p = 0.02, η2 = 0.57, ß = 0.95, while both groups had similar scores at the pre-test session, scores for the multi-modal training group were higher at post-training session. There was no significant interaction between list and group, F $_{(1,12)}$ = 15.68, p = 0.14, η2= 0.17, ß = 0.31, between list and session, F $_{(1,12)}$ = 0.09, p = 0.76, η2= 0.08, ß = 0.06, nor between list, group, and session, F $_{(1,12)}$ = 0.09, p = 0.18, η2 = 0.15, ß = 0.26.

Further analysis was conducted to understand the significant interaction between group and session. The mean score for the multi-okmodal training group was 18.29 in pre-intervention assessment and 21.29 in post-intervention assessment. The mean score for the control group was 18.57 in pre-intervention assessment and 17.23 in post-intervention assessment. A post-hoc analysis was conducted to evaluate the interaction between group and session and revealed that there no significant change between the performance of the multi-modal training and control groups in pre-, F $_{(1,26)}$ = 0.01, p = 0.92, η2= 0.0, ß = 0.05, and post-, F $_{(1, 26)}$ = 2.48, p = 0.18, η2 = 0.09, ß = 0.33, intervention. However, when the change in performance (delta) post-intervention was assessed for both groups using a one-way ANOVA, results revealed the change in the multi-modal training group was significantly higher than that in the control group, F $_{(1, 26)}$ = 14.82, p = 0.001.

Since phoneme discrimination training was not included in the multi-modal training battery, improvement in this subset of the test suggested that training might have generalized to this untrained task, namely phoneme discrimination.

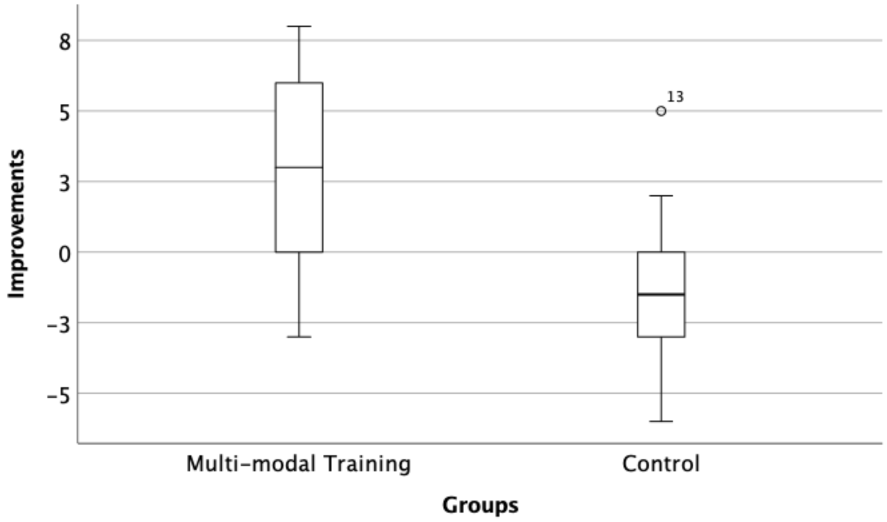


Figure 4-5: The graph illustrates the change in performance post intervention for both groups (multi-modal training and control). The y axis shows the improvements (calculated by subtracting scores post-intervention from score pre-intervention) and the x axis shows the groups. An outlier in the control group (shown in circle) was observed, but the case was not removed from the analysis due to the small sample size.

4.4.2.2 Speech-in-Noise Test (ALNT)

A 2-factor mixed ANOVA was conducted with factors group (multi-modal training and control) and session (pre-training and post-training) with the score of SNR at the SRT as the dependent variable. The results revealed no main effect of group on score of SNR, F $_{(1, 12)}$ = 1.02, p = 0.33, η2 = 0.08, ß = 0.15, nor main effect of session on SNR, F $(_{1,\ 12})$ = 0.51, p = 0.49, η2= 0.04, ß = 0.10. There was also no significant interaction between group and session, F $(_{1,\ 12})$ = 1.27, p = 0.28, η2= 0.10, ß = 0.18 (figure 4-6). Although there was a trend for a decrease (improvement) in SRT for the multi-modal training group (9.67 and 6.94 dB SPL for pre and post respectively) it did not reach statistical significance. This could be possibly due to the outlier skewing the SNR positively and reducing the effect size of the intervention. There was also no significant change between pre (5.67 dB SPL) and post (6.9 dB SPL) sessions for the control group and neither was a positive trend for improvement observed. The lack of statistical significance for improvements in speech-in-noise scores suggests that there was no generalization to speech-in-nose perception. Although outliers occurred in the post-training control group with no apparent trend possibly due to random error, their presence had little effect on significance.

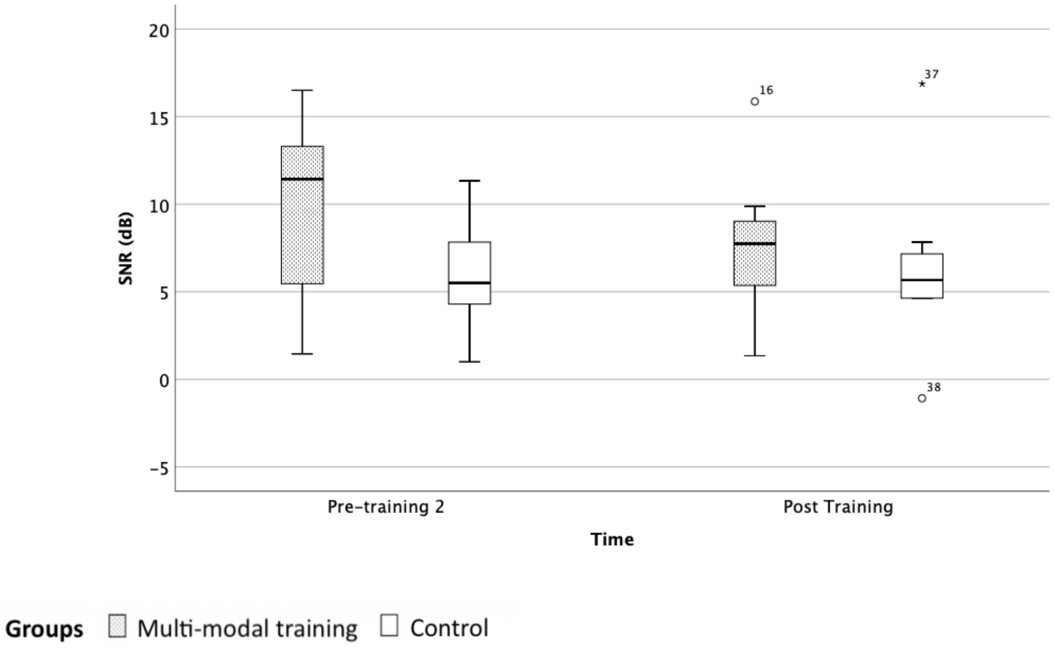


Figure 4-6: Box plot representation of the SNR at the SRT in pre and post intervention and over time in multi-modal training and control groups. The Y axis represents the SNR at SRT (lower scores mean better performance) and the X axis represents two time points (pre and

post intervention). Outliers (marked in small circle or star) were observed at the post-training test in the multi-modal training and control groups

4.4.2.3 Pitch Perception Assessment

A 2-factor mixed ANOVA (factors, group and session) was conducted for pitch discrimination scores (out of 30) as the dependent variable. There was no main effect of session on pitch perception, F (1, 12) = 2.43, p = 0.15, η2 = 0.17, ß = 0.30, nor a significance effect of group, F (1, 12) = 3.68, p = 0.08, η2 = 0.24, ß = 0.42 (figure 4-7). There was also no significant interaction between group and session, F (1, 12) = 1.55, p = 0.24, η2 = 0.12, ß = 0.21.

Figure 4-7: Box plot representation of pitch discrimination scores over time for both the intervention and control groups. The Y axis represents the raw score (out of 30) and the X axis represents two time points (pre and post intervention). Outliers (shown in circles) were observed in the multi-modal training group in the post-training session.

4.4.3 Power Calculation and Effect Size

Post-hoc power calculation and effect size were calculated for the A-CAPT since it is the only measure that showed statistically significant improvements in performance post the intervention. The effect size (partial eta squared) was determined to be 0.36. The power calculations for the one-way ANOVA, comparing the change in performance of the participants in the multi-modal training group to those in the control group post intervention, was calculated using GPower (Erdfelder et al., 1996) and was equal to 0.73. at 0.05 alpha.

4.4.4 Parents' and Children's Questionnaire

Average scores for parents' questionnaire in the multi-modal training group was 9.4 out of 10 (Appendix VIII). The questionnaire evaluated three main aspects namely, training approach, entertainment, and benefit of AT. All parents agreed that the training programme was entertaining in the current format for the children and families and almost all agreed that the training approach was beneficial when they were asked about their opinions on whether the training was beneficial or not. Only one of the seven parents thought that the activity was not appropriate for her son's age (13 years old).

Parents responded to the opened ended question indicating that they continued after trial has ended to incorporate different activities that could improve spoken language into their everyday family routine. Three parents noted that the training helped them to realize the importance of having an open discussion with their children (as when they used Diapix) rather than teaching them a particular skill. One said that before the training that she had thought that her son was considerably advanced in terms of spoken language but after going through the dialogue activity in Diapix, she realized that she needs to work on his conversational skills. Another mother was extremely surprised by how her son was able to discriminate between the pairs of the musical notes and this boosted her confidence and encouraged her to continue working with him to train his listening skills.

The children's questionnaire was brief and consisted of only two questions with a 'yes' or 'no' answer for each. All children responded with 'yes' to say that they found the activities enjoyable and also that they were eager to participate in future programmes. Although we attempted to collect data for the questionnaire from parents and children in the control group, they did not provide their responses.

4.4.5 Parental Involvement

Level of engagement of parents were assessed based their answers to the closed-ended and open-ended questions in the questionnaire reflecting their willingness to lead their child's learning experiences. All parents were interested to receive more materials particularly for the Diapix activity. Although we attempted to collect data for the questionnaire from parents in the control group, they did not respond.

Parental engagement was also assessed based on interaction with the researcher. In the multi-modal training group, parents were actively engaged with the researcher asking questions to validate their training techniques and voluntarily shared their progress via an online messaging platform. In contrast, parents in the control group rarely contacted the researcher and instead the researcher consistently reached out to them to ensure that they complete the assigned tasks.

4.5 DISCUSSION

The HIBA is a multi-modal AT intervention designed for use with children using CIs. The intention was to develop the children's hearing abilities and in turn their speech perception. The findings supported the prediction that the training would lead to perceptual improvements. For speech discrimination abilities, there was a significant improvement for the A-CAPT (untrained task). Despite that SNR at the SRT in the multi-modal training group decreased (improved), there was no evidence for generalization of learning to speech-in-noise perception (untrained task); the change in score did not reach statistical significance. Likewise, there was no significant improvement for pitch perception (trained task) despite the improved average scores for the multi-modal training group.

4.5.1 Training Approach

The defining principles of the training approach were that it was parent-led, multi-activity and interactive between parent and child. The training programme allowed the parents and children to have some flexibility in how they progressed through the programme allowing for an individualized approach. The intervention can be modified according to the subjects' needs (ability and age), environments (home dialects and settings) and daily routine since it was implemented at home with parents and thus it can be conducted at the participants' convenience. The programme for the control group was as flexible as for the intervention group and the tasks can be amended according to the participants' ability, age, environment, and daily routine.

We selected a mixture of approaches that were age appropriate and covered a range of listening skills that engaged both top-down and bottom-up processing. One of the components used in the training was previously assessed by different research team (Yucel et al., 2009) and involved pitch discrimination tasks. The other two training tasks involved communication and problem-solving tasks using Diapix, Arabic and language exercises using Alefbata application. This complex training approach included tasks on auditory discrimination, speech recognition and comprehension, attention, and communication skills. This dynamic training approach was also used to assess generalization of learning to phoneme discrimination and speech-in-noise perception

4.5.2 Training Tasks

Children in the multi-modal training group were trained on pitch discrimination that utilize bottom-up processing using electronic piano keyboards. Pitch perception skills were assessed before and after training in both the multi-modal training and control groups and no significant improvements were observed. This particular training programme had been previously tested (Yucel et al., 2009) on younger children in Turkey where they showed a significant improvement in performance in trained task (pitch perception) but did not see a benefit for the untrained task (speech perception). Eighteen children participated in their study; nine children who were newly implanted were assigned to the training group, while the others who were assigned to the control group were participating in a different study that examined sound quality perception, speech understanding, speech production, and communication mode. There were differences between their participants' demographic information that could have influenced the observed significant change in pitch perception post their intervention. The ages of the participants in the Yucel et al. (2009) study were significantly younger than those who participated in our study and their duration of CI use were much smaller (participants were newly implanted).

Another factor that may have influenced their outcomes was the dose of the intervention, which was much larger (two years) than the dose we implemented (four weeks). Although there is flexibility when selecting the dose of AT intervention (Tye-Murray, 2019) and benefits in trained tasks were observed in AT intervention regardless to the duration of training (ranging from 4 weeks to 2 years) (Rayes et al., 2019), longer exposure to training may enhance learning experience.

The children in the multi-modal training group were also trained on speech perception using complex cooperative communication tasks and language exercises that utilized top-down processing. Combining both top-down and bottom-up approaches was in line with previous recommendations for combining a mixture of training activities to maximize benefits and promote generalization of learning. For example, when non-auditory tasks (visual-spatial task) was trained, improvements in performance on the auditory domain was observed, suggesting that merely arousal and maintained attention could improve listening skills. (Amitay, Irwin, & Moore, 2006; Rayes et al., 2019).

Children who participated in the multi-modal training study arm showed an improved average SNR at SRT scores post intervention but the changes did not reach statistical significance. Children in the multi-modal training group had significantly poorer speech-in-noise perception at baseline than participants in the control group. The gap between the two groups diminished following the intervention period suggesting an improvement in speech-in-noise perception for the multi-modal training group (Figure 4-6).

Welch et al. (2015) also assessed speech perception in noise in their singing training study for children with hearing devices and, similar to our findings, the authors noted that there was improvement in speech-in-noise skills but was not statistically significant and concluded that no transfer of benefits from trained tasks to speech-in-noise perception. Mishra et al. (2015) also trained speech-in-noise perception using a digit-in-noise training for children with CIs and reported improved performance only on trained tasks (numbers in white noise and numbers in speech-shaped noise), while no significant improvements were generalized to untrained tasks (digit triplets in speech shaped noise). The recurrent lack of generalization of learning to speech-in-noise perception in the previous studies may suggest that it is a skill that requires on-task training.

Overall, the children in the trained group in this experiment were older and were implanted at older age than children in the control group. Since brain plasticity is most robust at younger ages, such discrepancy may have masked benefits of pitch discrimination in HIBA. For example, similar AT studies have shown benefits post intervention in pitch perception (Yucel et al., 2009 &Welch et al., 2015) task but as mentioned above the ages of the participants in both studies were younger than those who participated in HIBA.

4.5.3 Parental Engagement

4.5.3.1 Parents in the Multi-modal Training Group

One of the positive aspects that we observed in this study was the strong involvement of the parents in the in the multi-modal training group. The data collected through the questionnaire illustrated the willingness of parents to lead their child’s learning experience. Parents were more engaged with the researcher than the parents in the control group. The extent of parental involvement is likely to have influenced the outcomes in the study (Roberts & Kaiser, 2017). The enthusiasm of the parents in the multi-modal training group was clear and they were very keen to continue after the study had finished. They kept to intentionally incorporate different activities that aim to improve their children's spoken language in their daily routine, they also were happy to receive new training materials, and they were interested to receive guidance and support in form of verbal information or future participating in research studies. In agreement with our findings, Zaidman-Zait and Young (2008) explored parental involvement in habilitation post cochlear implantation and found that mothers believed that they played an important role in their child's intervention. They were keen to establish a healthy parent-child relationship by engaging in entertaining interactions with their children. These findings came from a similar cohort (CI users) to our study and provide an insight into the perception of parental involvement, however they were based on only two cases and children were younger (2-3 year-old) than those who participated in this study.

Parental involvement in the habilitation process of children with CIs was also investigated with a closer cohort of participants to our study (Bruin & Ohna, 2015). Fourteen parents (10 mothers and 4 fathers) of children with CIs (age between 3 to 11 years) shared their experiences after the CI surgery. Similar to our findings, all parents described deliberately incorporating various activities directed towards spoken language skills into family life, because they felt responsible for their child's future outcomes. Furthermore, parents expressed appreciation for any information that they gain because they felt acquiring knowledge guided them to support their children. Similarly, (Erbasi, Scarinci, Hickson, & Ching, 2018) reported that all of the 17 parents of children with hearing loss (ages between 6-9 years old) who participated in the study were feeling responsible for supporting their children to maximize their potential. This attitude was common amongst all parents regardless to their education level, cultural background, mode of communication, and type of hearing device (hearing aids or CIs). In addition, they reported that parents were optimistic and had high expectations for their children and believed their children would not achieve the highest outcomes by merely attending therapy sessions and attending school. Thus, they were actively involved in their children's habilitation journey.

4.5.3.2 Parents in the Control Group

In contrast to the parents in the multi-modal training group, parents in the control group rarely contacted the researcher with questions or clarifications, instead the researcher consistently reached out to them to ensure that they complete the assigned tasks. In addition, we attempted to collect data for the questionnaire from parents and children in the control group but they did not provide their responses. Such lack of interested maybe because the art tasks were not as challenging as the AT tasks not directly related to auditory and speech perception, and being aware of their placement in the control group might demotivated them. It is important to note that prior the training took place, the researcher met with the parents in the control group just like those in the multi-modal training group and emphasized on the importance of engaging with the children when working on art tasks. Tourangeau, Rips, & Rasinski, (2000) noted that people may avoid providing their responses to questionnaires maybe due to reluctance in disclosing their opinions or having doubts about the legitimacy or importance of the tasks that were assigned to. Another reason could be timing issues as the questionnaire were sent after the study was conducted and during the outbreak of COVID-19.

4.5.3.3 Benefits of Parental Engagement

The inconsistency between the parents' engagement level of the participants in multi-modal training group compared to the controls may explain the discrepancy between the performance of subjects within the two groups where children in the multi-modal training group showed various degrees of improvement while children in the control group showed no improvement and in some cases a decline in their performance.

Allowing parents to have an active role in their children care, have the potential to alter the way rehabilitation is delivered to children with hearing loss. The role of the clinicians may shift from providing therapy in clinic to instead performing continuous assessments, developing individualized intervention plans, and training parents or caregivers to deliver those plans to their children. After all, the contact hours of clinicians with children is limited while parents are continuously with the children. Overall, studies (Roberts & Kaiser, 2017) have shown that parents' engagement is a key factor for the development of speech and language abilities in children with speech delay. This personalized approach of care for the children with CIs and their families goes along with the new vision of care services. The National Health Services (NHS) in the UK 10-year plan is an example of an initiative that aims to empower people by transforming their experience of health care as they will be actively engaged in accessing, managing and contributing to their health care services using digital tools and technology (NHS, 2019).

4.5.4 Quality of the Study

The study at hand used a pseudorandomized allocation of subjects who were partially blinded to the groups' allocation, included active control group, reported the power calculation value for the measure that showed statistically significant improvement, and reported outcome measure selection. AT specific measures were also considered as the outcome measures selection were applicable to the aims of the study, training feedback was provided for all aspect of tasks, ecological validity was excellent since the AT was conducted at the participants home and with their parents which better represents normal listening environment, and compliance with training protocols was assessed. However, retention of improvement after the intervention was planned to be assessed but not conducted due to uncontrollable circumstances (COVID-19) that led to the earlier termination of the study.

The quality of evidence of this study was calculated following the guidelines presented in our published systematic review (Rayes et. al, 2019). The study design was graded based on a sum of scores that was given to each category within the general scientific measures and AT-specific measures (Table 4-3). The quality of the study would have been moderate (score 15 out 20) had we been able to recruit the targeted sample size (32); unfortunately recruiting fourteen subjects made the study underpowered.

Table 4-3: The sum of scores (score 16 out 20) that was given to each category within the general scientific measures and AT-specific measures. You need to explain the numbers.

Scientific Study Validity Criteria					**AT-Specific Study Validity Criteria**					**Study Quality Score**
Randomization	Control Group	Power Calculation	Blinding	Outcome Measure Reporting	Outcome Measure Selection	Training Feedback	Ecological Validity	Reporting of compliance	Follow up	
1	2	2	0	2	2	2	2	2	0	15

4.5.5 Study Bias and Limitation

The sample size in this study was small, such limitation may have masked training gains in some measures and alternatively inflated gains in others. It is possible that a larger sample will reveal that training has little effect and many of the gains can be attributed to CI use. However, such speculation could be untrue in this study because the children who participated in this intervention were older and have been using CIs for longer duration.

Another limitation that was observed in this study was the lack of blinding. Even though the subjects were blinded to whether they were in the multi-modal training group or control group in the first and second pre-training assessment, we could not maintain blinding throughout the whole period of the study as the materials utilized were clearly either to enhance auditory and speech perception or not. In addition, it was impossible to blind the observer as the data were collected by only one researcher. To minimize bias, training was administered by the parents but not by the researcher. As a result, children in the multi-modal training and the control groups received the same time of interaction with the researcher who administered assessments. Therefore, the duration of the assessments were about 45 minutes per child whether in the control or the trained group, indicating that improvements in the trained group. cannot be attributed to increased familiarity with the assessments or the researcher. Finally, although improvement was measured in the intervention group, we do not know which aspect of the training led to such improvement and whether parental interaction, arousal, or motivation were the key factor that influenced such gain.

4.6 CONCLUSION

HIBA, the multi-modal AT for children with CIs, has the potential to improve auditory perception. Generalisation of benefits was observed for speech cue discrimination post-intervention in the multi-modal training group but not in the art group. Our findings suggest that children with CIs and their parents may benefit from regular and sustained access to age-appropriate AT materials and activities. Findings also indicate that empowering families and giving them an active role in their children’s care plan is applaudable as parents were compliant, fully engaged and able to implement home-based intervention when provided with clear tasks and plans. Parents were ready to take control of their children’s progress and actively shape their future. The study followed the published guidelines to achieve a higher level of quality of evidence and the quality of the study reached the highest level despite the limitations that was caused by the COVID-19 pandemic (e.g., reduced recruitment rate and the lack of a retention assessment phase).

CHAPTER 5: GENERAL DISCUSSION

5.1 OVERVIEW AND MOTIVATION

(Re)habilitation post implantation is a key factor that could significantly improve speech outcomes in children with CIs (Kennedy et al., 2006; Kral, 2013; Moeller, 2000; Yoshinaga-Itano et al., 1998). However, access to (re)habilitation services can be challenging for children and it can be dependent on the region and country in which they live. Saudi Arabia is an example of a country where it is difficult to access (re)habilitation services for many reasons such as children being diagnosed with hearing loss at significantly older ages delaying their initial hearing aid fittings and their enrolment in early intervention services. In addition, children who resided in in city areas had better opportunities to be fitted with hearing aids and enrolled into early intervention services than those who lived in rural areas; reduced awareness and ineffective methods for disseminating information also contributed to lack of access to such services (Alyami et al., 2016; Milaat et al., 2001). Scarcity of such essential services in Saudi Arabia was the main motivation for conducting this research. The aim was to determine whether HIBA, a multi-modal, parent-led training intervention can improve auditory and speech outcomes in Arabic-speaking children with CIs in Saudi Arabia.

An evidence-based approach was taken to develop HIBA. First, following the guidelines available in the review of the effectiveness of computer-based AT in adults with CI (Henshaw & Ferguson, 2013), the outcomes of AT in children with CI were investigated through conducting a systematic review (Rayes et al., 2019) (see chapter 2 for details). The review concluded that AT was beneficial for children with CIs based on reported improvements observed for on-trained tasks in all of the reviewed studies

(9 studies) regardless of the duration (ranging from 4 weeks to 2 years) or type of the approach (bottom-up or top-down). In addition, generalization of training or transfer of benefits to other learning domains (6 out of 9 studies) was observed in all the studies that assessed generalization. For example, Kronenberger et al. (2011) observed that the training of working memory skills led to improved language processing skills; the Good et al., (2017) study revealed that music training led to improved emotional speech prosody perception. Similarly, retention of benefits post AT was assessed in only 3 studies and it was revealed that improvements were sustained for a period ranging from 2 weeks up to 2 months post AT intervention.

Even though the quality of evidence was deemed to be low across the reviewed studies, having consistent positive outcomes in the three key measures that were used to assess the benefits of AT namely, improvement in trained tasks, transfer of benefits to untrained tasks, and retention of benefits post intervention, were encouraging and formed the impetus for the research in this thesis. Findings of this systematic review guided the development of HIBA (chapter 4) and thus measures such as randomization power calculation and active controls were utilized. In addition, to reporting outcome measures and the approaches of training and providing reinforcement and feedback to children and their families and assessing their compliance with training protocols was carefully incorporated.

The A-CAPT (chapter 3) was developed and validated to ensure that there was an appropriate outcome measure to assess the planned intervention. A-CAPT assessed consonant perception in children and its development fills a gap because of the sparsity of published tests validated with Arabic speaking children in Saudi Arabia. Consonant discrimination was considered to be important because it provides frequency-specific information on discrimination abilities that can help researchers and clinicians understand if and how perception is changing. Therefore, a closed-set phoneme discrimination test was developed based on the British English CAPT (Vickers et al., 2018).

5.2 THEORIES OF LANGUAGE ACQUISITION

The Nativist theory of language acquisition proposed that humans have a biological predisposition for learning languages and only a minimal input is needed for language learning. This theory minimizes the importance of the role of linguistic input and the language environment in language acquisition as it suggests that language acquisition can be acquired without the role of imitation or observation. Chomsky in (1959) claimed that children are born with a blue print of language or built-in language acquisition device (LAD) that makes mastering rules and grammar of any language is easily accomplished with minimal exposure to language. In contrast to the Nativist theory, the behaviourist theory claims that language acquisition is just like any other behaviour that can be observed and learnt through recurrent exposure. Behaviorist theory (Skinner, 1985) proposed that new-borns' brains are tabula rasa, without any built-in mental abilities for language acquisition. According to this theory, children learn the language through exposure to an input which elicits a response that is conditioned through reinforcement and rewards (Mohamad Nor & Rashid, 2018).

Combining both the nativist and the behaviourist theories resulted in the interaction theory, which HIBA was developed in accordance to. The interaction theory proposes that both biological and environmental factors are required for acquiring language. Interactionists believe that regular interaction between children and their natural environment is the optimal method for language acquisition, and parents-children interaction is crucial in shaping the learning process and maintaining children's attention (Chapman, 2000; Matychuk, 2005; Piper, 1998). Vygotsky (1978) believed that enriched environment and recurrent exposure to conversations with adults not necessary parents, enhance language acquisition via collaborative dialogue, where adults could model behaviours and provide verbal instructions, reinforcement and feedback. Behaviour change technique (BCT) is a method that fall under the interaction theory and used in behaviour change interventions for modifying behaviours such as eating habits or communication responses. BCTs were introduced to speech and language therapists and were encouraged to apply them in practice (Rees, Wood, & Cavin, 2016).

Training tasks in HIBA were designed with the interactionist approach in mind. The training empowered parents and encouraged them to assume their role in developing their children's language; it was implemented in children's most familiar settings (at home) with variety of activities that exposed participants to music, dialogue and semi-formal instructions. This multi-modal training paradigm aimed to provide children with enriched environment that promote language learning.

5.3 PARENTAL ROLE POST CI

Parental involvement in the post-implant auditory rehabilitation process is a key predictor of CI outcomes in children (Moeller, 2000). Parental involvement can be characterized by ensuring that the child regularly uses the CI and that the CI is functioning properly, providing auditory stimulation, consistently speaking to/with the child and engaging him/her in conversation, providing a language-rich environment, stimulating speech production of the child, interaction with the child's teachers or therapists, participating in the development and implementation of the intervention program, and attending therapy sessions. Such tasks could be perceived by some parents as a burden that is difficult to handle. Thus, it might be easier and less stressful for those parents to try to use tasks like the DiaPix which stimulate conversation and dialogue in a relaxed game-like environment rather than adhering to a more formal AT program that requires greater time commitment, group interaction, or a device (computer or tablet).

5.4 TRAINED TASKS IN HIBA

Pitch perception was assessed as a trained task in HIBA using a complex piano tone pitch discrimination test. Children participated in music training that involved pitch and rhythm discrimination tasks; participants were provided with take-home electric keyboards, which were used for listening to different pairs of notes. Although overall the change in performance post intervention did not reach statistical significance, between group assessment showed a trend towards improvement in participants in the multi-modal training group but not in the control group.

The music training programme in HIBA was implemented previously by Yucel et al. (2009). However, they conducted the intervention on younger children and continued the programme for two years. By the end of the second-year their music-training group had developed more rapidly than their untrained control group in all aspects of musical skills including pitch perception. Welch et al. (2016) also assessed pitch perception in their singing and vocal exploration study; pitch perception significantly improved over time (20 weeks) for children, particularly for those with hearing loss (less so for normal-hearing children). Duration of intervention may have influenced the findings on pitch perception for children in this study. Yucel et al. (2009) and Welch et al. (2016) conducted their studies for much longer durations compared to HIBA which was implemented for only four weeks. Since there was a trend toward improvement in pitch perception in HIBA, it would be interesting to re-conduct the study for longer duration, and assess if pitch training would lead to significantly improved pitch perception. Another factor that may have an effect on the results was age of participants. Participants in Yucel et al. (2009) and Welch et al. (2016) were considerably younger than the participants in HIBA. Neuroplasticity is at its greatest at younger ages and it lessens as listeners get older. Could neuroplasticity and age suppress the expected improvement in pitch perception as seen in previous studies?

The lower than expected sample size also could have masked the expected gain from the pitch training programme as fewer participants were recruited for this experiment which reduced the overall power of the study. Welch et al. (2016) included twenty-nine children, but twelve of which had hearing impairments and only six of which used CI while the other seventeen children had normal hearing; their sample size was heterogenous and not exclusive to children with CI. Yucel et al. (2009) recruited eighteen children with profound hearing loss who used CI, nine of which was assigned to the training group and nine were assigned to the control group. A stronger effect of HIBA intervention might have been detected if the study were sufficiently powered and the needed sample size determined by the apriori sample calculation was achieved.

5.5 UNTRAINED TASKS (GENERALIZATION) IN HIBA

Generalization of benefits to untrained tasks, phoneme discrimination and speech-in-noise perception, were assessed with two measures, the A-CAPT and ALNT. Analysis of the A-CAPT revealed a significant improvement in children's performance in the intervention group but not in the control group. Since exercises that directly train phoneme discrimination were not included in the training battery, this finding indicated that the benefits of the multi-modal training were generalized to phoneme perception.

It was unclear which component or combination of components led to the observed improvement due to the design of HIBA. Previous work by Roman et al. (2016) showed that training of identification and discrimination of non-speech stimuli such as enviromental sounds and music have led to transfer of learning to phonetic discrimination skills. However other studies did not observe generalization of learning from music training to speech perception (Welch et al., 2015; Yucel et al., 2009). Yucel et al. (2009) observed a transfer of learning in one of the speech measures, i.e. speech recognition through an open-set speech test but not the other that assessed speech detection, discrimination, and comprehension. Welch et al. (2015) did not report any transfer of learning to speech perception; the authors noted that focused singing training with children with hearing loss was not possible as participants were a heterogeneous mix of CI users, hearing aids users, and normal hearing children.

The current evidence may indicate that music training and specifically training of pitch perception may be associated with improved phoneme perception, however interactive communication and language training may also had an influence on the observed improvements in phoneme discrimination skills post HIBA intervention.

There was a trend for improvement in speech-in-noise perception (ALNT) for the multi-modal training group compared to the control group, however results indicated that the change did not reach statistical significance ruling out evidence for generalization of learning to this untrained task. Mishra et al. (2015) investigated generalization in speech-in-noise training. The group reported a near transfer as learning effects were established and generalized to similar but untrained conditions (number recognition in white noise), however no significant improvement was observed in untrained tasks (digit triplets in speech shaped noise). These findings could suggest that the training of speech-in-noise skills should be considered when developing an AT intervention that is intended to improve such skills. Again, the current study was unempowered and a stronger effect of HIBA intervention might have been detected if a larger sample size was utilized.

5.6 ACHIEVEMENTS AND IMPACT

5.6.1 Milestone I: Systematic Review

This systematic review was the first review to critically appraise the literature of AT in children with CI, which offer valuable information for both clinicians and researchers.

The review concluded that AT leads to improvements in outcomes related to trained tasks across all the included studies. This finding may inspire clinicians to make use of AT with their paediatric CI users. However, the review also revealed that the level of evidence was overall low. The lack of higher quality studies should not be interpreted as low effectiveness of AT but rather represents the practicalities of conducting such research with young children including recruiting appropriate sample sizes based on the apriori power calculation, utilizing control (preferably active controls), applying blinding, selecting proper outcome measures, monitoring participants' compliance, providing constrictive feedback, and assessing generalization of learning and long-term benefits.

The systematic review provided researchers with guidelines for designing AT interventions, developing training interventions, and selecting outcome measures. In addition, the review evaluated a variety of research designs and summarized essential measures that when utilized, would increase the reliability of the studies.

These guidelines were adopted for the design of the HIBA programme, producing a research evaluation of a higher level of evidence than before. It was not possible to fully meet all the requirements due to the COVID-19 pandemic which restricted the recruitment process and forced us to terminate the study prior assessing retention of improvement as planned.

5.6.2 Milestone II: The A-CAPT

There are a limited number of published tests that were validated with Saudi-Arabic speaking children. The scarcity of tools that assess consonant perception in Arabic speaking children in Saudi Arabia was the main motivation of this part of the project. Since speech perception tests are the most common outcome measures used to assess speech development in children with hearing impairment (Schaefer et al., 2017), absence of such tools may hinder the quality of service that clinicians can offer to those children. Without tools that can assess discrimination of speech cues and provide frequency-specific information with known reliability, it would be difficult for clinicians or researchers to verify the benefits of hearing devices or to assess effectiveness of habilitation interventions. Therefore, developing the A-CAPT was not only applicable for this research but also as a valuable tool for clinical and research use beyond this research.

Modern Standard Arabic language was used for the development of the test materials because it allows the test to be applicable to a greater number of populations.

In the validation stage of the A-CAPT, the scores were generally very high suggesting that the pictures and words were intelligible for the majority of normal hearing participants aged 5 years and older. The initial validation of the A-CAPT was conducted in the UK at the KFA, a Saudi-funded primary school. The children at this school who took part in this experiment were from Saudi Arabia and were exposed to the same type of Modern Standard Arabic as those children who participated in the final phase of the study that was conducted in Saudi Arabia. The only difference between these two groups was the fact that most of the children who participated in the study that was conducted in the UK were bilingual while most of the children who participated in the study conducted in Saudi Arabia were monolingual. Such a difference is not expected to be an issue since all the children were native Arabic speakers who were equally exposed to Modern Standard Arabic. This approach of using the formal form of the language is appropriate for other diglossic languages such as Arabic, Modern Greek, Swiss German, and Haitian Creole. Despite the range of dialects within the Saudi-Arabic language, children with different backgrounds were familiar with the materials that were presented in a closed-set format. Using this format where four alternative options are available makes it easier to elicit familiar responses from children and makes it applicable for Arabic speaking children regardless of their dialects.

This may also indicate that the test can be used with Arabic speaking children in non-Arabic speaking countries; it has been reported that Arabic is the fastest growing language in the United States (Brown, 2016). In addition, providing such tests can minimize the reported lack of assessment speech materials for children in the UK who speak languages other than English (Cattani et al., 2014), which is particularly important in large cities where the proportion of children where English is not the first language can be higher than for those who have English as the family language (Mehta et al., 2017).

5.6.3 Milestone III: HIBA

The lack of higher quality research that assesses AT intervention in children with CIs and the limited access to it was the main motivation behind this work. HIBA was developed as an evidenced-based multi-modal parent-led AT intervention programme. Clinicians and researchers are keen to interventions that are based on research with high ecological validity that relate to real-life listening. In addition, HIBA was developed for an underserved population, implanted Arabic-speaking children in Saudi Arabia where studies have shown that habiliation services for children with hearing loss are scarce (Alyami et al., 2016; Milaat et al., 2001), and HIBA is a step toward raising awareness and enforcing aural habilitation services for Arabic speaking children with CI in Saudi Arabia.

Limitations

Knowledge of the vocabulary that was used in the A-CAPT may vary between the participants in HIBA since the range of the participants' ages was wide (5-13 years). Although being unfamiliar with the test's vocabulary can be an issue when assessing speech perception, monitoring performance overtime and comparing it to a baseline would not be affected because for such within-subject comparisons the vocabulary level is unlikely to have changed over such short time scale. However, the analysis could have been affected by benefits of the training being masked due to the average age for the children in the multi-modal training group being higher (10.13 years) than the control group (8.29 years). Such discrepancy could be linked to brain plasticity which is greater at younger ages.

In addition, the art training for the control group may affect their performance as children and their parents appeared to be less motivated compared to the participants in the multi-modal training group. The parent-researcher interaction was minimal for participants’ in the control group and could have influenced the interaction between the parents and their children. Such inconsistency in parent-researcher interaction between the two groups may indicate that parents who were following the AT protocol were more motivated than those who were using the art training protocol, or it might indicate that the type of training (art) was less motivating for parents. This lack of motivation may explain the decline in the performance of some of the participants in the control group.

The way that HIBA was designed makes it difficult to tease out which training tasks or combination of training tasks contributed to the measured benefits. This issue was similar to an AT training study that (Ingvalson et al., 2014) used Earobics (Cognitive Concepts, 1997), which trains both phonological awareness and working memory skills simultaneously. The group of children who received the training showed significant improvements on language measures post intervention while the children in the control group showed no gains. The authors suggested that phonological and working memory training most likely led to improved language performance, but they could only speculate on which aspects of the training were most influential.

Blinding is a methodological approach to minimize research bias but it was difficult to apply in the HIBA trial that was conducted in Saudi Arabia because only the author was involved in the conduct of the study. All baseline assessments were conducted prior to randomization to group so participants and the researcher were not aware of the subjects’ group allocation. However, when the intervention programme started, it was clear which group the participants were assigned to. The intervention group was assigned tasks that involved auditory and speech perception while the control group was assigned tasks that involved art and craft. The researcher met with families to train them on their intervention approaches and also dealt with any queries or problems so it was not possible to apply blinding. In addition, the participants knew which groups that they were in, but they weren’t aware of whether one group was expected to lead to improvements above the other.

The sample size was small despite the efforts put into the recruitment process and the investment in the training materials including the advanced purchase of musical keyboards and art and craft materials. The recruitment was halted due to the COVID-19 pandemic so further batches of participants could not be recruited to meet the appropriate predicted sample size (from apriori power calculation). There are risks associated with being underpowered and it can lead to false findings, however the findings were interpreted with caution and post-hoc power calculated to better understand the findings. In addition, the second baseline was used in the analysis of the three outcome measures in HIBA. Such restriction was put in place despite the fact that we have a control group. Had we used the first baseline in the analysis, significant improvements post intervention in all measures would have been observed. However, with the small sample size we have, we opted to take the strictest potions to reduce bias and minimize false positive errors.

It is also informative to measure retention of benefits post intervention to understand any long-term carry over effects. Unfortunately, this part of the research was not possible due to the COVID-19 lockdown restrictions. The lack of assessment of retention contributed to reduction of the overall score on the quality of evidence of the study.

Finally, the findings of HIBA suggested that the duration of training (4 weeks) was possibly too short and masked some of the potential gains of HIBA. Other researchers (Welch et al., 2015; Yucel et al., 2009) who trained music and pitch discrimination for significantly longer (three months to two years) reported significant benefits while the measured benefits of pitch discrimination training in HIBA did not reach statistical significance. It would be interesting to re-conduct the pitch discrimination training for children with CIs to confirm if a longer intervention period leads to significant improvements.

5.7 Recommendations and Follow-up Work

To further investigate the impact of HIBA on speech and pitch perception in children with CIs, I would design a larger scale trial. This trial should have the appropriate larger sample size and would run for a school term as conducted by Welch et al., (2015). The participants should be assigned to six groups, a music and pitch training group, a communication and dialog training group, Modern Standard Arabic language training group, a multi-modal training, and a two active control group. A control group where parents are given general advice about language stimulation such as sharing books and interactive games, as well as a control group involving interactive non-auditory tasks. This would help to understand which components of HIBA affect selected outcome measures. A team of researchers needs to collaborate to achieve a double-blinded study to minimize bias and improve the quality of evidence. For the subjective questionnaire this should be given to participants and families at the final test session to ensure that responses are collected from all the participants including those in the control group. In addition, an interview with each family would be conducted to find out what they liked or did not like during trial. Finally, to assess long term benefits of the intervention, final assessment of retention following a period without intervention should be conducted.

5.8 CONCLUSION

Performance of children with CIs vary post-implantation as some children exhibit limited benefit and their speech perception is poor while others can attain speech perception abilities comparable to their normal hearing peers. Many of the factors that contribute to the variability in the outcomes of children with CIs are predetermined and in cases cannot be amended. However, AT has shown to improve speech perception in children with CIs.

Here, we developed HIBA, an evidenced-based multi-modal parent-led AT intervention programme, to improve speech and pitch perception in children with CI based on research with high ecological validity that relate to real-life listening. HIBA was carefully designed to be amongst the studies with higher level of quality of evidence. Had we been able to recruit the sample size recommended by the a priori calculation and conduct post training retention assessment, the HIBA study would have scored high (appose to moderate) in the grading scale used to assess the quality of evidence-based AT studies. Moreover, to properly assess the outcomes of HIBA as an intervention, the A-CAPT was developed and successfully validated and used for the first time. Here, we offer this reliable measure as a novel phoneme discrimination test in Arabic.

Benefits were observed in HIBA for speech cue discrimination measured by A-CAPT. Our findings suggest that children with CIs and their parents may benefit from regular and sustained access to age-appropriate auditory training materials and activities. Results also indicate that empowering families by giving them an active role in their children’s care plan is commended and that parents were compliant, fully engaged, and able to implement home-based intervention when provided with clear plans.

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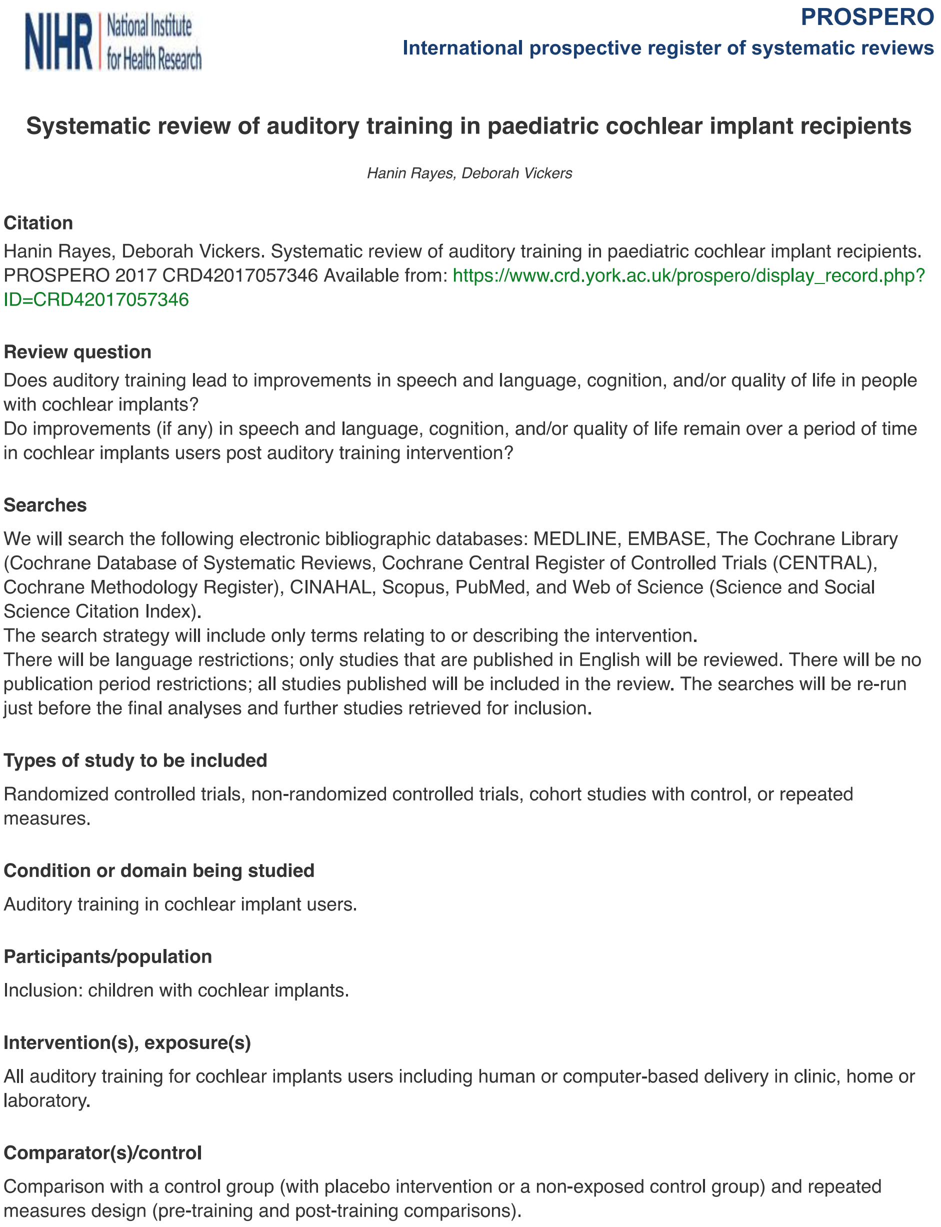
APPENDIX

Appendix I: Registering the systematic review at PROSPERO database



PROSPERO
International prospective register of systematic reviews

Systematic review of auditory training in paediatric cochlear implant recipients

Hanin Rayes, Deborah Vickers

Citation

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Review question

Does auditory training lead to improvements in speech and language, cognition, and/or quality of life in people with cochlear implants?
Do improvements (if any) in speech and language, cognition, and/or quality of life remain over a period of time in cochlear implants users post auditory training intervention?

Searches

We will search the following electronic bibliographic databases: MEDLINE, EMBASE, The Cochrane Library (Cochrane Database of Systematic Reviews, Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Methodology Register), CINAHAL, Scopus, PubMed, and Web of Science (Science and Social Science Citation Index).
The search strategy will include only terms relating to or describing the intervention.
There will be language restrictions; only studies that are published in English will be reviewed. There will be no publication period restrictions; all studies published will be included in the review. The searches will be re-run just before the final analyses and further studies retrieved for inclusion.

Types of study to be included

Randomized controlled trials, non-randomized controlled trials, cohort studies with control, or repeated measures.

Condition or domain being studied

Auditory training in cochlear implant users.

Participants/population

Inclusion: children with cochlear implants.

Intervention(s), exposure(s)

All auditory training for cochlear implants users including human or computer-based delivery in clinic, home or laboratory.

Comparator(s)/control

Comparison with a control group (with placebo intervention or a non-exposed control group) and repeated measures design (pre-training and post-training comparisons).

Context

Auditory training intervention must be interactive requiring users' involvement in individual or group basis. Range of auditory training settings will be included such as interventions delivered by clinicians in clinical settings and interventions delivered via a personal computer, tablet or other technological devices which can be conducted in either laboratory, schools or home environment.

Main outcome(s)

Improvements in speech perception (words and sentences recognition in quiet and noise), cognitive abilities (working memory, executive function, and attention), and/or quality of life (family or self reported feedback related to improved communication, if any).

Additional outcome(s)

Retention of improvements on trained or untrained measures post auditory intervention is ceased, and generalization of improved skills from trained tasks to other learning domains

Data extraction (selection and coding)

Study selection: Inclusion and exclusion criteria were established based on PICOS strategy (CRD, 2008).
Participants: cochlear implants recipients, both adults and children;
Intervention: interactive individual or group computer-based or clinician-based auditory training; Controls: pre/post training outcomes or comparison with a control group;
Outcomes: measures of speech perception, cognitive ability, and quality of life;
Study design: Randomized controlled trials, non-randomized controlled trials, repeated measures, or cohort studies with controls.
Data, including participants (age and compliance), sample size, study design, training type, training stimuli, controls, randomization, outcomes , study findings, funding and limitation, will be extracted by the two authors,

Risk of bias (quality) assessment

The two authors will independently assess the risk of bias based on the following characteristics: randomization, blinding, controls, power calculation, selective reporting of outcome measures, training feedback, participants self assessment and generalization improvements if any.

Strategy for data synthesis

Meta-analysis will not be feasible due to the wide range of outcome measures across the selected studies.
We plan to report the findings from the selected trials as a narrative stating the study findings ((no)improvements on the trained task and untrained tasks) including sample size, study design, training type, training stimuli, and retention post training.

Analysis of subgroups or subsets

None planned.

Contact details for further information

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Review team members and their organisational affiliations

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Type and method of review

Intervention, Systematic review

Anticipated or actual start date

02 February 2017

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26 June 2017

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None.

Conflicts of interest

None known

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England

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Review Completed not published

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Subject index terms

Cochlear Implantation; Cochlear Implants; Cognition; Humans; Language; Quality of Life; Speech; Speech Perception

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Details of any existing review of the same topic by the same authors

Stage of review at time of this submission

Stage	Started	Completed

Appendix II: Inclusion and Exclusion Criteria According to PICOS

Inclusion and exclusion criteria were defined as follow:

- **Participants:** Children (<18 years old) with cochlear implants
- **Intervention(s):** All auditory training for cochlear implants users including human or computer-based delivery in clinic, home, school, or laboratory.
- **Comparator(s)/Control** Comparison with a control group (with placebo intervention or a non-exposed control group) and repeated-measures design (pre-training and post-training comparisons).
- **Outcome(s):** Improvements in speech perception (words and sentences recognition in quiet and noise), cognitive abilities (working memory, executive function, and attention), and/or quality of life (family or self-reported feedback related to improved communication, if any). Retention of benefits when AT ceases and generalization of learning.
- **S (Study Design):** Randomized-Control Trials (RCT), non-RCT, repeated measures, or cohort studies with controls.

Appendix III: Guidelines for Level of Evidence

Henshaw & Ferguson (2013) developed guidelines for evaluation AT studies, which categorize the level of evidence according to study quality scores (a sum of graded predefined measures) as follow:

- Scores between 0–5 are deemed very low, indicating that the estimation of effect is unreliable.
- Scores between 6–10 are deemed low, indicating that further evidence is very likely to impact on our confidence in the estimation of effect and are likely to alter the estimate.
- Scores between 11–15 are deemed moderate, indicating further evidence is likely to impact on our confidence in the estimation of effect and may alter the estimate.
- Scores between 16–20 are deemed high, indicating further evidence is very unlikely to alter our confidence in the estimation of effect.

Appendix IV: This table shows the final version of the A-CAPT lists, the phonetic transcription for each stimulus and its translation to English.

A-CAPT Lists					
Easy List			Hard List		
Arabic	English Translation	Phonetic Transcription	Arabic	English Translation	Phonetic Transcription
سور	Fence	/suːr/	ناس	People	/naːs/
دور	Role	/duːr/	ماس	Diamonds	/maːs/
نور	Light	/nuːr/	داس	Trampled	/daːs/
حور	Poplar	/ħuːr/	باس	Bass	/baːs/
جد	Grandfather	/jad/	عم	Uncle	/ʕam/
سد	Dam	/sad/	فم	Mouth	/fam/
يد	Hand	/yad/	كم	How many	/kam/
خد	Cheek	/xad/	دم	Blood	/dam/
خط	Line	/xatˤ/	حاج	Pilgrim	/ħa:dʒ/
خس	Lettuce	/xas/	حاب	Love	/ħa:b/
خل	Vinegar	/xal/	حاد	Sharp	/ħa:d/
خد	Cheek	/xad/	حار	Hot	/ħa:r/
قاس	Garsh	/qa:s/	كب	Spilled	/kab/
قال	He said	/qa:l/	كف	Palm	/kaf/
قام	Stood-up	/qa:m/	كم	How many	/kam/
قاد	Drove	/qa:d/	كح	Lakh	/kaħ/
نار	Fire	/naːr/	طين	Mud/clay	/tˤiːn/
دار	House	/daːr/	لين	Lin	/liːn/
طار	Flew	/tˤaːr/	تين	Figs	/tiːn/
حار	Hot	/ħaːr/	دين	Religion	/diːn/
بط	Duck	/batˤ/	حر	free	/ħar/
خط	Line	/xatˤ/	بر	Desert	/bar/
نط	Jump	/natˤ/	جر	drag	/jar/
مط	Stretch	/matˤ/	مر	Passed	/mar/
سوس	Mite/plaque	/suːs/	طيب	Perfume	/tˤiːb/
سود	Black	/suːd/	طيح	Fall down	/tˤiːħ/
سور	Fence	/suːr/	طين	Mud/clay	/tˤiːn/
سوق	Mall/market	/suːq/	طير	bird	/tˤiːr/
موت	Death	/mawt/	بر	Desert	/bar/
موز	Banana	/mawz/	برد	Cold	/bard/
موج	Waves	/mawdʒ/	بط	duck	/batˤ/
مر	Passed	/mar/	برق	Lightning	/barq/

Appendix V: This table shows the participants' demographic, age, age at implantation, duration of CI use, number of participants who are bilateral users of CI, number of females. Ages were shown in years.

	Subject	Age	Age at Implant	Duration of CI Use	Number Bilateral	Number of Females
Multi-modal Training	1	10.6	3.6	7	0	0
	2	7.9	4.9	3	0	1
	3	12.8	5.8	7	0	0
	4	11.5	4.5	7	0	0
	5	10.2	4.1	6.1	0	1
	6	11.9	5.2	6.7	1	0
	7	6	3	3	0	1
	Mean	10.13	4.44	5.69	1	3
	SD	2.39	0.96	1.86		
Control	8	7	3	4	0	1
	9	5	3.5	2.5	1	1
	10	13	3	10	0	0
	11	7	2	5	0	0
	12	5	1.3	3.7	0	1
	13	9	4	5	0	1
	14	12.1	4.5	7.6	0	1
	Mean	8.3	3.04	5.4	1	5
	SD	3.22	1.11	2.57		

Appendix VI: This parents’ questionnaire assessed parents’ perception toward the training approach and perceived benefits through asking 6 questions 5 of which were close-ended questions while the sixth one was an open-ended question that encouraged parents to describe their experience with the training program in their own word.

Questions	**Answer**		
	Yes	**No**	**Somewhat**
Do you think the training tasks were entertaining for your child?	7	0	0
Do you think the training sessions were beneficial to your child?	6	0	1
Do you think the duration of the training session (30 min) were appropriate?	6	0	1
Did you like performing the training sessions independently at home?	6	0	1
Would you participate in other auditory training activities in the future?	6	0	1
Please describe your experience with this auditory training program below?			

Appendix VII: This children’s questionnaire assessed the training approach and engagement in a child-friendly approach. Scoring was based on a binary response of whether they enjoyed the program or not given a pass or fail score for each child.

<u>Children’s Questionnaire</u>

Did you have fun?	☐ Yes	☐ No
Would you participate in similar activates again?	☐ Yes	☐ No
Which was your favourite activity?	☐ Finding the differences	☐ Words building/rhyming
	☐ Keyboard/Music	☐ None

Appendix VIII: Parents and children responses to questionnaires. Scores for parents' views were calculated according to their responses to the five closed-ended questions, in a form of scale from 0 to 2. Maximum score for each question was 2 and maximum score for the

Questionnaire		
Participant Code	**Parent Scores**	**Children Scores**
1	10	Pass
2	10	Pass
3	10	Pass
4	6	Pass
5	10	Pass
6	10	Pass
7	10	Pass
Mean	9.43	100%
SD	1.51	