Systematic Review of Auditory Training in Paediatric Cochlear Implant Recipient

Hanin Rayes

Department of Speech Hearing and Phonetic Sciences, Faculty of Brain Sciences, University

College London, London. UK

Ghada Al-Malky

Ear Institute, Faculty of Brain Sciences, University College London, London. UK

Deborah Vickers

Department of Clinical Neurosciences, Clinical School, University of Cambridge,

Cambridge. UK

Department of Speech Hearing and Phonetic Sciences, Faculty of Brain Sciences, University

College London, London. UK

Corresponding Author

Hanin Rayes

Department of Speech Hearing and Phonetic Sciences. Faculty of Brain Sciences.

University College London, London. UK

Hanin.rayes.16@ucl.ac.uk

+447376080286

Conflict of Interest: there are no conflicts of interest

Funding: This research is funded by a Ph.D. scholarship from King Abdulaziz University.

ABSTRACT

Objective: The purpose of this systematic review is to evaluate the published research in Auditory training (AT) for paediatric CI recipients. This review investigates 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.

Method: A systematic search of seven databases identified 96 papers published up until January 2017, nine of which met the inclusion criteria. Data were extracted and independently assessed for risk of bias and quality of study against a PICOS framework.

Results: All studies reported improvements in trained AT tasks including speech discrimination/identification and working memory. Retention of improvements overtime was found whenever it was assessed. Transfer of learning was measured in four out of six studies, which assessed generalization. Quality of life was not assessed. Overall, evidence for the included studies was deemed to be of low quality.

Conclusion: Benefits of AT were illustrated through the improvement in trained tasks and this was observed in all 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 needed.

Keywords: aural rehabilitation, auditory training, children, cochlear implants, systematic review.

Systematic Review of Auditory Training in Paediatric Cochlear Implant Recipients

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.

Cochlear implants (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 exeptionally 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 auditory-training (AT) programmes may help.

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).

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 Filippo & Scott, 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

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.

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.

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).

Research Aims

The primary aim of this systematic review was to investigate whether AT is effective at improving performance scores for pediatric 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 pediatric patients using CIs and provide researchers with the latest AT findings for pediatric CI recipients (See Appendix A).

METHODS

A systematic review protocol was prepared and registered with PROSPERO (2017: CRD42017057346), 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 B).

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 1).

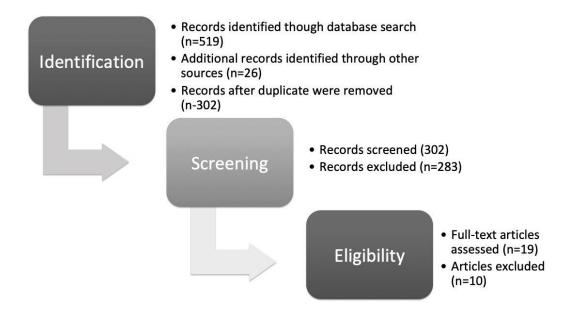


Fig.1 Process of papers selection

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 C) (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).

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.

RESULTS

The analysed studies and their findings were summarised in two tables. Table 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 summarizes the main findings including, improvement, retention, generalization of learning, and compliance. The tables can be found at the end of the document.

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).

Quality of the Studies

Quality of the studies in addition to their level of evidence is listed in Table 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.

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.

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 program 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 visualspatial awareness.

Ingvalson *et al.* (2013) used Earobics ("Earobics: Auditory development and phonics program [Computer software]," 1997), which trains both phonological awareness and workingmemory 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.

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 focussed 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 program 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 program, 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.

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 auditorymemory 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 program 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.

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 WM 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 WM measure was retained for up to 1 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.

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 pilot studies. 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.

AT Approaches.

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 program. 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 training pitch discrimination, rhythm discrimination, and sequence repetition. All four studies reported improved skills on trained asks.

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.

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.

DISCUSSION

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 WM training does not improve other skills that are not WM specific, including speech perception. However, there is no evidence either that such findings apply to CI users since the number of WM 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.

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 outcomes measures used across studies, generalization and retention of benefits can be the focus of future studies as a primary AT outcomes regardless to the measures used in the studies, and eventually be investigated in a meta-analysis.

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.

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 program should also be incorporated and be considered as key indicators to the success of any AT programme.

Acknowledgments

We would like to thank the librarian Alex Stagg at the UCL Ear Institute for his assistance with database selection, formulating relevant keywords and conducting the systematic literature search. We also would like to thank two anonymous reviewers for their valuable comments and feedback. Debi Vickers was supported by an MRC Senior Research Fellowship in Hearing (MR/S002537/1).

REFERENCES

Amitay, S., Irwin, A., & Moore, D. R. (2006). Discrimination learning induced by training with identical stimuli. *Nature Neuroscience*, 9(11), 1446-1448.

https://doi.org/10.1038/nn1787

Anderson, S., & Kraus, N. (2013). Auditory Training: Evidence for Neural Plasticity in Older
Adults. *Perspectives on Hearing & Hearing Disorders Research & Diagnostics*, 17, 3757. https://doi.org/10.1044/hhd17.1.37

- Anvari, S. H., Trainor, L. J., Woodside, J., & Levy, B. A. (2002). Relations among musical skills, phonological processing, and early reading ability in preschool children. *The Journal of Experimental Child Psychology*, 83(2), 111-130.
- Atkins, D., Best, D., Briss, P. A., Eccles, M., Falck-Ytter, Y., Flottorp, S., . . . Zaza, S. (2004).
 Grading quality of evidence and strength of recommendations. *British Medical Journal*, 328(7454), 1490. <u>https://doi.org/10.1136/bmj.328.7454.1490</u>
- Barrett, K. C., Ashley, R., Strait, D. L., & Kraus, N. (2013). Art and science: how musical training shapes the brain. *Frontiers in Psychology*, *4*, 713. https://doi.org/10.3389/fpsyg.2013.00713
- Barton, C., & Robbins, A. M. (2015). Jumpstarting auditory learning in children with cochlear implants through music experiences. Cochlear Implants International, 16 Suppl 3, S51-62. <u>https://doi.org/10.1179/1467010015z.00000000267</u>
- Bauer, P. W., Sharma, A., Martin, K., & Dorman, M. (2006). Central auditory development in children with bilateral cochlear implants. *Archives of Otolaryngology–Head & Neck Surgery*, 132(10), 1133-1136. <u>https://doi.org/10.1001/archotol.132.10.1133</u>
- Besson, M., Schon, D., Moreno, S., Santos, A., & Magne, C. (2007). Influence of musical expertise and musical training on pitch processing in music and language. *Restorative neurology and neuroscience*, 25(3-4), 399-410.
- Blamey, P., Arndt, P., Bergeron, F., Bredberg, G., Brimacombe, J., Facer, G., . . . Whitford, L. (1996). Factors affecting auditory performance of postlinguistically deaf adults using cochlear implants. *Audiology and Neuro-Otology*, 1(5), 293-306.
- Blamey, P. J., Maat, B., Baskent, D., Mawman, D., Burke, E., Dillier, N., . . . Lazard, D. S.(2015). A Retrospective Multicenter Study Comparing Speech Perception Outcomes for

Bilateral Implantation and Bimodal Rehabilitation. *Ear and Hearing*, *36*(4), 408-416. https://doi.org/10.1097/AUD.000000000000150

Brattico, E., Tervaniemi, M., & Picton, T. W. (2003). Effects of brief discrimination-training on the auditory N1 wave. *NeuroReport*, 14(18), 2489-2492.

https://doi.org/10.1097/01.wnr.0000098748.87269.a1

- Briaire, J. J., & Frijns, J. H. (2006). The consequences of neural degeneration regarding optimal cochlear position in scala tympani: a model approach. *Hearing Research*, 214(1-2):17-27. <u>https://doi.org/10.1016/j.heares.2006.01.015</u>
- Burk, M. H., & Humes, L. E. (2008). Effects of long-term training on aided speech-recognition performance in noise in older adults. *Journal of Speech Language and Hearing Research*, 51(3), 759-771. <u>https://doi.org/10.1044/1092-4388(2008/054)</u>
- Burk, M. H., Humes, L. E., Amos, N. E., & Strauser, L. E. (2006). Effect of training on wordrecognition performance in noise for young normal-hearing and older hearing-impaired listeners. *Ear and Hearing*, 27(3), 263-278.

https://doi.org/10.1097/01.aud.0000215980.21158.a2

- Burns, P. B., Rohrich, R. J., & Chung, K. C. (2011). The Levels of Evidence and their role in Evidence-Based Medicine. Plastic and Reconstructive Surgery, 128(1), 305-310. https://doi.org/10.1097/PRS.0b013e318219c171
- Chen, J. K., Chuang, A. Y., McMahon, C., Hsieh, J. C., Tung, T. H., & Li, L. P. (2010). Music training improves pitch perception in prelingually deafened children with cochlear implants. *Pediatrics*, 125(4), e793-800. <u>https://doi.org/10.1542/peds.2008-3620</u>

- Connor, C. M., Craig, H. K., Raudenbush, S. W., Heavner, K., & Zwolan, T. A. (2006). The age at which young deaf children receive cochlear implants and their vocabulary and speechproduction growth: is there an added value for early implantation? *Ear and Hearing*, 27(6), 628-644. <u>https://doi.org/10.1097/01.aud.0000240640.59205.42</u>
- Culp, M. E. (2017). The Relationship Between Phonological Awareness and Music Aptitude. Journal of Research in Music Education, 65(3), 328-346.

https://doi.org/10.1177/0022429417729655

De Bruyn, L., Philips, B., Moelants, D., Coussement, P., Leman, M., Dhooge, I. (2011). E087 Sound Caterpillar – an interactive, musical auditory training game for hearing impaired children. *International Journal of Pediatric Otorhinolaryngology*. 75. 77-77. https://doi.org/10.1016/S0165-5876(11)70395-0

De Filippo, C. L., & Scott, B. L. (1978). A method for training and evaluating the reception of

ongoing speech. The Journal of the Acoustical Society of America, 63(4), 1186-1192.

Earobics: Auditory development and phonics program [Computer software]. . (1997).

Cambridge, MA: Cognitive Concepts. https://doi.org/10.1044/0161-1461.3001.108

- Dubno, J.R., & Levitt, H. (1981). Predicting consonant confusions from acoustic analysis. Journal of the Acoustical Society of America, 69(1), 249–61.
- Finley, C. C., Holden, T. A., Holden, L. K., Whiting, B. R., Chole, R. A., Neely, G. J., . . . Skinner, M. W. (2008). Role of electrode placement as a contributor to variability in cochlear implant outcomes. *Otology and Neurotology*, 29(7), 920-928.https://doi.org/10.1097/MAO.0b013e318184f492
- Firszt, J. B., Holden, L. K., Skinner, M. W., Tobey, E. A., Peterson, A., Gaggl, W., . . . Wackym,P. A. (2004). Recognition of speech presented at soft to loud levels by adult cochlear

implant recipients of three cochlear implant systems. *Ear and Hearing*, 25(4), 375-387.
Friedland, D. R., Venick, H. S., & Niparko, J. K. (2003). Choice of ear for cochlear implantation: the effect of history and residual hearing on predicted postoperative performance. *Otology and Neurotology*, 24(4), 582-589. Fu, Q. J., & Galvin, J. J. (2008).
Maximizing cochlear implant patients' performance with advanced speech training procedures. *Hearing Research*, 242(1-2), 198-

208.<u>https://doi.org/10.1016/j.heares.2007.11.010</u>

- Fu, Q. J., Galvin, J.J., Wang, X., & Nogaki, G. (2004). Effects of auditory training on adult cochlear implant patients: a preliminary report. *Cochlear Implants International, 5 Suppl 1*, 84-90. <u>https://doi.org/10.1179/cim.2004.5.Supplement-1.84</u>
- Fu, Q. J., Galvin, J. J., Wang, X., & Wu, J. L. (2015). Benefits of music training in mandarinspeaking pediatric cochlear implant users. Journal of Speech Language and Hearing Research, 58(1), 163-169. https://doi.org/10.1044/2014_jslhr-h-14-0127
- Galvin, J. J., Fu, Q. J., & Shannon, R. V. (2009). Melodic contour identification and music perception by cochlear implant users. *Annals of the New York Academy of Sciences*, *1169*, 518-533. https://doi.org/10.1111/j.1749-6632.2009.04551.x
- Good, A., Gordon, K. A., Papsin, B. C., Nespoli, G., Hopyan, T., Peretz, I., & Russo, F. A.(2017). Benefits of music training for perception of emotional speech prosody in deaf children with cochlear implants. *Ear and hearing*, *11*, 11.

https://doi.org/10.1097/AUD.00000000000402

Hagr, A., Garadat, S. N., Hassan, S. M., Malki, K., Al Ohali, Y., Al Ghamdi, N., . . . Al Hamid,S. (2016). The effect of the Arabic computer rehabilitation program "Rannan" on sound

detection and discrimination in children with cochlear implants. *Journal of the American Academy of Audiology*, 27(5), 380-387. <u>https://doi.org/10.3766/jaaa.15039</u>

Henshaw, H., & Ferguson, M. A. (2013). Efficacy of individual computer-based auditory training for people with hearing loss: a systematic review of the evidence. *Plos One*, 8(5), e62836. <u>https://doi.org/10.1371/journal.pone.0062836</u>

Herholz, S. C., & Zatorre, R. J. (2012). Musical training as a framework for brain plasticity: behavior, function, and structure. *Neuron*, 76(3), 486-502. <u>https://doi.org/10.1016/j.neuron.2012.10.011</u>

- Holden, L. K., Vandali, A. E., Skinner, M. W., Fourakis, M. S., & Holden, T. A. (2005). Speech recognition with the advanced combination encoder and transient emphasis spectral maxima strategies in nucleus 24 recipients. *Journal of Speech, Language, and Hearing Research*, 48(3), 681-701. <u>https://doi.org/10.1044/1092-4388(2005/047)</u>
- IEEE (1969) IEEE Recommended Practice for Speech Quality Measurements. IEEE Transactions on Audio and Electroacoustics, 17, 225–246.
- Ingvalson, E. M., & Wong, P. C. (2013). Training to improve language outcomes in cochlear implant recipients. *Frontiers in Psychology*, *4*, 263.

https://doi.org/10.3389/fpsyg.2013.00263

Ingvalson, E. M., Young, N. M., & Wong, P. C. (2014). Auditory-cognitive training improves language performance in prelingually deafened cochlear implant recipients. *International Journal of Pediatric Otorhinolaryngology*, 78(10), 1624-1631.

https://doi.org/10.1016/j.ijporl.2014.07.009

Jeng, F. C., Hu, J., Dickman, B., Montgomery-Reagan, K., Tong, M., Wu, G., & Lin, C. D. (2011). Cross-linguistic comparison of frequency-following responses to voice pitch in American and Chinese neonates and adults. Ear and Hearing, 32(6), 699-707.

https://doi.org/10.1097/AUD.0b013e31821cc0df

- Kane, M., Schopmeyer, B., Mellon, N. K., Wang, N., & Niparko, J. K. (2004). Prelinguistic communication and subsequent language acquisition in children with cochlear implants. *Archives of Otolaryngology–Head and Neck Surgery*, 130(5), 619-623. https://doi.org/10.1001/archotol.130.5.619
- Kant, A., & Adhyaru, M. (2009). Home auditory training program (HAP) for cochlear implantees and hearing impaired children using hearing aids-an outcome of a three-year research project. *Indian journal of otolaryngology and head and neck surgery : official publication of the Association of Otolaryngologists of India*, 61(1), 54-8.
- Kaplan, H., Bally, S., Brandt, F., Busacco, D., & Pray, J. (1997). Communication Scale for Older Adults (CSOA). *Journal of the American Academy of Audiology*, 8, 203–21
- Khan, A. M., Handzel, O., Burgess, B. J., Damian, D., Eddington, D. K., & Nadol, J. B., Jr. (2005). Is word recognition correlated with the number of surviving spiral ganglion cells and electrode insertion depth in human subjects with cochlear implants? *Laryngoscope*, *115*(4), 672-677. https://doi.org/10.1097/01.mlg.0000161335.62139.80
- Kiessling, J., Pichora-Fuller, M. K., Gatehouse, S., Stephens, D., Arlinger, S., Chisolm, T., . . . von Wedel, H. (2003). Candidature for and delivery of audiological services: special needs of older people. *International Journal of Audiology, 42 Suppl 2*, 2s92-101.
- Klingberg, T., Fernell, E., Olesen, P. J., Johnson, M., Gustafsson, P., Dahlström, K., . . .Westerberg, H. (2005). Computerized Training of Working Memory in Children WithADHD-A Randomized, Controlled Trial. *Journal of the American Academy of Child &*

Adolescent Psychiatry, 44(2), 177-186. <u>https://doi.org/10.1097/00004583-200502000-</u> 00010

- Killion, M.C., Niquette, P.A., Gudmundsen, G.I., Revit, L.J., Banerjee, S. (2004). Development of a quick speech-in-noise test for measuring signal-to-noise ratio loss in normal-hearing and hearing-impaired listeners. *Journal of the Acoustical Society of America*, 116, 2395– 2405.
- Kral, A., Kronenberger, W. G., Pisoni, D. B., & O'Donoghue, G. M. (2016). Neurocognitive factors in sensory restoration of early deafness: a connectome model. *The Lancet Neurology*, 15(6), 610-621. http://dx.doi.org/10.1016/S1474-4422(16)00034-X
- Kraus, N., & Chandrasekaran, B. (2010). Music training for the development of auditory skills. *Nature Reviews Neuroscience*, *11*(8), 599-605. <u>https://doi.org/10.1038/nrn2882</u>
- Kraus, N., Skoe, E., Parbery-Clark, A., & Ashley, R. (2009). Experience-induced malleability in neural encoding of pitch, timbre, and timing. *Annals of the New York Academy of Sciences*, 1169, 543-557. https://doi.org/10.1111/j.1749-6632.2009.04549.x
- Kricos, P. B., & Holmes, A. E. (1996). Efficacy of audiologic rehabilitation for older adults. J Am Acad Audiol, 7(4), 219-229. Krishnan, A., Xu, Y., Gandour, J., & Cariani, P. (2005).
 Encoding of pitch in the human brainstem is sensitive to language experience. Cognitive Brain Research, 25(1), 161-168. <u>https://doi.org/10.1016/j.cogbrainres.2005.05.004</u>
- Krizman, J., Marian, V., Shook, A., Skoe, E., & Kraus, N. (2012). Subcortical encoding of sound is enhanced in bilinguals and relates to executive function advantages. *Proceedings of the National Academy of Sciences of the United States of America*, 109(20), 7877-7881. <u>https://doi.org/10.1073/pnas.1201575109</u>

Kronenberger, W. G., Pisoni, D. B., Henning, S. C., Colson, B. G., & Hazzard, L. M. (2011).
Working memory training for children with cochlear implants: a pilot study. *Journal of Speech Language and Hearing Research*, *54*(4), 1182-1196. <u>http://doi.org/10.1044/1092-4388(2010/10-0119)</u>

Manson, K. (2017). *Executive Function In Deaf Children: An Intervention Study*.
(doctoral dissertation), University College London, London, United Kingdom. Markman,
T. M., Quittner, A. L., Eisenberg, L. S., Tobey, E. A., Thal, D., Niparko, J. K., . . . The,
C. I. T. (2011). Language development after cochlear implantation: an epigenetic model. *Journal of Neurodevelopmental Disorders*, 3(4), 388-404.

https://doi.org/10.1007/s11689-011-9098-z

Melby-Lervag, M., Redick, T. S., & Hulme, C. (2016). Working Memory Training Does Not Improve Performance on Measures of Intelligence or Other Measures of "Far Transfer": Evidence From a Meta-Analytic Review. *Perspectives on Psychological Science*, *11*(4), 512-534. <u>https://doi.org/10.1177/1745691616635612</u>

Mens, L. H., & Berenstein, C. K. (2005). Speech perception with mono- and quadrupolar electrode configurations: a crossover study. *Otology and Neurotology*, *26*(5), 957-964. https://doi.org/10.1097/01.mao.0000185060.74339.9d

- Mishra, S. K., Boddupally, S. P., & Rayapati, D. (2015). Auditory learning in children with cochlear implants. *Journal of Speech, Language, and Hearing Research*, 58(3), 1052-1060. <u>https://doi.org/10.1044/2015_JSLHR-H-14-0340h</u>
- Nadol, J. B., Jr., Young, Y. S., & Glynn, R. J. (1989). Survival of spiral ganglion cells in profound sensorineural hearing loss: implications for cochlear implantation. *Annals of*

Otology, Rhinology, and Laryngology, 98(6), 411-416.

https://doi.org/10.1177/000348948909800602

- Newman, C.W., Weinstein, B.E., Jacobson, G.P., & Hug, G.A. (1990). The Hearing Handicap Inventory for Adults: psychometric adequacy and audiometric correlates. *Ear and Hearing*. 11, 430–433
- Nikolopoulos, T. P., Wells, P., & Archbold, S. M. (2000). Using Listening Progress Profile (LIP) to assess early functional auditory performance in young implanted children. *Deafness & Education International*, 2(3), 142-151. <u>https://doi.org/10.1179/146431500790561125</u>
- Nilsson, M., Soli, S.D., & Sullivan, J.A. (1994). Development of the Hearing in Noise Test for the measurement of speech reception thresholds in quiet and in noise. *The Journal of the Acoustical Society of America*, 95, 1085–1099.
- Niparko, J. K., & Blankenhorn, R. (2003). Cochlear implants in young children. *Mental retardation and developmental disabilities research reviews*, 9(4), 267-275. <u>https://doi.org/10.1002/mrdd.10089</u>
- Niparko, J. K., Tobey, E. A., Thal, D. J., Eisenberg, L. S., Wang, N. Y., Quittner, A. L., & Fink,
 N. E. (2010). Spoken language development in children following cochlear implantation. *JAMA : the journal of the American Medical Association, 303*(15), 1498-1506.
 https://doi.org/10.1001/jama.2010.451
- Nogueira, W., Litvak, L. M., Saoji, A. A., & Buchner, A. (2015). Design and evaluation of a cochlear implant strategy based on a "Phantom" channel. *Plos One*, *10*(3), e0120148. <u>https://doi.org/10.1371/journal.pone.0120148</u>

- Oba, S. I., Fu, Q. J., & Galvin, J. J. (2011). Digit training in noise can improve cochlear implant users' speech understanding in noise. *Ear and Hearing*, *32*(5), 573-581. https://doi.org/10.1097/AUD.0b013e31820fc821
- Patel, A. D. (2011). Why would Musical Training Benefit the Neural Encoding of Speech? The OPERA Hypothesis. *Frontiers in Psychology*, *2*, 142.

https://doi.org/10.3389/fpsyg.2011.00142

- Perin da Siliva, M., Comerlatto Junior, A., Andreoli Balen, S., & Bevilacqua, M. (2012).
 Software use in the (re)habilitation of hearing impaired children. *Jornal Da Sociedade Brasileira De Fonoaudiologia*, 24:34–41. http://dx.doi.org/10.1590/S2179-64912012000100007
- Pfingst, B. E., Franck, K. H., Xu, L., Bauer, E. M., & Zwolan, T. A. (2001). Effects of Electrode Configuration and Place of Stimulation on Speech Perception with Cochlear Prostheses. *Journal of the Association for Research in Otolaryngology*, 2(2), 87-103. <u>https://doi.org/10.1007/s101620010065</u>
- Ponton, C. W., Don, M., Eggermont, J. J., Waring, M. D., & Masuda, A. (1996). Maturation of human cortical auditory function: differences between normal-hearing children and children with cochlear implants. *Ear and Hearing*, 17(5), 430-437
- Pulsifer, M. B., Salorio, C. F., & Niparko, J. K. (2003). Developmental, audiological, and speech perception functioning in children after cochlear implant surgery.
 Archives of Pediatrics & Adolescent Medicine, 157(6), 552-558.
 https://doi.org/10.1001/archpedi.157.6.552
- Richardson, W.S., Wilson, M.C., Nishikawa, J., Hayward, R.S. (1995) The well-built clinical question: a key to evidence-based decisions. *ACP Journal Club*, 123(3):A12–3

- Rochette, F., & Bigand, E. (2009). Long-term effects of auditory training in severely or profoundly deaf children. *Annals of the New York Academy of Sciences*, 1169, 195-198. <u>https://doi.org/10.1111/j.1749-6632.2009.04793.x</u>
- Rochette, F., Moussard, A., & Bigand, E. (2014). Music lessons improve auditory perceptual and cognitive performance in deaf children. *Frontiers in Human Neuroscience*, 8, 488. <u>https://doi.org/10.3389/fnhum.2014.00488</u>
- Roman, S., Rochette, F., Triglia, J. M., Schön, D., & Bigand, E. (2016). Auditory training improves auditory performance in cochlear implanted children. *Hearing Research*, 337, 89-95. <u>http://dx.doi.org/10.1016/j.heares.2016.05.003</u>
- Ross, B., & Tremblay, K. (2009). Stimulus experience modifies auditory neuromagnetic responses in young and older listeners. *Hearing Research*, 248(1-2), 48-59. <u>https://doi.org/10.1016/j.heares.2008.11.012</u>
- Rubinstein, A., & Boothroyd, A. (1987). Effect of two approaches to auditory training on speech recognition by hearing-impaired adults. *Journal of Speech & Hearing Research*, 30(2), 153-160.
- Rubinstein, J. T., Parkinson, W. S., Tyler, R. S., & Gantz, B. J. (1999). Residual speech recognition and cochlear implant performance: effects of implantation criteria. *American Journal of Otolaryngology*, 20(4), 445-452.
- Schow, R. L., & Nerbonne, M. A. (2007). *Introduction to Audiologic Rehabilitation*: Pearson Allyn and Bacon.
- Seyyedi, M., Viana, L. M., & Nadol, J. B. (2014). Within-Subject Comparison of Word Recognition and Spiral Ganglion Cell Count in Bilateral Cochlear Implant Recipients.

Otology and Neurotology, 35(8), 1446-1450.

https://doi.org/10.1097/MAO.000000000000443

- Shahin, A. J. (2011). Neurophysiological Influence of Musical Training on Speech Perception. *Frontiers in Psychology*, 2, 126. https://doi.org/10.3389/fpsyg.2011.00126
- Sharma, A., Dorman, M., Spahr, A., & Todd, N. W. (2002). Early cochlear implantation in children allows normal development of central auditory pathways. *Annals of Otology, Rhinology, & Laryngology - Supplement, 189*, 38-41.
- Sharma, A., Dorman, M. F., & Spahr, A. J. (2002). Rapid development of cortical auditory evoked potentials after early cochlear implantation. *NeuroReport, 13*(10), 1365-1368.
 Sharma, A., Martin, K., Roland, P., Bauer, P., Sweeney, M. H., Gilley, P., & Dorman, M. (2005). P1 latency as a biomarker for central auditory development in children with hearing impairment. *Journal of the American Academy of Audiology, 16*(8), 564-573.
 Sharma, A., Tobey, E., Dorman, M., Bharadwaj, S., Martin, K., Gilley, P., & Kunkel, F. (2004). Central auditory maturation and babbling development in infants with cochlear implants. *Archives of Otolaryngology–Head & Neck Surgery, 130*(5), 511-516. https://doi.org/10.1001/archotol.130.5.511
- Sharma, M., Purdy, S. C., & Kelly, A. S. (2009). Comorbidity of auditory processing, language, and reading disorders. *Journal of Speech Language and Hearing Research*, 52(3), 706-722. <u>https://doi.org/</u>10.1044/1092-4388(2008/07-0226)
- Shepherd, R. K., & Hardie, N. A. (2001). Deafness-induced changes in the auditory pathway: implications for cochlear implants. *Audiology and Neuro-Otology*, 6(6), 305-318. <u>https://doi.org/10.1159/000046843</u>

- Shepherd, R. K., Hartmann, R., Heid, S., Hardie, N., & Klinke, R. (1997). The central auditory system and auditory deprivation: experience with cochlear implants in the congenitally deaf. *Acta Oto-Laryngologica Supplement, 532*, 28-33. Skinner, M. W. (2003).
 Optimizing cochlear implant speech performance. *Annals of Otology, Rhinology, and Laryngology, 191*, 4-13. <u>https://doi.org/10.1177/00034894031120S903</u>
- Skinner, M. W., Holden, L. K., Whitford, L. A., Plant, K. L., Psarros, C., & Holden, T. A. (2002). Speech recognition with the nucleus 24 SPEAK, ACE, and CIS speech coding strategies in newly implanted adults. *Ear and Hearing*, 23(3), 207-223.
- Stecker, G. C., Bowman, G. A., Yund, E. W., Herron, T. J., Roup, C. M., & Woods, D. L. (2006). Perceptual training improves syllable identification in new and experienced hearing aid users. *Journal of Rehabilitation Research and Development*, 43(4), 537-552.
- Sweetow, R. W., & Palmer, C. V. (2005). Efficacy of individual auditory training in adults: a systematic review of the evidence. *Journal of the American Academy of Audiology*, 16(7), 494-504.
- Sweetow, R. W., & Sabes, J. H. (2006). The need for and development of an adaptive Listening and Communication Enhancement (LACE) Program. *Journal of the American Academy of Audiology*, *17*(8), 538-558.
- Sweetow, R. W., & Sabes, J. H. (2007). Technologic advances in aural rehabilitation: applications and innovative methods of service delivery. *Trends in Amplification*, 11(2), 101-111. <u>https://doi.org/0.1177/1084713807301321</u>
- Tremblay, K., Kraus, N., McGee, T., Ponton, C., & Otis, B. (2001). Central auditory plasticity: changes in the N1-P2 complex after speech-sound training. *Ear and Hearing*, 22(2), 79-90. Tremblay, K., Ross, B., Inoue, K., McClannahan, K., & Collet, G. (2014). Is the

auditory evoked P2 response a biomarker of learning? *Frontiers in Systems Neuroscience*, 8, 28. <u>https://doi.org/10.3389/fnsys.2014.00028h</u>

- Tremblay, K. L., Inoue, K., McClannahan, K., & Ross, B. (2010). Repeated Stimulus Exposure Alters the Way Sound Is Encoded in the Human Brain. *Plos One*, *5*(4), e10283. https://doi.org/10.1371/journal.pone.0010283
- Tremblay, K. L., Shahin, A. J., Picton, T., & Ross, B. (2009). Auditory training alters the physiological detection of stimulus-specific cues in humans. *Clininial Neurophysiology*, *120*(1), 128-135. <u>https://doi.org/10.1016/j.clinph.2008.10.005</u>
- Tye-Murray, N., Sommers, M. S., Mauze, E., Schroy, C., Barcroft, J., & Spehar, B. (2012).
 Using patient perceptions of relative benefit and enjoyment to assess auditory training. *Journal of the American Academy of Audiology*, 23(8), 623-634.
 https://doi.org/10.3766/jaaa.23.8.7
- Ventry, I.M., & Weinstein, B.E. (1982). The Hearing Handicap Inventory for the Elderly: A New Tool. *Ear and Hearing*, 3, 128–134.
- Vongpaisal, T., Caruso, D., & Yuan, Z. (2016). Dance Movements Enhance Song Learning in Deaf Children with Cochlear Implants. *Frontiers in Psychology*, 7, 835. https://doi.org/10.3389/fpsyg.2016.00835
- Welch, G.F., Saunders, J., Papageorgi, I., & Himonides, E. (2012). Sex, gender and singing development: Making a positive difference to boys' singing through a national programme in England. In S. Harrison, G.F. Welch, & A. Adler (Eds.), *Perspectives on Males and Singing* (pp37-54). London: Springer.

- Welch, G. F., Saunders, J., Edwards, S., Palmer, Z., Himonides, E., Knight, J., . . . Vickers, D. A. (2015). Using singing to nurture children's hearing? A pilot study. *Cochlear Implants International, 16 Suppl 3*, S63-70. <u>https://doi.org/10.1179/1467010015z.00000000276</u>
- Wilson, B. S., Finley, C. C., Farmer, J. C., Jr., Lawson, D. T., Weber, B. A., Wolford, R. D., ... Schindler, R. A. (1988). Comparative studies of speech processing strategies for cochlear implants. *Laryngoscope*, 98(10), 1069-1077. <u>https://doi.org/10.1288/00005537-198810000-00009</u>
- Wu, J. L., Yang, H. M., Lin, Y. H., & Fu, Q. J. (2007). Effects of computer-assisted speech training on Mandarin-speaking hearing-impaired children. *Audiology and Neuro-Otology*, 12(5), 307-312. <u>https://doi.org/10.1159/000103211</u>
- Yucel, E., Sennaroglu, G., & Belgin, E. (2009). The family oriented musical training for children with cochlear implants: speech and musical perception results of two year follow-up. *International Journal of Pediatric Otorhinolaryngology*, *73*(7), 1043-1052. https://doi.org/10.1016/j.ijporl.2009.04.009
- Zhang, T., Dorman, M. F., Fu, Q. J., & Spahr, A. J. (2012). Auditory training in patients with unilateral cochlear implant and contralateral acoustic stimulation. *Ear and Hearing*, 33(6), e70-79. <u>https://doi.org/10.1097/AUD.0b013e318259e5dd</u>
- Zhou, Y., Chai Sim, K., Tan, P., & Wang, Y. (2012). MOGAT: Mobile games with auditory training for children with cochlear implants. MM 2012 - Proceedings of the 20th ACM International Conference on Multimedia. 429-438.

https://doi.org/10.1145/2393347.2393409.

Zimmerman-Phillips, S., Osberger, M. J., & Robbins, A. M. (2001). Sylmar, CA Patent No. Advanced Bionics Corporation.

APPENDIX A

Research Questions

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?
- o 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?

APPENDIX B

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 posttraining 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 C

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. Uncertain

Study		Particip ants		Training					Findings		
Authors	Design	N	Age	Stimuli	Skills trained	Frequency and duration	Place of training	Outcome measures	Improved?	Retention	Generalizat ion
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	3-7у	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	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 	Yes	Not assessed	Not Assessed

				drumbeats, speech sounds, syllables, and phonemes; blending words, syllable				Language Scales (OWLS) (Carrow- Woolfolk, 2008)			
Kronenberge r, 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 WM 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 noise. (Angel Sound)	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); Digit triplets in speech shaped noise (untrained) 	Yes	Yes for up to 3 weeks	near transfer but not far transfer
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 instrume nt; in clinic/ Lab	- Same as training stimuli but different sets used only as outcome measures	Yes in all except ASA	Yes	Yes (Phoneme Discriminat ion)
Welch et al. (2015)	Non-RCT	12 9CI/3H A)/17N H	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,	Repeated	7CI/3H	5-11	Discrimination task,	Identification	30 min per 1	PC based	- Vowel and	Yes	yes for 2	Not
Lin, and Fu (2007)	measure	A	yrs	trained to identify final vowels. Discriminating between phonemes. For vowels, acoustic speech features included (F 1 and F 2) and duration;	and discrimination of speech sound	session 5 days a week for 10 weeks	in home	consonants discrimination - Chinese tone recognition		months	assessed
Yucel, Sennaroglu, and Belgin (2009)	Non-RCT	9 CI EG /9 CI CG	36-96 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 Cl 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 1 – Description of study details

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- 	Yes	Not assessed	Not Assessed	Not explicitly reported but can be deduced 1

Table 2 – information with r	respect to assessment criteria
------------------------------	--------------------------------

	Woolfolk, 2008)				
Kronenberger et al. (2011)	 Digits forward and backward Spatial span forward and backward; BRIEF: Sentence repetition 	Yes	yes (all WM 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	Yes (only	Not	No transfer to	Not reported
	questionnaire.	trained)	assessed	speech	
	- MAIS and MUSS				
	- Phonetic discrimination,				
	- Word identification,				
	- Comprehension of simple				
	auditory instructions, and				
	- Sentence repetition				