Music Perception and Appreciation in Trained Cochlear Implant Users: A Systematic Review

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Abstract

A cochlear implant is a device designed to improve the hearing thresholds of those with severe-profound hearing loss. Consisting of a microphone, signal processing unit, magnetically-coupled radio-frequency transducer and electrode array a cochlear implant is able to provide electrical stimulation in the cochlea to stimulate nerves in responses to sound. Threshold (T) levels and comfort (C) levels are obtained in order to identify appropriate stimulation levels for quiet and loud sounds. Verification for cochlear implants is achieved using behavioural and subjective testing as well as objective testing. As technology in cochlear implants has developed to enable provision of access to more than just speech, it has become more advantageous and appropriate to assess users with environmental (non-speech) sounds. Modern cochlear implants are able to process and deliver usable stimuli in response to a wide range of non-speech sounds, and among the most important of these is music. Music is a complex organisation of aural stimuli often intended to provoke emotion. Music is a core human experience (Schulkin & Raglan, 2014). Music can demonstrate near-limitless variation in rhythm, pitch, timbre, loudness, texture, duration, and localisation. The ability to process this combination of sonic properties can be thought of as one's ability to perceive music, and it is this perceptual ability which is often affected by hearing impairment and, in particular, the auditory stimulation provided by cochlear implants. The ability to enjoy music can be thought of as one's ability to appreciate music. In this thesis, a systematic review is justified in order to evaluate the field of study involved in assessing the effect of training on music perception and appreciation in cochlear implant users. Three studies were included by way of online search with strict inclusion and exclusion criteria. The studies found significant potential benefit for musical training in CI users. Future research is justified considering the relatively small number of high-quality studies available and the lack of diversity in populations involved. Audiological professionals are advised to continually educate

themselves regarding optimal CI surgery techniques, amplification paradigms, musical properties and training programs for music perception and appreciation in order to provide the best outcomes for CI patients.

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SNHL	– Sensorineural Hearing Loss
CHL	– Conductive Hearing Loss
WHO	– World Health Organisation
HI	– Hearing Impairment
CI	– Cochlear Implant
BTE	– Behind-the-Ear Hearing Aid
HA	– Hearing Aid
dB	– Decibel
dBHL	– Decibel Hearing Level
dBSPL	– Decibel Sound Pressure Level
HHI	–Hearing Handicap Inventory
MCI	– Melodic Contour Identification
VEI	– Vocal Emotion Identification
EPP	– Emotional Prosody Perception
IPP	– Inter-professional Practice
NCIQ	– Nijmegen Cochlear Implant Questionnaire
MBEMA	– Montreal Battery

1. Hearing impairment

It was estimated that in 2015 in New Zealand there were 330,269 people aged ≥14 with hearing impairment (HI) and that number was expected to rise to around 449,453 by 2061 (Exeter, Wu, Lee, & Searchfield, 2015). According to the World Health Organisation (2020) greater than 6.1% of the world's population is affected by a disabling HI. This means an estimated 432 million adults are affected by a HI greater than 40dB in the better hearing ear while and estimated 34 million children are affected by a HI greater than 30dB in the better hearing ear (World Health Organisation, 2020). A 1999 study in the South Australian population estimated that 17% of those aged ≥15 had a HI of ≤25dBHL, gradually decreasing in prevalence as severity increased, such that 0.5% of the population had a HI of ≤65dBHL (Wilson et al., 1999).

1.1 The Impact of Hearing Impairment

Hearing impairment is a disability with largely varying impacts that can be described using the World Health Organisation's International Classification of Functioning, Disability and Health (ICF) model (World Health, 2001). The ICF model consists of simplifying factors of a disability by describing the health condition, body functions and structures affected, activities disrupted, participation disrupted, environmental factors, personal factors and third-party disability. An example of the ICF model in the context of hearing impairment is shown in Figure 1 below.

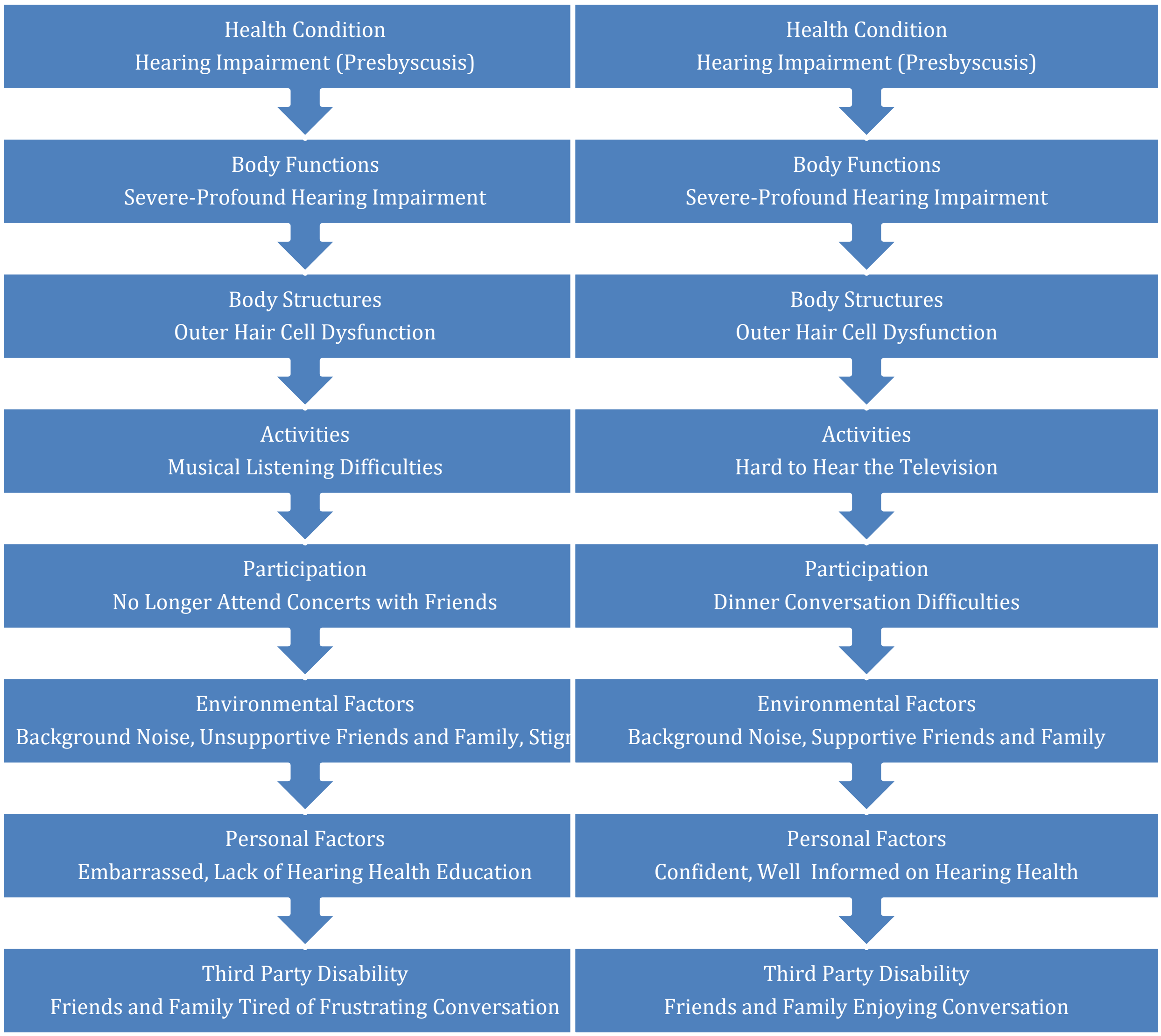


Figure 1: THE IMPACT OF HEARING IMPAIRMENT USING THE ICF MODEL

In the example, two people have been diagnosed with presbycusis due to outer hair cell dysfunction. A severe-profound SNHL of the same configuration is affecting both of them, however due to differences in their activities, participation, environmental and personal factors, the person on the right is able to live with the hearing impairment much more positively and sustainably when compared to the other. It is obvious then, that considerations for variability in clients, even with identical hearing losses, may result in varying degrees of rehabilitation considerations. For the first person we may consider educating the client as well as friends and family about the prevalence of hearing loss and the negative effects of stigma as well as fitting hearing aids and considering cochlear implantation. For the second person (again, with the same hearing loss) we may encourage the positive outlook on hearing impairment which they have already expressed and monitor the hearing impairment as well as considering hearing aids and cochlear implantation. Candidacy for assistive listening devices such as hearing aids consists of three main considerations. Firstly, the presence of a hearing impairment. Secondly, the impact of the hearing impairment on everyday function. Finally, the willingness of the patient to improve their hearing capabilities or lessen the negative impact of the hearing impairment. For viability of rehabilitation using this method, all three must be present. Some of the impacts of hearing impairment are apparent in the example above, however there are many more including but not limited to social difficulties, communication barriers, safety hazards due to lowered awareness, isolation, depression, anxiety, short-temperedness, acceleration of cognitive decline, speech production difficulties, lowered quality of life, otalgia (ear pain), learning difficulties, bullying and unequal opportunities.

1.2 Hearing Impairment Aetiology

It is vital that, with a growing prevalence of HI, research into rehabilitation progresses at an appropriate pace in order to provide the best outcomes for those with HI. HI can be acquired or congenital (from birth). The common causes of hearing loss are shown in Table 1 (adapted from (Eggermont, 2017).

Table 1: COMMON CAUSES OF HEARING LOSS

Acquired	**Congenital**
• Occupational and recreational noise induced hearing loss (NIHL) • Presbycusis (Age related) • Trauma • Meniere's disease • Tympanosclerosis • Ototoxicity • Bacterial infection • Otosclerosis (some genetic factors) • Viral infection • Diabetes • Vestibular Schwannoma	• Structural abnormality such as enlarged vestibular aqueduct syndrome (EVAS), microtia, or atresia • Non-syndromic causes (e.g. autosomal dominant, autosomal recessive, or mitochondrial disorders) • Syndromic causes (e.g. Alport syndrome, Branchio-Oto-Renal syndrome, CHARGE syndrome, Jervell & Lange-Nielsen syndrome, Pendred syndrome, Perrault syndrome, Stickler syndrome, Treacher-Collins syndrome, Usher syndrome, Waardenburg syndrome) • Auditory neuropathy spectrum disorder • TORCH (i.e. Toxoplasmosis, Other (Syphilis, Varicella Zoster, Parvovirus B19), Rubella, Cytomegalovirus (CMV), Herpes)

While public perception can sometimes be that a hearing impairment can be instantly solved by the fitting of a hearing aid, it is far more complex than that. The spectrum of audiological rehabilitation spans from education and auditory training, to assistive listening devices and hearing aids, and all the way to cochlear or auditory brainstem implantation. With such a wide spectrum, extensive assessment must be conducted in order to provide optimal care. Factors which are considered in the selection of rehabilitation include but are not limited to configuration and severity of hearing impairment, aetiology, funding, environmental factors and personal factors. If the rehabilitation method chosen is not appropriate for the individual, we will see less benefit from the rehabilitation. These factors and their impact on rehabilitation are particularly relevant for CI users. When considering CI candidacy, diagnosis of the specific site of lesion becomes important. An example of this is the differences in site of lesion between auditory neuropathy spectrum disorder (ANSD) and other retrocochlear pathologies such as vestibular schwannoma or auditory nerve dysfunction. A cochlear implant will be able to provide amplification for someone with ANSD if the site of lesion is at the inner hair cells. A cochlear implant will not be able to provide adequate amplification for someone with vestibular schwannoma, auditory nerve dysfunction, or brainstem ANSD, as the site of lesion is further up the auditory chain than the site of amplification. In order to assess the validity of cochlear implantation a CT scan or MRI is essential.

2. Cochlear implants

2.1 Fundamentals

Cochlear implantation allows people with a HI to perceive sounds through acoustic-digital transduction at a level that hearing aids can be incapable of providing with acoustic-digital-acoustic transduction. Stimulating the auditory nerve directly, a CI attempts to replicate/replace the function of the cochlea by transducing sound from an acoustic input signal (sound) to an electrical output signal. This eliminates the necessity for normal function of the impedance matching system in the middle ear as well as normal function of the inner ear. A cochlear device consists of multiple components. The external component is fitted behind the ear and looks similar to a behind-the-ear (BTE) hearing aid. Consisting of a microphone, amplifier, and sound processor, this component is responsible for tailoring the intensity and frequency of a given input signal to an individual’s needs before it is sent to the internal component. The internal component consists of a coil surgically implanted under the skin on the temporal bone through which the signal can be transduced via induction. The internal component also consists of an electrode array which is surgically threaded along the length of the basilar membrane typically with the intention of residing in scala tympani (a chamber in the cochlea) in order to directly stimulate the spiral ganglion (sensory nerves) located throughout the modeolus therefore eliminating the need for normal middle ear and cochlear function for hearing.

2.2 Cochlear Implant Candidacy

An audit of 17 countries with 28 respondents showed that funding was available for unilateral cochlear implantation in 60% of countries, with the other 40% of funding coming from medical insurance or self-funding. The most common candidacy measure used was audiometric results using thresholds of 75-85dB above 1Khz as the cut-off point. It has been suggested this is too strict and a four-tone average (FTA) of 500Hz, 1kHz, 2kHz and 4kHz should be used with a cut-off of 70dBHL FTA as success has been shown for implanted subjects when compared to hearing aid subjects (Leigh, Dettman, Dowell, & Sarant, 2011).

A systematic review of paediatric cochlear implant candidacy showed that patients with profound hearing impairment experienced similar outcomes to those with severe hearing impairment, supporting the relaxation of candidacy criteria based on pure-tone thresholds alone (de Kleijn et al., 2018). In cases where some natural hearing is present, CI surgery will only be considered after first attempting to rehabilitate a patient using hearing aids. It is obvious that, if surgery can be avoided it should be for safety of the patient as well as preservation of normal cochlear function if possible.

Areas of candidacy which are becoming more accepted include those with asymmetric losses. Self-funded cochlear implants tend to have more relaxed candidacy criteria (Vickers, De Raeve, & Graham, 2016). Pre-implantation neurocognitive measures such as working memory capacity, inhibition-concentration, information processing speed, and nonverbal reasoning can have an impact on post-implantation sentence recognition, particularly degraded speech signals, and therefore should be considered when determining CI candidacy (Moberly, Castellanos, & Mattingly, 2018).

Interaural time differences are the term given to the miniscule delay in hearing a sound in one ear when compared to the other. Interaural level differences is the term given to the sound level differences in hearing a sound in one ear when compared to the other. The brain is trained to recognise these phenomenons, allowing an individual to localise sound, heightening awareness of the environment. In order to localise sound (identify the direction of sound source), achieve summative loudness, make use of the head shadow effect, optimise hearing in noisy situations (binaural squelch) and avoid the “unaided ear effect” (neural degradation due to lack of stimulation of the auditory nerve pathway) it is vital that those with a HI can be provided with hearing in both ears. This involves fitting those with unilateral HI as well as encouraging those with bilateral HI to use two hearing aids instead of one despite the potential cost.

Trends in cochlear implant candidacy include the increasing prevalence of bimodal rehabilitation; utilising both cochlear implant and hearing aid technology in unison. This is most common in those with an asymmetrical HI in which the acoustic device compensates for the relatively poor low frequency stimulation capabilities of a CI by providing amplification for the contralateral ear.

A relatively early pilot study of children well experienced with CI use, aged seven to 15 concluded that when educating parents about unilateral CI use for their child, bimodal amplification should be discussed as use of a contralateral hearing aid can improve speech-in-noise perception and other potential benefits (Ullauri, Crofts, Wilson, & Titley, 2007).

An early systematic review concluded that methodological limitations resulted in inconclusive evidence when comparing bilateral CI implantation and bimodal amplification, regardless however, bilateral amplification in any mode outperforms unilateral amplification when correctly fitted and verified (Ching, Massie, Van Wanrooy, Rushbrooke, & Psarros, 2009).

A more recent retrospective study of experienced CI users showed a significant decrease in hearing impairment and improvement in quality of life in those who were provided with contralateral acoustic amplification (Sanhueza, Manrique-Huarte, Calavia, Huarte, & Manrique, 2019).

With increasing supporting research as well as many resources on which to rely for accurate and optimal fitting it is obvious why audiologists and otolaryngologists are trending toward bimodal amplification for their patients.

It is important to note that, post-cochlear-implantation, the hearing experience of the recipient will be vastly different to that of a person with normal hearing. This is due to the limitations of the device. In an ideal world, a cochlear implant would be capable of providing stimulation along the length of the entire basilar membrane allowing for hearing at frequencies the same as someone with normal hearing (roughly 20Hz-20kHz) while replicating the same amplification, frequency sensitivity and compression qualities present in the hearing system of someone with normal hearing.

Although modern cochlear implants are greatly advanced when compared to the first model of the device which was designed only to notify the recipient if a sound was present or not (with no frequency content or loudness differences), there are many limitations.

Those who are fitted with cochlear implant devices later in life as well as those who have unilateral HI with normal hearing on the contralateral side have the best perspective in comparing natural hearing with cochlear implant mediated hearing. Often CI users explain a "robotic" sensation of hearing owing to the relatively low frequency specificity of the CI electrode array and a lack of "bass" or "tinny" soundscape owing to limitations of the extent of electrode array insertion. It is difficult to accurately replicate the hearing experience of a CI user in order for someone with normal hearing to understand as there are too many variables to contend with. Because of this, subjective assessments are vital to CI success.

2.3 Coding Strategy

In the same way a hearing aid is fitted according to a prescription in order to provide optimal amplification for an average person with a given HI, cochlear implants utilise different sound processing formulae known as coding strategies to achieve the same goal. Temporal coding and the tonotopic organisation of the cochlea (colloquially known as volley and place) are psychophysical concepts which explain the capability of an individual with normal hearing to identify and discern sounds according to their frequency content. Temporal coding alone is limited by the capacity of the auditory nerve and its ability to fire at a given rate. In order to stimulate frequencies ≥1kHz for a CI user, multiple electrodes are required to engage, making use of the tonotopic organisation of the cochlea. Electrodes are fired either simultaneously or individually, as well as varying in firing rate and amplitude depending on the coding strategy employed. The BTE component of the CI processes analog sound using one of a number of common strategies aimed at speech encoding. Early strategies known as multi-peak (MPEAK) and spectral-peak (SPEAK) have given way to continuous-interleaved sampling (CIS; (Somek, Fajt, Dembitz, Ivković, & Ostojić, 2006) or the advanced combination encoder (ACE; (Holden, Vandali, Skinner, Fourakis, & Holden, 2005). These encoding strategies are optimised for speech understanding and have the potential to alter the input signal greatly, resulting in hearing which is vastly different to someone with normal hearing or someone who uses hearing aids. This is where complex environmental signals such as music may require training for the listener to better tolerate or make use of their hearing through a CI.

2.4 Surgery

Cochlear implants typically extend through the cochlea at a length of 1.25 turns. Anatomically the average length of a human cochlea is typically 2.5-2.75 turns. This means that, in theory, extremely low frequency stimulation is difficult to attain given the tonotopic organisation of the cochlea. More recent evidence shows that longer electrode arrays are capable of reaching closer to the apex of the cochlea resulting in more stimulation of neurons in the area responsible for hearing low frequency sounds (Roy, et al., 2016).

A review of the literature concluded that there is some support for the ability of longer electrodes to stimulate broader frequency regions along the basilar membrane however evidence for translation into real-world benefits such as music perception and appreciation is limited (Boyd, 2011). So-called “soft surgery” aimed at the preservation of residual hearing refers to the efforts of surgical staff such as otolaryngologists to attempt to conduct CI procedures in such a way that any remaining natural hearing a patient may have will still be present post-implantation. The preservation of residual hearing in these cases results in the patient hearing digitally through a CI while retaining some natural acoustic hearing, typically in the low frequencies.

Factors considered for soft surgery include but are not limited to CI design/technology, surgical approach, surgical trauma, steroid administration and multifarious patient factors (Bruce & Todt, 2018). A systematic review of hearing preservation literature found that soft surgery techniques are feasible noting that a round window insertion approach and straight electrode array may be preferrable to a cochleostomy approach with a perimodiolar electrode array (Snels, IntHout, Mylanus, Huinck, & Dhooge, 2019). According to the review, the literature regarding soft surgery currently has many limitations and, in order to evolve and progress the field of study, prospective studies comparing hearing preservation techniques are required.

A literature review of paediatric CI users showed that 65% of recipients with conventional electrode arrays retained functional low frequency natural hearing in their first accessible post-operative audiogram and 82% of recipients retained detectable hearing thresholds at any frequency (Carlson et al., 2017).

CI candidacy is impacted by the trend toward hearing preservation, as concerns for destruction of natural hearing are lessened, implantation is no longer reserved for those who are profoundly deaf (Boisvert, Reis, Au, Cowan, & Dowell, 2020). Although it is now possible to preserve residual hearing in some cases, results are variable due to many factors (Moran, Dowell, Iseli, & Briggs, 2017), including the patient's initial hearing thresholds (pre-implantation), the type of electrode implanted (perimodiolar, lateral wall or mid-scala), and surgical procedure (cochleostomy, round-window or extended round window) (Wanna et al., 2018).

A recent systematic review of hearing preservation benefits concluded that there is support (although limited) for the efforts of audiological and otolaryngological professionals to preserve patient hearing, stating that there are real-life benefits for patients. Variance in assessors and timeframes resulted in great difficulty accurately comparing the interventions of interest (hearing preservation techniques), promoting the need for interprofessional teams to make efforts to standardise methodologies for future comparisons. As the field advances, guidelines for optimal hearing preservation techniques should arise and be tailored to individual patient needs and variables in order to optimise patient outcomes (Schaefer, Sahwan, Metryka, Kluk, & Bruce, 2021).

3. Musical Concepts

The definition of music is broad and ambiguous. It has been described as a universal human competence, a necessary and integral dimension of human development and, like speech, a product of our biology and social interactions (Cross, 2001). Music is important to multiple age groups. It allows individuals to express themselves, satisfy their emotional needs, and maintain quality of life (North, Hargreaves, & O'Neill, 2000). The experience of listening to music can enhance connectivity between brain regions, indicating provocation of cognitive function (Menon & Levitin, 2005). This is particularly important in people with hearing impairment as progression of cognitive decline has been associated with hearing impairment (Dawes et al., 2015).

Music can be considered a main feature of social gatherings such as concert-going and at house parties, or as an accompaniment in many circumstances such as watching television, driving in the car, or riding in an elevator. The ability to hear and enjoy the full frequency range of music in these circumstances has been shown to increase positivity, alertness and focus (Sloboda, O'Neill, & Ivaldi, 2001).

Music and speech differ greatly in their acoustic characteristics. Typically, speech, even when shouted, rarely exceeds 85 dB SPL whereas music can reach levels of 100-110 dB SPL regardless of genre (Chasin, 2003). Assistive listening devices optimised at sound levels for typical speech will perform poorly with live music because of this. These levels exceed the safe listening level (stated in Regulation 11 of the Health and Safety in Employment Regulations 1995 in NZ) of a maximum of 8 hours continuous listening per day at a level of 85 dB A (Department of Labour (2002)). Every 3 dB step above this level requires a halving of listening time (e.g. 88 dB A should only be listened to for 4 hours maximum per day in order to protect from NIHL). A bitter irony exists in the notion that participating in music can cause hearing impairment which in turn limits our ability to participate in music.

Compression and limiting within hearing devices allow the listener to be in these environments without too much discomfort, but can distort the sound heavily, creating a poor listening experience for the user. The important frequency spectrum for speech understanding is considered by audiologists to be between 250 Hz and 8 kHz (Katz, Chasin, English, Hood, & Tillery, 2015). In order to enjoy and hear the full harmonic range of certain instruments, normal hearing or minimal hearing loss at frequencies > 8 kHz is required. It is obvious then that people who have hearing loss tend to experience music differently to those with normal hearing. Emotional intent, one of the vital aspects of music, is often lost in translation between the composer and someone who has hearing loss, due to misinterpretations and difficulties in differentiating pitch, timbre, texture, and rhythm (Darrow, 2006). Those with severe to profound hearing loss rely on elements such as visual music accompaniment, vibrotactile music (sound vibrations which can be felt) and atmosphere (social gathering) in order to find enjoyment and appreciation in music more so than a person with normal hearing.

Those who are fitted with a cochlear implant experience music much differently to those with normal hearing. This distortion is thought to be a causing factor for low music enjoyment and participation levels post-implantation. The extent of literature in the field of music appreciation and perception in CI users is extremely low relative to the importance the public places on music's role in quality of life (Jiam, Caldwell, & Limb, 2017).

A piece of music can be separated into many variable contributing factors including but not limited to pitch (melody, harmony, key, mode, progression, tonality & atonality), rhythm (tempo & syncopation), timbre (instrument, range & wave-type), loudness, vibrotactile, visual accompaniment, emotion (key, mode & vocal expression), lyrics, dynamics (reduced by compression), duration, texture (reverberation, panning & spreading), genre and structure. Many of these factors rely heavily on recognition and discrimination of varying sound types, giving those with hearing impairment varying degrees of difficulty in attaining desired music perception and appreciation.

4. Assessment of Musical Ability of Cochlear Implant Users

Traditional measures of success for CI recipients include specific frequency thresholds (pure-tone audiometry), speech-recognition testing and subjective questionnaires such as the Hearing Handicap Inventory (HHI). Understanding the importance of the musical listening experience allows us to see the value in assessing music perception and appreciation post cochlear implantation. Audiological professionals are increasing assessment of non-speech environmental sounds, and as such, novel assessments of CI success are emerging, particularly those taking into account music perception and appreciation. Perception is the ability of an individual to become aware of a given input using their senses. In the case of music perception, we are referring to the awareness of musical content described above. Assessors of basic musical perception such as pitch and timbre recognition are simple to administer and are often the focus of audiological efforts however more complex features of music are difficult to measure owing to underdeveloped assessors, technological limitations and absence of awareness/understanding. Music appreciation is related to the experience and emotional & social response to music (Looi, Gfeller, & Driscoll, 2012) and is generally assessed using qualitative measures.

5. Findings of Historic Reviews

In order to understand the breadth of the reviews that have been conducted to date on the topics of music perception and appreciation assessment in CI users. This section will provide a brief synopsis of a number of studies, and summarises the themes that were common to multiple reviews.

In 2004 a review was undertaken by Hugh J. McDermott entitled *Music Perception with Cochlear Implants: A Review* in which seven significant findings from past research on the topic were discussed (McDermott, 2004). Rhythm was proven to be perceived well by cochlear implant recipients. Melody recognition was poor in cochlear implant recipients, and timbre perception was unsatisfactory. The pleasure of listening to music was rated lower for cochlear implant recipients than for those with normal hearing. Training programs were shown to improve the acceptance of music for cochlear implant recipients. It was also found that pitch perception may be improved by development of cochlear implant sound processors for recipients.

Perception of music was likely to be better for cochlear implant recipients who had residual acoustic sensitivity, particularly in the lower frequencies.

A review entitled *Cochlear Implant-Mediated Perception of Music* (Limb, 2006) aimed to compile literature regarding recent advances in CI sound processing at the time. The findings showed that assessing specific features of music which CI users tend to have difficulty with such as rhythm, melody and timbre is useful. Assessing CI users in this way did not re-create the experience of listening to music in real-world situations. Music was described as one of the most complex acoustic stimuli able to be perceived by humans and may be the pinnacle of listening available to CI users if achieved. Cochlear implant surgeons were urged to understand the importance of achieving complete and atraumatic electrode array insertion when possible, maximising the range of absolute frequencies and therefore musical pitches to be heard. CI candidates and recipients were encouraged to be well informed that music perception is poor in recipients (at the time of the study), although improvements were being actively pursued. CI-mediated music perception could be vastly improved with electrical-to-cochlear pitch mapping, pitch processing strategies particularly inter-electrode stimulation and preservation of natural hearing *(Limb, 2006).*

A review of speech and language training for CI users entitled *Perceptual Learning and Auditory Training in Cochlear Implant Recipients* (Fu & Galvin, 2007) showed that high importance should be placed on developing protocols for aural training in order to provide efficient and effective aural rehabilitation for CI users. Stimuli used to train CI users may not have been variable enough to produce a general improvement for CI users, and it was found that the improvement from training varied greatly between participants. Objective neurophysiological measures were suggested in developing more efficient training while development of subjective measures was suggested to allow for assessment of real-world benefit from aural training. Aural training was described as relatively cost effective when compared to the cost of the device, and auditory training, when paired with speech processor optimisation, yielded dramatic gains in speech understanding. Improvements in both speech understanding and music perception were seen when inexpensive and effective auditory training was provided for CI users (Fu & Galvin, 2007).

A review entitled *Experience-Induced Malleability in Neural Coding of Pitch, Timbre And Timing* (Kraus, Skoe, Parbery-Clark, & Ashley, 2009) showed that subcortical auditory processes can be altered and trained. Auditory processing interacted with other sensory processing and varied with language and musical experience. It was suggested that processing early in the human auditory system is shaped by music and language experience likely due to efferent fibres (top-down influence). Auditory processing, multi-sensory integration and apparent cognitive-sensory reciprocity were factors with potential to be utilised by those with an impaired auditory system for sensory learning (Kraus et al., 2009).

In 2012 a review was undertaken by Looi, Gfeller and Driscoll entitled *Music Appreciation and Training for Cochlear Implant Recipients: A Review* which focused on the effect of musical training on cochlear implant users (Looi et al., 2012). This review shares the scope of this thesis but used a less stringent screening protocol. Their findings indicated that music enjoyment and appraisal is a vital measurement in considering outcomes for cochlear implant recipients, and that musical training can improve the listening experience and should be considered for all cochlear implant recipients.

A multi-disciplinary literature review entitled *Music Perception of Cochlear Implant Recipients with Implications for Music Instruction: A Review of Literature* (Hsiao & Gfeller, 2012) consolidated studies from the fields of audiology, speech-language pathology and music therapy revealed that great variance in music perception is apparent between individual CI users, emphasising the importance of individual assessment. With an individually tailored training methodology, children were proven to succeed in many forms of musical activity. It was expressed that it is vital for audiologists, the inter-professional practice team as well as family and friends to understand that the music perception limitations witnessed are representative of the technological constraints of the device, not through fault or weakness of the child. Ideal listening environments using quality equipment at moderate volume levels were shown to enhance music perception and enjoyment. Greater amounts of time spent listening to music and undergoing training had potential to improve many aspects of music perception. Visual cues and repetition of songs were shown to enhance participation and learning (Hsiao & Gfeller, 2012). In order to distinguish whether improvements post-training are attributable to genuine improved auditory perception or other factors such as cognitive processing, attention and memory a study was conducted with ten CI users. It was found that improvements were not solely attributed to the other factors meaning that a true improvement in auditory perception is witnessed after training (Oba, et al., 2013).

A review of cochlear implant technology entitled *Technological, biological, and acoustical constraints to music perception in cochlear implant users* (Limb & Roy, 2014) showed that multiple and diverse research streams are necessary for substantial improvements in music perception. CIs and coding strategies designed specifically for music and speech rather than speech alone should logically improve music perception. Music rehabilitation and training was described as a field requiring further development to improve music perception outcomes. Post-implantation training was shown to stimulate brain 're-organisation' therefore improving music perception. Music perception is defined as one of the highest forms of hearing a human can possess, not just entertainment. A lack of understanding of this notion was linked to a lack of training availability for CI users. Music perception assessments were promoted as a great tool for measuring the limitations of CI, again supporting the notion that assessment of music perception is recording the limits of the CI not the CI user (Limb & Roy, 2014).

A review entitled *Music-based training for pediatric CI recipients: A systematic analysis of published studies* (Gfeller, 2016) showed that only the first steps of research into music training for CI users had been completed. It was stated the field would greatly benefit from years of study undertaken in collaborative efforts by CI centres/programs in order to progress from its infancy. Music training was not promoted as superior to speech and language training, but the resulting perceptual improvements were described as encouraging. Factors that were encouraged to be considered in future trials include stimuli type, training format, training frequency, training duration, age (chronological, hearing and mental), hearing history, outcome interpretation, participant characteristics. Typical patterns seen in children with normal hearing were shown to aid in informing these factorial selections. Poor logistical planning in terms of finance, cooperation, facilities & equipment, adequate sample sizes and participant characteristics were factors warned to cause future trials to fail. Fine-tuning the auditory system with musical training had promise but needed more reviewing to determine how well these skills will be instilled in paediatric patients specifically. Training quality was reliant on valid, reliable and age-appropriate elements of music and speech, participation maintenance and identification of non-musical factors with potential to influence outcomes. Progression of research was described as potentially reliant on multidisciplinary collaborations (Gfeller, 2016).

A review of literature available in PubMed and Scopus databases entitled *Assessment and Improvement of Sound Quality in Cochlear Implant Users* (Caldwell, Jiam, & Limb, 2017) aimed to summarise information surrounding the experience of listening to complex sounds such as speech emotion and music for CI users. The findings showed that in the past decade the scope of CI research has been enlarged to include examination of more discrete features of sound such as pitch, amplitude and rhythm. Sound quality was described as essential in understanding auditory performance but the was a lack of research into utilizing its benefits. Perception of sound quality for CI users was limited by factors that were environmental, user specific and intrinsic to CI use, including pitch distortion and dynamic range compression. Objective, systematic and quantitative measures were shown to be lacking in the research. Promising strategies aimed at improving sound quality perception were in existence. It was suggested that development of measurement tools and rehabilitation strategies that include apical cochlear stimulation, place-pitch maps and noise reduction processing should be included in aural training (Caldwell et al., 2017).

A systematic review of music perception in adult (aged ≥18 years) cochlear implant users entitled *Music Appreciation after Cochlear Implantation in Adult Patients: A Systematic Review* (Riley, Ruhl, Camacho, & Tolisano, 2018) showed that CI users generally enjoy listening to music. Presence of temporal cues (such as those provided by percussive instruments) and music with a fast tempo increased the mentioned enjoyment. Simplifying the spectral content of music was found to improve enjoyment, meaning complex harmonics and dynamic ranges were less preferred. Music with vocals and linguistic cues was also preferred (Riley et al., 2018).

A systematic review entitled *Systematic Review of Auditory Training in Pediatric Cochlear Implant Recipients* (Rayes, Al-Malky, & Vickers, 2019) showed that auditory training improved the abilities of CI users in trained tasks in all nine studies which were reviewed. It was stated that the type of auditory training should be tailored to an individual however any training may be better than none. Benefit assessments were mainly conducted of the trained tasks and not untrained tasks which could assess generalised benefit. Only three of the studies in the review assessed benefit retention, which was then recommended for future studies. Lack of randomisation, a power calculation, and lack of blinding is common due to the population and constraints of the studies resulting in low to moderate study quality (Rayes et al., 2019).

Upon completing the literature review within this thesis, an extremely similar review was published. The well-conducted study entitled *Efficacy of Music Training in Hearing Aid and Cochlear Implant Users: A Systematic Review and Meta-Analysis* (Shukor, Lee, Seo, & Han, 2020) included both HA and CI users, which is a wider scope than the current review. Significant improvements in musical perception were observed with training. It was discovered that musical training may be more beneficial for children <18 years old. Patients using a CI only received more benefit from training than those using both a cochlear implant and hearing aid together (bimodal). No significant differences were found between the outcomes of those with previous musical experience and those without. A long training duration was recommended (>12 months) for better outcomes. Musical training was found to have potential in providing benefit in overall aural rehabilitation and speech-language development. Musical training was proven as an effective aural rehabilitation approach. It was stated that more randomised controlled trials (RCTs) are necessary to confirm the effectiveness of musical training in a field where RCTs are relatively scarce (Shukor et al., 2020).

In summary, music enjoyment is vital (Looi et al., 2012), and CI recipients enjoy music (Riley et al., 2018), particularly if it has simple spectral content and strong rhythm (Riley et al., 2018). There is, however, great variance in music perception among CI users (Hsiao & Gfeller, 2012). While preservation of residual hearing is important to facilitate music perception (McDermott, 2004; Limb, 2006), it is also amenable to music training (Hsiao & Gfeller, 2012; Gfeller, 2016; Shukor et al., 2020) and auditory training in general (Fu & Galvin, 2007, Rayes et al., 2019). Music training was also found to improve the experience of listening to music (McDermott, 2004; Looi et al., 2012). Aural rehabilitation and speech and language development was also improved by music training (Gfeller, 2016; Shukor et al., 2020), auditory training (Rayes et al., 2019), and simple exposure to music (Kraus et al., 2009). While aural training holds promise as a cost-effective intervention (Fu & Galvin, 2007), a general theme was that more research into its benefits was required (Limb & Roy, 2014; Gfeller, 2016), particularly that which is high quality (Gfeller, 2016; Shukor et al., 2020), and multidisciplinary (Hsiao & Gfeller, 2012, Gfeller, 2016). Such research could be aided by having better subjective and objective measures of music perception (Fu & Galvin, 2007; Caldwell et al., 2017).

6. Objective of Current Review

Many of the reviews mentioned above include bimodal hearing device users, specific populations (age) and low-quality studies. The current review is necessary in order to provide a more recent overview of the field particularly focused on the quality of the studies and unimodal CI users only. The population of interest is current cochlear implant users of any age. A study will be included if some/all participants use bimodal amplification so long as only the cochlear implant was used during testing or if results for CI users only are available separately. Interventions of interest can vary greatly and were assessed case by case, in general interventions were appropriate and shared the current review's focus on improving music perception and/or appreciation. Music perception and appreciation can be assessed in many different ways owing to the complexity of the concept. Only assessors relevant to music perception and appreciation were analysed, meaning typical assessors such as speech recognition were omitted.

The objective of this review is to investigate the following questions:

1. To what extent does training impact a CI users' ability to perceive and appreciate music?
2. What are the important contributing factors/influences on training success?
3. How can future studies be conducted in order to provide optimal outcomes for participants and advancement of the field?
4. How do the findings relate to previous studies and reviews?
5. How can the findings be used by professionals to improve rehabilitation for clients?

7. Review Protocol

7.1 Search

The current review protocol was assessed by a thesis supervisor and was not registered online. The University of Canterbury (UC) “multi-search” tool (based on “Summon”; Serials Solutions, Seattle, WA, USA) was the choice of search engine for the current review. This decision was made in order to access search results from a variety of databases such as PubMed and Scopus and filter results to an appropriate time period with ease. The search for the current study used the keywords “cochlear implant music training” and was conducted on 02/11/2020 at 11:30 am. Using the UC multi-search tool, results were refined in order to include studies only with the full text available online, exclude studies not published in English and exclude studies prior to January 2012. This date was chosen in order to create an appropriate scope for the current study taking into consideration previous reviews and technological advancement. Covidence (Covidence. Melbourne, Australia) is the name of the software used in order to screen and provide pilot forms by which to extract data from the studies and export figures and tables showing the process and results of the systematic review.

7.2 Screening

The current study used the eligibility criteria (inclusion & exclusion protocol) described in Table 2 in order to narrow the scope to an appropriate size.

Table 2*:* ELIGIBILITY CRITERIA FOR THE CURRENT SYSTEMATIC REVIEW.

Inclusion Protocol	**Exclusion Protocol**
• Population: CI users (unilateral or bilateral), any age, any country • Randomised Controlled Trial • Music training intervention • Conducted in January 2012 or later • Intervention effect estimate	• Opinion/anecdotal journals • Non-RCT studies • Bimodal amplification without ability to separate CI results • Full text unavailable online • English language version unavailable

The studies were uploaded to the Covidence software where both the reviewer and supervisor voted for or against each study to be included according to the criteria based on the abstract alone. Each study required two votes for "yes" in order to progress to the next stage of screening. Two votes "no" resulted in exclusion from the current review, while one vote "yes" and one vote "no", or any vote "maybe", resulted in a secondary round of screening in which the reviewer and supervisor discussed and agreed on reasons for inclusion or exclusion. The studies which were included after the abstract screening step were then assessed according to their full text in order to confirm their appropriateness for inclusion in the review, the same voting structure is used in this stage of screening on order to reveal the final cohort of studies to be included according to the protocol. 100 studies were imported from the search results as only a limited amount of relevant material was found form the preliminary assessment. The citation for each study was exported from the web search into EndNote X9 software (Clarivate Analytics, Philadelphia, PA, USA) for management and citation. The "search web for full text" function was used in order to attach PDF documents containing the full text to each reference. Full text files that were not found using this method were found by following the search link from the initial web search.

7.3 Data Extraction

We found several advantages and disadvantages to using the Covidence software for data extraction and synthesis. Covidence uses standardised online data extraction forms (pilot forms) which can be altered in order to best suit the study.

7.4 Quality Assessment

The first Covidence form is based on the Cochrane methodology for assessing risk of bias, this will be the basis of our quality assessment. Each potential bias was investigated in the studies and given a rating of low, high or unclear risk of bias. Incorrect procedures for participant selection, sequence generation and allocation concealment can all result in selection bias, meaning that participants are either unrepresentative of the target population or are aware of and can influence their intervention group allocation which can have a significant impact on performance. Incorrect or absent blinding of participants and personnel can lead to performance bias, this is when participants are aware of the intervention and outcome of interest enabling extreme risk for impacting results, the same bias can occur when personnel are unblinded. Incorrect or absent blinding of outcome assessments can lead to detection bias, if a participant is aware of the outcome of interest there is great potential for result alteration whether intentional or not. Incomplete outcome data can lead to attrition bias, and attrition must be well documented and accounted for in order to mitigate the potential risks associated. This can be simple or complicated depending on the prevalence and severity of attrition. Selective reporting can lead to reporting/publishing bias, this is when the author, usually unknowingly, influences the outcome of the study. This can occur for many reasons such as inexperience, time restrictions or something more sinister such as sponsorship pressure. Reporting/publishing bias is generally noticeable when the author does not strictly follow a protocol or does not report results correctly or fully. A study that has no bias present can be considered high-quality, a study containing minor biases can be considered average-quality and a study which has strong biases can be considered low-quality. An evidence base built from high-quality studies is considered robust, accurate and can be used to draw upon for best practice. An evidence base built of low-quality studies is considered weak, biased and should be carefully considered before drawing upon for best practice.

7.5 Analysis

The next stage of data extraction using Covidence software involved identification and reporting of methods, population, intervention and outcomes. The software provides blank tables (pilot forms) for each study which the user can customise and specify which data to extract. Generally, in a systematic review a PICO style extraction is the first step. PICO style extractions involve reporting the population, intervention, comparison and outcomes of a study (Eriksen & Frandsen, 2018). It is then important to identify other potentially significant data such as study design, publishing date, results and funding. The studies consisted of randomised controlled trials with one using a crossover design and the other two using a parallel group design. In order to best fit the scope of this study, only outcome measures relevant to music perception and appreciation were included.

7.6 Synthesis

Typically, a meta-analysis is performed in order to formally pool results and provide an accurate representation of the effectiveness of the interventions. In this case the study designs and interventions/assessors vary greatly, the sample size is small and raw data is unavailable making this process much more difficult. A more appropriate approach is a narrative synthesis in which the same concepts are explored through systematic reporting. A narrative synthesis involves developing a theory of how the intervention works, why and for whom; developing a preliminary synthesis of findings of included studies, exploring relationships within and between studies and assessing the robustness of the synthesis (Akers, Aguiar-Ibáñez, & Baba-Akbari, 2009).

8. Results

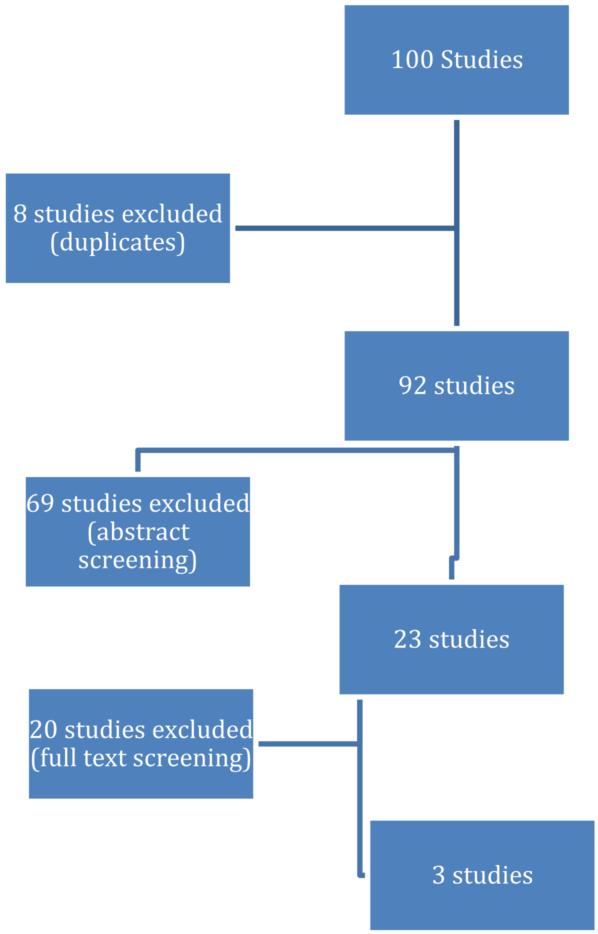
8.1 Search

The search yielded 1175 results sorted by relevance. The first 100 of these results were assessed preliminarily in order to estimate the number of studies which would make it through the screening stage for eligibility.

8.2 Screening

Figure 2 below shows the screening process for the current review.

Figure 2: SCREENING RESULTS FOR THE CURRENT REVIEW



Eight studies were immediately recognised by Covidence software as duplicates and were accordingly excluded. Abstract screening revealed 69 studies which either did not meet the inclusion criteria or contained attributes featured in the exclusion protocol. The remaining 23 studies were scrutinised in great depth relating to content in their full text versions. Only three studies were able to meet the criteria of the current review:

- "A Randomized Controlled Crossover Study of the Impact of Online Music Training on Pitch and Timbre Perception in Cochlear Implant Users" (Jiam, Deroche, Jiradejvong, &

Limb, 2019), published in The Journal of the Association for Research in Otolaryngology;

- "Benefits of Music Training for Perception of Emotional Speech Prosody in Deaf Children With Cochlear Implants" (Good et al., 2017) published in Ear and Hearing; and
- "Comparison of Two Music Training Approaches on Music and Speech Perception in Cochlear Implant Users" (Fuller, Galvin, Maat, Başkent, & Free, 2018), published in Trends in Hearing.

Table 3 below summarises the excluded studies and the reasoning for their exclusion. Some studies had multiple reasons for exclusion however only the main reasoning is shown below. Covidence software did not allow for recording of exclusion reasoning in the abstract screening process and as such, have been listed as "irrelevant" below.

Table 3: EXCLUSION REASONING FOR THE CURRENT REVIEW

Exclusion Reason	**Duplicate**	**Irrelevance (abstract screening)**	**Wrong Intervention**	**Wrong Study Design**	**Full Text Unavailable Online**	**Wrong Patient Population**	**Total**
Studies Excluded	8	69	2	13	1	4	97

As seen above, the most common reason for exclusion was "wrong study design". This was mainly due to the inclusion criteria requiring a randomised controlled trial study design. Two studies consisted of an intervention inappropriate for the current study. One study was not available online and was therefore excluded. For studies were focused on populations inappropriate for this review, in general this was due to the inclusion of bimodally amplified participants in the population with results that were inseparable from the CI population.

8.3 Data Extraction

A brief descriptive summary of each study is expressed in table 4 below.

Table 4: DESCRIPTIVE SUMMARY OF THE INCLUDED STUDIES

Study	**Study Design**	**Population**	**Intervention(s)**	**Comparison/ Outcome**
Jiam (2019)	RCT (crossover)	15 CI users aged 28-84 (mean 63), 17 participants with NH aged 22-75 (mean 37)	Musical training (online) 4 weekly 2-hour sessions recommended	Pitch identification and timbre identification
Fuller (2018)	RCT (Parallel)	19 post-lingually deafened CI users Age: 56-80 (mean 69.1)	Musical therapy or Pitch/timbre training. 6 weekly 2-hour sessions	Vocal emotion identification, Melodic contour identification (organ and piano), Quality of life, Subjective survey.
Good (2017)	RCT (Parallel)	18 CI users Age: 6-15 (mean 10.2)	Musical training 30-minute private piano lesson and two 30-minute home practise sessions per week for 24 weeks.	Montreal Battery for Evaluation of Musical Abilities (MBEMA), Emotional prosody perception.

Further description of the extracted data is available in the synthesis below.

8.4 Quality Assessment

Table 5 below shows the risk of bias assessment for the eligible studies.

Table 5: RISK OF BIAS ASSESSMENTS

	Jiam (2019)	**Good (2017)**	**Fuller (2018)**
Sequence Generation	Low	High	Low
Allocation Concealment	Low	High	Low
Blinding of Participants & Personnel for All Outcomes	Unclear	Unclear	Unclear
Blinding of Outcome Assessors for All Outcomes	Low	Low	Unclear
Incomplete Data for All Outcomes	High	High	Low
Selective Outcome Reporting	High	Low	Low
Other Biases	Unclear	Low	Low

The authors in Jiam (2019) randomly selected participants for two interventions; the method for randomisation was reported to be computer generated resulting in a low level of risk of selection bias due to sequence generation. The allocation was concealed in each respective participant's file resulting in a low risk of selection bias due to allocation concealment. Blinding of personnel and participants was not mentioned in this study resulting in an unclear risk of bias. Blinding of assessments was not reported in this study however was considered unlikely to have an impact on outcomes, resulting in a low risk of detection bias. 26 CI users were enrolled in the study with

15 successfully reaching the endpoint. Those who could not finish the study were well reported, citing declination to participate (n= 5), discontinuation of intervention (n= 4) and lack of participation in musical training exercises (n= 4) as reasons for incompletion. Of the 15 who completed the study, 11 Participants did not meet the 2-hour per week training goal outlined in the intervention guidelines. This means the intervention was not fully completed. It was decided by the authors to include those who did not fully complete the intervention in the analysis, with an average of 0.88 to 1.89 hours of training per week. The amount of attrition and incomplete data witnessed causes the study to be extremely vulnerable to attrition bias (high risk). There is a high risk of reporting bias in the study given that participants were included in the analysis despite incompletion of the intervention. The protocol for this study is very well described, the outcome measures used were all specified and consistent, however the study is at a high risk of reporting bias as a decision made by the authors which had a great impact on the results and analysis. Other bias arises when the author of this study mentions that all CI users whether bilaterally amplified, unilaterally amplified or bimodally amplified were forced to use the same test conditions (using only one CI during testing and removing/shutting down contralateral device). This makes the study easier to analyse and allows the study to meet the inclusion criteria for the current review, however this may have given those who rely on bimodal or bilateral hearing a disadvantage when forced to undergo test conditions. Other bias is also present when considering the mode of intervention administration. Online computer-based training was completed at home by the participants which invites bias as participants may have greatly varying experiences including but not limited to exercise choice, internet bandwidth, computer processing speed, screen size, speaker/headphone type, background noise and environmental

distractions. Because these variables were not monitored or measured, we are left with an unclear risk of bias in this category.

The authors in Good (2017) reported a pseudo-randomised process for selecting participants for each intervention in that those with a location preference were allocated to their preferred intervention whereas those without a location preference were fully randomised using computer generated sequencing. Although this method may be considered the most appropriate given the limitations of the location of intervention, a high risk of selection bias is present wherever full randomisation is not conducted in the sequence generation process. When a location preference is offered to participants in a study, allocation concealment is compromised, also leading to a high risk of selection bias. Blinding of personnel and participants was not mentioned in this study resulting in an unclear risk of bias. Participating families were told that the study's focus was on assessing whether art-based training would lead to various cognitive benefits such as auditory perception, this has a blinding effect in that participants and family are unaware of the intervention and outcome of interest. Blinding of assessments was not specifically reported in this study, however the act of informing participants and their families of a different study focus is considered a form of blinding, reducing the impact on outcomes. These factors lead to a low risk of detection bias. Seven of the 25 participants enrolled in the study dropped out citing inability to commit to training as the reason. Completion of the intervention of interest required participants to self-regulate the content and duration of their training. This was not monitored by the authors, leaving the study extremely vulnerable to variances in intervention completion (or incompletion) and as such, a high risk of attrition bias is present. The protocol for this study is very well described, the outcome measures used were all specified and consistent resulting in a low risk of publication bias due to selective reporting.

The authors in Fuller (2018) reported randomisation in selecting participants for the given interventions however this process was not described. Given the relative ease of digital randomisation it is safe to assume that this process was conducted appropriately resulting in a low risk of selection bias due to randomisation. Allocation concealment was practised until baseline testing where each participant was told of their intervention allocation resulting in a low risk of selection bias due to allocation concealment. Blinding of participants and personnel was not present in the study however it was reported that this had a low risk of impacting outcomes. In the discussion however, it is reported that future studies should use blinding in order to eliminate potential for detection bias. These factors lead to an unclear risk of detection bias. The same pattern was observed for blinding of outcome assessors i.e., absence of blinding, reported low risk of detection bias, blinding encouraged for future studies. Again, this led to an unclear risk of detection bias due to outcome assessor blinding. It is apparent in the study that all participants were able to successfully complete all tasks and allocated interventions with no incomplete data leading to the conclusion that there is a low risk of attrition bias. The protocol for this study is well described, the outcome measures used were all specified, appropriate and consistent resulting in a low risk of publication bias due to selective reporting.

8.5 Analysis

The Jiam et al., (2019) study ("A Randomized Controlled Crossover Study of the Impact of Online Music Training on Pitch and Timbre Perception in Cochlear Implant Users") utilised a crossover RCT study design in an attempt to measure the effect of musical training on music perception. 32 participants were involved in the study consisting of 15 CI users and 17 participants with normal hearing. Participants were randomly allocated to two groups to receive one of two interventions. Both groups completed pitch and timbre identification tasks before training, after completing one training block and after completing the second training block. Assessment was undertaken in a standardised environment in order to eliminate undesired variability.

The pitch assessment required participants to identify which of two tones played consecutively was higher in pitch. A practise block was presented first with 20 trials, followed by two test blocks consisting of 140 trials each. Participants with normal hearing were presented with a different set of stimuli (more difficult) owing to the existing proven degradation of pitch perception in CI users. Comparisons made between the pitch perception of those with normal hearing and those with CI must be heavily scrutinised given that the stimuli used in the assessor was different for each population. An experiment conducted with a fixed number of trials where all of which are individual events consisting of two possible outcomes with unchanging probability (one pitch is higher than the other) is referred to as binomial, otherwise described as a two alternative forced choice task (2AFC).

The timbre assessment required participants to identify one of sixteen instruments after listening to a 0.75 second presentation. Logic X Pro software was the choice of presentation method however it is unclear whether the presentations were of recorded instruments or utilising software instruments. The significance of this is unknown as recordings of acoustic instruments will have different sound qualities to that of digitally replicated instruments. Training consisted of two 4-week blocks, the control block involved non-musical audiobook listening for two hours per week, the intervention of interest involved a wide variety of online musical exercises to be completed as modules at each participant's own pace with an expectation of two hours participation per week. When conducting a systematic review, it is important to identify crossover designs in which the intervention may have an impact which lasts the duration of the study. In this case the intervention of interest has the potential to have lasting effects and bias results in the second round of assessment. In order to mitigate this potential bias, it is standard practice that results from only the first group of participants who complete the intervention of interest are included in the analysis. This can cause drastic changes in the interpretation of results and as such, the following table has been created in order to demonstrate one of the main flaws in conducting a crossover RCT study design in this context.

Table 6: TASK P VALUES BY ROUND OF ASSESSMENT IN Jiam et al., 2019.

Task	**Baseline**	**Round 2 (p value)**	**Round 3 (p value)**
Pitch Task		0.694	<0.001 (0.018 relevant to round 2)
Timbre Task (Arm A)		<0.001	0.649
Timbre Task (Arm B)		0.119	0.013

Using traditional analysis, we can see that the intervention has shown a significant effect for the pitch task at round 3 of testing. Using the mitigation method discussed above, we are forced to dismiss this result and use the results from round 2 of testing. It is then apparent that the intervention had an insignificant overall effect ($p = 0.694$) for the pitch task. It is reported that arm had insignificant interactions with round in the pitch task however the p value was unreported. This is critical as it disproves the hypothesis of the authors that the intervention of interest would have a significant effect on the pitch task and the control intervention would not, meaning there is no clear evidence that the intervention of interest is effective for improving performance in the pitch task. Other effects investigated for the pitch task are shown in the table below.

Table 7: OTHER EFFECTS OBSERVED FOR THE PITCH TASK IN Jiam et al., 2019

Factor	P value	significance
Hearing Status	<0.001	Y
Arm	0.317	N
Round x Hearing	0.673	N
Round x Arm	0.860	N
Hearing x Arm	0.002	Y
3-Way	0.921	N

The effect of hearing status seen above refers to the comparison of results in the pitch task for the CI users and those with normal hearing, this was expected as CI users are expected to have larger thresholds in discriminating pitch when compared to those with normal hearing. The main effect of hearing and arm seen above refers to the difference in performance between the participants with normal hearing in the arm B compared to the participants with normal hearing in arm A. This is thought by the authors to be unrelated to the interventions and other variable explanations in the study and is attributed to differences in existing capabilities. This demonstrates that, particularly with a relatively small sample size, effects can be observed that are sometimes unrelated to the factors and variables of interest in the study, creating difficulty in determining true effects. The timbre task had more promising results, showing a significant improvement (p= <0.001) for arm A at round 2 of testing after receiving the intervention of interest and an insignificant effect (p= 0.649) after receiving the control intervention. Arm B showed an insignificant effect (p= 0.119) after receiving the control intervention and a significant improvement (p= 0.013) after receiving the intervention of interest. This is critical as there is evidence that the intervention of interest is effective in improving timbre recognition and the control intervention is not. The table below shows other significant effects observed in the timbre task.

Table 8: OTHER EFFECTS OBSERVED FOR THE TIMBRE TASK IN Jiam et al., 2019

Factor	P value	Significance
Class	<0.001	Y
Round	<0.001	Y
Hearing Status	<0.001	Y
Class x Round	0.004	Y
Class x Hearing	<0.001	Y
Hearing x Arm	0.018	Y
Class x Hearing x Arm	0.045	Y (marginal)

Eight more unreported factors were investigated (including 4-way analysis) and found to be insignificant. Round was found to have a significant effect (p= <0.001) however improvement was not observed between every round for each arm due to the different interventions. Hearing status was a significant factor again in the timbre task (p= <0.001). Similar to the pitch task, this can be explained by existing differences in existing capability between CI users and those with normal hearing. Attributed to the existing capabilities of individuals involved in the studies, a significant effect was observed for hearing status and arm (p= 0.018) as the participants with normal hearing in arm B, again tended to outperform the participants with normal hearing in arm A (p= 0.095, marginal significance reported) and the CI users in arm A tended to outperform the CI users in arm B (p= 0.079, marginal significance reported). Instruments used as stimuli in the timbre task were categorised into four “classes”; woodwinds, brass, percussion and strings. Those with normal hearing tended to perform best in the percussive instrument class (p= <0.001) relative to the other three instrument classes. This was followed by strings (p= <0.001) relative to brass and woodwinds, brasses and woodwinds. This pattern was also observed in the CI users, performing best in the percussive class (p= <0.001) relative to the other three classes. The deficits seen in CI users' capabilities were more apparent in the strings (-2.8 d prime score) and percussive (-2.7 d prime score) classes and less apparent in the woodwind (-1.3 d prime score) and brass (-1.2 d prime score) classes.

The authors note that the general impression is that CI users rely on percussive features of music, this is proven by the better performance in the percussive task, however CI users are shown to have greater deficits in this instrument class compared to woodwinds and brass instruments when compared to those with normal hearing. There was a correlation found between performance in the pitch task relative to the timbre task, those who had better pitch discrimination generally performed better in the timbre task, however statistical proof was not reported. This correlation was evident in both the cohort with normal hearing and the CI users. No statistically significant correlation was present for the four existing musical experience factors investigated (musical training in years, age at training onset, instrument use in hours per week and music listening in hours per week). No statistically significant correlation was discovered for gender, onset of deafness (pre/post-lingual) or device configuration (unilateral/bilateral CI). The authors provide evidence that a procedural learning effect is potentially present, this is where the act of repeating a task allows an individual to become familiar with the study environment, assessor type, response demands and to develop strategic plans in order to achieve better results. This is similar to the "lasting effect" in crossover trials discussed earlier however the effects noticed here are related to the participant and not the intervention administration directly. This effect is not unique to crossover RCT as the “lasting effect” is but leaves the results vulnerable to bias.

The Fuller et al. (2018) study (“Comparison of Two Music Training Approaches on Music and Speech Perception in Cochlear Implant Users”) consisted of a parallel group RCT study design conducted in an attempt to measure the effect of musical therapy and pitch/timbre training on music perception and appreciation. 19 Dutch speaking, post-lingually deafened, experienced (>12 months) CI users were randomly assigned to three groups to receive one of three interventions, musical therapy, pitch/timbre training or the control (writing, cooking and woodworking). Participants were assessed before and after training. Some of the assessments used were the same as in a previous study by the same author in which participants with normal hearing were assessed while listening through a CI simulator. Upon testing, those with bimodal amplification removed their acoustic hearing aid device allowing the study to meet the inclusion criteria of the current study, potentially give these participants a disadvantage due to undergoing test conditions different to everyday conditions. Assessment was conducted in a heavily controlled environment in an attempt to minimise variability in testing conditions. Verbal answers were recorded to reduce any risk of uncertainty in marking results. Assessments of word and sentence recognition conducted by the authors are considered to be in the vein of speech recognition and outside the scope of music perception and appreciation relevant to the current review. Participants completed assessment of vocal emotion identification, melodic contour identification (piano), melodic contour identification (organ) and quality of life before and after their allocated intervention as well as a subjective survey given only to the musical therapy intervention group after each session. These assessments are within the scope of the current review and as such, are included in the analysis.

Melodic contour identification is described as the ability of an individual to perceive changes in pitch and identify whether a given sound is higher in pitch, lower in pitch or matching in pitch when compared to another sound. It is crucial for a CI user that their device is optimised for MCI in order to accurately perceive pitch change. Vocal emotion identification, also known as vocal emotion recognition or emotional prosody perception, is the capability or of an individual to discern emotional intent from a given speech signal based on its' acoustic properties. Acoustic cues which give rise to recognisable emotion include amplitude, timing, pitch (frequency of F0) and voice quality (soft, harsh or shrill). These can be portrayed by a speaker with varying vocal efforts such as subglottal pressure and laryngeal tension (Frick, 1985). A three-stage model has been proposed in order to describe the process of vocal emotion identification for a listener. The first stage consists of the sensory processing function also known as bilateral auditory processing where the hearing system plays a vital role in conveying the information to the auditory cortex. Stage two consists of projection of auditory information between brain regions where it may lateralise to the right hemisphere of the brain. Stage three consists of higher cognitive processes where an evaluative judgement is made on the emotional content of the sound source (Schirmer & Kotz, 2006). Those with a HI by definition have an impaired ability to accurately carry out the first stage of vocal emotion identification and may suffer deficits in the further two stages due to degraded or underdeveloped neural pathways stemming from lack of stimulation. Vocal emotion identification is vital in understanding the intention of a given passage of speech, it is obvious then that even recognisable speech can still lead to social difficulties.

Assessment consisted of four target emotions as stimuli; joy, anger, relief and sadness presented by two female and two male voices. Each presenter spoke each emotion twice per participant resulting in 32 total trials. This is considered to be a four-alternative forced choice closed set assessment. Time efficiency in assessment completion is vital in order to maintain maximum attention and effort in participants. It has been shown that, in assessing psychophysical parameters of children, a four alternative forced choice assessment is preferrable due to time efficiency when compared to a two alternative forced choice assessment although precision of results and presence (or absence) of biases remain similar (Vancleef et al., 2018). Acclimatisation to the task (familiarisation) was achieved using preliminary testing with different presenting voices, affirmation of correct answers and audiovisual feedback for incorrect answers. Participants were shown the correct answer and reminded of their incorrect answer before moving on to the next trial in the acclimatisation period in order to allow participants to better understand the task. No feedback was given to participants in the main assessment.

Melodic contour identification assessment consisted of nine target stimuli presented as five tones each in the following patterns; flat, rising, falling, flat/rising, flat/falling, falling/rising, falling/flat, rising/flat and rising/falling. This is considered to be a nine-alternative forced choice closed set assessment (9AFC). This method was borrowed from (Galvin, Fu, & Oba, 2009). The tones were presented for a duration of 250 ms with a 50 ms break and difference of either one, two or three semitones. A one semitone step in terms of frequency can be calculated by multiplying the frequency of the reference tone by approximately 1.0595, otherwise described as a 5.95% increase or decrease in frequency between adjacent tones. The lowest frequency tone presented was at 220 Hz. Several conditions were measured; piano alone, organ alone, piano with matching masker (220 Hz), piano with matching masker (880 Hz), organ with piano masker (220 Hz) and organ with piano masker (880 Hz). Both of these assessments required participants to provide a physical response by clicking their chosen answer on a screen containing all possible answers.

The musical therapy group received six weekly training sessions lasting two hours consisting of listening to music, listening to emotional & musical speech, singing, playing an instrument and improvising music. The environment for this training group was described as social, dynamic and multimodal. The pitch/timbre training group received six weekly sessions lasting two hours consisting of computerised MCI presented through electronically synthesised instruments; piano, organ, violin, trumpet, clarinet and glockenspiel. Beginning with intervals of six semitones the difficulty was increased gradually down to intervals of only one semitone for the CI cohort. For the purposes of training diversification, an instrument identification training task and "daily-life" sound identification training task were also administered. During these training tasks a similar method was employed as the acclimatisation period where audiovisual feedback was provided for incorrect answers and affirmation for correct answers. The control group also received six weekly training sessions lasting two hours consisting of writing, cooking and woodworking with no musical element involved, however the training environment was described as similar to the music therapy group.

A subjective survey with a focus on quality of life was administered before and after training. The survey, known as the Nijmegen cochlear implant questionnaire (NCIQ), was developed in order to measure health aspects specific to CI users (Hinderink, Krabbe, & van den Broek, 2017). The questionnaire consists of three main fields of interest; sound perception and speech production, psychological function (self-esteem) and social activity and interaction.

The mean differences between pre/post training for the vocal emotion identification task are shown in the table below

Table 9: MEAN DIFFERENCES POST TRAINING FOR THE VEI TASK IN Fuller et al., 2018.

Training Group	Baseline	Endpoint
Musical Therapy Training		+1.1%
Pitch/Timbre Training		-0.9%
Control		-4.2%

As seen in the table above, only minor differences in performance were observed post-training. Overall training showed no significant effect (p= 0.067). In order to assess the significance of the +1.1% change in performance for the musical therapy group, a one-way ANOVA was performed. A significant effect (P= 0.022) was observed showing that musical therapy training is effective in improving vocal emotion identification. The mean differences between pre/post training for the melodic contour identification task are shown in the table below.

Table 10: MEAN DIFFERENCES POST TRAINING FOR THE MCI TASK IN Fuller et al., 2018

Training Group	**Mean Difference**	**Mean Difference (with A3 Masker)**	**Mean Difference (with A5 Masker)**
Musical Therapy Training	+6.3	-2.6	+5.3
Pitch/Timbre Training	+8	+21	+15.4
Control	-8	-6.8	-1.9

As seen in the table above, larger mean differences were observed post-training in the pitch/timbre training group than the musical therapy training group. Overall a training effect did not reach significance and was not observed (P= 0.192). A two-way RM ANOVA was performed on the data for the pitch/timbre training group in order to determine the significance of the post-training change using masker and training as the factors for both the piano and organ assessor. The piano assessor showed a marginally significant effect due to training (p= 0.045); the masker had an insignificant effect in this case (p= 0.149). The organ assessor revealed a different pattern, falling short of significance for training (p= 0.064) and showing a significant effect for the masker (p= 0.049). The importance of correct and accurate statistical analysis is demonstrated here given that although it appears there is a large difference in mean performance for the pitch/timbre training group, the training effect only marginally meets significance for the piano assessor. The mean differences post-training for the NCIQ (subjective quality of life questionnaire) are shown in the table below.

Table 11: MEAN DIFFERENCES POST TRAINING FOR THE NCIQ QUESTIONNAIRE IN Fuller et al., 2018

Training Group	Sound Perception (basic)	Sound Perception (advanced)	Speech Production	Self-Esteem	Activity Limitations	Social Interactions	Total
Musical Therapy	-2.5	+11.1	-2.5	-1	+8.4	+8.1	+3.6
Pitch/Timbre	-5.8	+3.7	-2.1	-7.5	-8.7	-12.9	-5.5
Control	+1.7	+1	+7.9	-2	-3.6	+1.6	+1.1

Overall, no significant effect was observed for training (p= 0.928). The same can be said for each training group assessed individually. Providing evidence that musical training may not have an effect on quality of life. Finally, significant effects were shown for training for the subjective survey given only to the musical therapy group in order to track personal progression impression. Six questionnaires were completed in total (one after each training session). Questionnaires three, four, five and six all showed a significant effect (p= <0.05) of training in reference to questionnaire one. Questionnaires five and six also showed a significant effect (p= <0.05) for training in reference to questionnaire two. This provides evidence that musical therapy training has the potential to improve self-assessed performance in music perception and appreciation. In both of these analyses, music perception and participation (playing piano) in the training were rated significantly (p= <0.05) higher than musical speech perception.

The Good et al. (2017) study ("Benefits of Music Training for Perception of Emotional Speech Prosody in Deaf Children with Cochlear Implants") utilised a parallel group RCT study design in an attempt to measure the effect of musical training on music perception. 18 CI users between the ages of six and fifteen were randomly assigned to one of two groups where they would receive six months of music training or six months of art training (control group).

The Montreal Battery for Evaluation of Musical Abilities (MBEMA) is a tool used to objectively measure the musical abilities of an individual, comprised of scale, contour, interval, rhythm and incidental memory. Each participant completed the MBEMA as well as an emotional prosody perception assessment at three stages; before training, during training (after 12th lesson) and after training (after 24th lesson). The MBEMA required participants to complete two practise trials with immediate feedback followed by 20 test trials without feedback for each of the test categories. The design of the MBEMA in this context required participants to listen to two versions of the presentation and asked whether the presentations were the “same” or “different”. Each presentation of each sub-test had a 50% chance of being the same and a 50% chance of being different, this assessment is considered to be a two-alternative forced choice closed set assessment also known as binomial.

Emotional prosody perception, another term for vocal emotion identification described above, is the ability of an individual to recognise/identify the emotional intent of a speaker. It is obvious that a lack of this ability can lead to minor misunderstandings or more serious confusion in various social situations. Volume, facial movement, pitch, timbre and pacing are some of the factors that contribute to an individual’s capability to perform accurate emotional prosody perception. CI users experience a deficit in many of these areas leading to potentially extreme social difficulties. Emotional prosody perception was assessed using a novel assessment created specifically for the study, consisting of voice recordings from one male adult, one female adult, one male child and one female child. Presentations of the adult stimuli were separated from the child stimuli in “blocks”. The sentence presented was borrowed from “Individual differences in the nonverbal communication of affect: The diagnostic analysis of nonverbal accuracy scale” (Nowicki & Duke, 1994) and is as follows: “I am going out of the room right now, but I’ll be back later”. The stimuli were presented in either a low level of emotional expression or a high level of emotional expression and consisted of four emotions: sad, happy, fearful and angry. Eight possible answers were available to the participant for a given trial, this is known as an eight alternative forced choice assessment (8AFC). Two separate conditions were also presented in separate blocks: audio only and audiovisual (accompanied by facial cues). A total of 60 trials were administered in a randomised order per participant.

The music training group completed 30-minute piano lessons segmented into two modules, one focused on scales, hand positioning and other traditional piano training methods, and one focused on learning basic songs while being encouraged to practise vocally. The art training group completed 30-minute painting lessons segmented into two modules, one focused on traditional painting methods/theory and one focused on completion of an artwork. These sessions were completed weekly for a total of 24 weeks. Both groups were expected to complete two 30-minute sessions of practise at home weekly. Results for the MBEMA task are shown in the table below.

Table 12: TRAINING EFFECTS FOR THE MBEMA TASK IN Good et al., 2017

Training Group	**Midpoint Assessment**	**Endpoint Assessment**
Musical Training	Insignificant (p= 0.161)	Significant (p= 0.002 relevant to baseline, p= 0.003 relevant to midpoint)
Art Training (Control)		Insignificant (p= 0.7)
Overall		Significant (p= 0.002)

The musical training group (intervention of interest) showed significant improvements, with a main effect of time at the endpoint both relevant to baseline assessment (p= 0.002) and midpoint assessment (p= 0.003). The same can not be said for the art training group (control intervention) which showed an insignificant effect of time at the endpoint (p= 0.7). When the data was combined (both intervention groups), an overall main effect of time was observed (p= 0.002). Subgroups of the MBEMA were analysed individually and a main effect of time was observed for the contour task (p= 0.037), the rhythm task (p= 0.03), the memory task (p= 0.04) and (marginally) the interval task (p= 0.068). This provides evidence that musical training is effective in improving general musical abilities. The results in performance for the emotional prosody perception task are shown in the table below.

Table 13: TRAINING EFFECTS FOR THE EPP TASK IN Good et al., 2017

Training Group	**Midpoint Assessment**	**Endpoint Assessment**	**Audio Only**	**Audiovisual**
Musical Training	Significant (p= 0.047)	Significant (p= 0.015 relevant to both baseline and midpoint, p= 0.004 relevant to baseline, p= 0.08 relevant to midpoint only)	Significant (p= 0.034 relevant to both baseline and midpoint, p= 0.14 between baseline and midpoint, p= 0.038 between midpoint and endpoint)	Marginally Significant (p= 0.067 relevant to both baseline and midpoint, p= 0.04 between baseline and midpoint, p= 0.77 between midpoint and endpoint)
Art Training		Insignificant (p= 0.24)	Insignificant (p= 0.322)	Insignificant (p= 0.215)
Overall		+7.2% mean difference Significant (p= 0.001)		

Overall a significant improvement in performance was seen with a main effect of time (p= 0.001) and a mean difference in score percentage of +7.2%. Significant improvements were observed in the musical therapy group with a main effect of time at the endpoint relevant to the baseline and midpoint (p= 0.015), at the endpoint relevant to the baseline (p= 0.004) and at the endpoint relevant to the midpoint (p= 0.08). The same cannot be said for the art training group which showed an insignificant effect of time at the endpoint (p= 0.24). The assessments were also analysed according to the modality of stimuli presented by separately analysing the audiovisual and audio only tasks. No significant gains were again observed in the art training group with an insignificant effect of time at the endpoint for both the audio only (p= 0.322) and the audiovisual (p= 0.215) tasks. For the audio only task, the musical training group showed significant improvements with a main effect of time at the endpoint (p= 0.034) and between the midpoint and endpoint (p= 0.038) but showed an insignificant effect of time between the baseline and midpoint (p= 0.14). For the audiovisual task, the musical training group showed significant improvements with a main effect of time between the baseline and midpoint (p= 0.04) but showed only a marginal effect of time at the endpoint (p= 0.067) and an insignificant effect of time between the midpoint and endpoint (p= 0.77).

8.6 Synthesis

In order to fulfill the requirements of a systematic review, analysis must not only report findings from the individual studies but explore inter-study relationships. The inter-study factors analysed in this review are the assessor type, population factors, study setting and intervention factors. Collating reported effect sizes and mean differences (where available) is the most efficient method for carrying out this task. Population characteristics of each study are shown in the table below as well as the main effect size witnessed post-training in an attempt to identify relationships between intervention success and population variables.

Table 14: CHARACTERISTICS AND EFFECT SIZES FOR THE INCLUDED STUDIES: Jiam et al., 2019, Good et al., 2017 and Fuller et al., 2018.

<table>
<tr><th>Study Author</th><th>Age (years)</th><th>Onset of HI</th><th>Duration of CI use (years)</th><th>Population Size</th><th colspan="4">Main Effect Size (p value)</th></tr>
<tr><td>Jiam (2019)</td><td>22-75 (mean 37)</td><td>6 pre-lingual, 9 post-lingual</td><td>1-28 (mean 6.78)</td><td>20 (sex unreported)</td><td colspan="2">Pitch
<0.001*</td><td colspan="2">Timbre
<0.001, 0.013</td></tr>
<tr><td>Good (2017)</td><td>6-15 (mean 10.2)</td><td>15 pre-lingual, 1 post-lingual, 2 unknown</td><td>1.02-9.53 (mean 6.25)</td><td>18 (12 male, 6 female)</td><td colspan="2">MBEMA
0.002</td><td colspan="2">EPP
0.015****</td></tr>
<tr><td rowspan="2">Fuller (2018)</td><td rowspan="2">56-80 (mean 69.1)</td><td rowspan="2">19 post-lingual</td><td rowspan="2">3-13 (6.3)</td><td rowspan="2">19 (sex unreported)</td><td>VEI
0.022**</td><td colspan="2">MCI (piano)
0.045***</td><td rowspan="2">MCI (organ)
0.064***</td></tr>
<tr><td>NCIQ
0.928</td><td colspan="2">MTGQ
<0.05</td></tr>
</table>

**overall effect not proven to be related to intervention of interest, **musical therapy group only (intervention of interest), ***pitch timbre training group only (musical therapy group showed no significant effect of training), ****musical training group only (intervention of interest), VEI = vocal emotion identification, MCI = melodic contour identification, NCIQ = Nijmegen cochlear implant questionnaire, MTGQ = musical therapy group questionnaire, MBEMA = Montreal Battery for Evaluation of Musical Abilities, EPP = emotional prosody perception.*

A large disparity was observed in the age groups of the three studies. Adult populations were the focus of Jiam (2019) and Fuller (2018) with mean ages 37 and 69.1 respectively, whereas a paediatric population was the focus of Good (2017) with a mean age of 10.2. This demonstrates the importance of taking age into consideration for studies reporting an “adult population” which still have the potential for great variance. The procedures which audiological professionals undertake in order to provide optimal amplification for a client can vary greatly between children, young adults, middle-aged adults and senior adults. This consideration must also be taken into account when building an evidence base for best clinical practice. Because of the large disparity between age groups, we have the potential to identify a population in which the intervention is more or less effective.

Analysis of the population factors above however, is only appropriate for matching or similar assessors. For example, the MBEMA is a unique assessor in that it measures a broad range of musical capabilities of the participant, comparing this to an assessment of a specific area of music perception such as MCI is inappropriate. It is apparent that the only shared measure between the studies is the vocal emotion identification (emotional prosody perception) tasks. Fuller (2018) shows that in a paediatric population, musical therapy has a significant effect on vocal emotion identification ($p = 0.022$) and Good (2017) shows that in an adult population, musical training has a significant effect on emotional prosody perception ($p = 0.015$). This is promising as musical training is proven to lead to improvements in vocal emotion identification in both paediatric and adult populations.

The musical therapy in Fuller (2018) consisted of six, weekly 2-hour sessions of listening to music, listening to emotional & musical speech, singing, playing an instrument and improvising music whereas the musical training in Good (2017) consisted of 24, weekly 30-minute piano lessons segmented into two modules, one focused on scales, hand positioning and other traditional piano training methods, and one focused on learning basic songs while being encouraged to practise vocally as well as two self-managed 30 minute practise sessions. It is reported that the self-managed practise sessions were not monitored and, as such provide a great possibility for variances in training completion and experiences. Although the training methods mentioned above have the same goal, they are structured vastly differently in both timing and content. Given that an insignificant effect was observed for the audio only task in Good (2017) for the musical training group ($p= 0.14$) between baseline assessment and the midpoint (after 12 weeks of training), the relatively long training time period was justified when a significant effect was later observed at the endpoint ($p= 0.034$). This is interesting as it highlights the importance of ongoing training and encourages the concept of musical training for CI users beyond the study, this may have something to do with the acclimatisation and learning capabilities of the paediatric population involved.

Different levels of significance were witness at different stages in Good (2017) showing that improvement due to musical training is not linear and may vary in effectiveness over time. Consistent, ongoing training is indicated. The Good (2017) EPP assessment was considered an 8AFC task whereas the Fuller (2018) VEI task was considered a 4AFC task. Both of these assessors proved accurate enough to see clear effects post-training, confirming their appropriateness for the studies.

The paediatric population in Good (2017) consisted of a large majority of pre-lingually deafened children with one having post-lingual deafness and 2 unknowns. This is a stark contrast to Fuller

(2018) which consisted of a population of post-lingually deafened adults only. It is encouraging to see that, regardless of onset of HI, training was proven to be effective. Jiam (2019) consisted of a more balanced population with six pre-lingually deafened adults and nine post-lingually deafened adults.

A large variance was seen overall in duration of CI use ranging from one to 28 years, this is particularly evident in Jiam (2019). An extremely large variation was seen in the Jiam (2019) population ranging from one to 28 years of use. The duration of CI use, in terms of means, was relatively similar between the studies (6.78, 6.25, 6.3 (years)). Future studies are indicated in populations with varying levels of CI experience in order to assess the effectiveness of training depending on experience.

Although pitch perception and melodic contour identification are similar in concept to each other, they have distinct differences which make them inappropriate for inter-study analysis. For example, the pitch task in Jiam (2019) required participants to identify which of two tones were higher in pitch (2AFC) whereas the MCI task in Fuller (2018) required participants to identify which of nine melodic contours was presented (9AFC). Arguably, the 9AFC task utilised in Fuller (2018) is more relevant to music perception given that melodic contour, commonly referred to as melody, is a highly featured aspect of music. This is not to say that the 2AFC task in Jiam (2019) is irrelevant to music perception as improved capabilities in discriminating tones will provide a basis and support MCI capabilities.
Although an effect of time was observed for the pitch task ($p= <0.001$) in Jiam (2019), it is not evident that the training was responsible and as such, a main effect of training cannot be

concluded. The effect observed in Fuller (2018) however, was unique to the pitch/timbre training group who showed large gains in mean percentage for the MCI task reaching significance in the piano presentation medium (p= 0.045) but falling short of significance in the organ presentation medium (p= 0.064). In this case the training consisted of a large amount of experience gained with the instrument used in the assessor (piano), showing that with specific measures of music perception, specific training may be administered accordingly. Although these studies generally have the same outcome goal of improving music perception and appreciation, each of them attempted to accomplish this in different ways resulting in a large amount of variation in the undertaking of interventions.

A noticeable theme within the studies is the concept of self-training, it is present through the Meludia task in Jiam (2019) and self-managed practices in Good (2017). When allowing individuals to self-manage their education there can be great variability in participation and completion rates owing to many different factors including but not limited to access to required technology, disorders which cause attention deficits, motivation, time management and honesty (in reporting completion). A systematic review of self-regulated learning found that only a quarter of self-regulated learning programs involved evidence based supportive structures and that such supports for self-regulated learning led to statistically significant benefits when compared to unsupported self-regulated learning (Brydges et al., 2015). The efficacy of self-regulated learning is proven however can be enhanced with proper guidance and should be considered in future research.

Population sizes were relatively small when compared to healthcare research with a range of 18-20 CI users between the studies. The sample sizes (population sizes) were not appropriately calculated to prove adequate numbers for meaningful statistical analysis in Good (2017) and Fuller (2018). Good (2017) mentioned the small sample size but provided no appropriate calculation for evidence. With an assumed effect size of 0.3 and priori of 0.8, Jiam (2019) calculated an appropriate sample size of 20 participants per population. This recommended number of participants was not reached with only 15 participants in the CI group and 17 in the group with normal hearing. Future studies are encouraged to use proper sample size calculation in order to strengthen the findings.

Only Good (2017) reported the sex types of the participants consisting of 12 males and six females, however sex reported as having an insignificant effect. CI configurations varied greatly within the studies, featuring 32 unilaterally implanted, 16 bilaterally implanted and four bimodally amplified participants overall. In order to provide consistency for assessors and attempt to control inter-participant variations which are not of interest, participants in all of the studies were forced to utilise only one of their CI devices as well as blocking the contralateral ear when necessary. Those with bilateral CI and bimodal amplification may have experienced a disadvantage given that they are not being assessed in their regular device configuration. The studies do not report whether or not this method was also encouraged during the training periods, it is assumed that bilateral and bimodal users were able to use their everyday configuration during training times. In general, promise was shown for improvement in music perception and appreciation tasks post-training.

9 Discussion

A systematic review which results in an undesirable sample size of studies is still worthy of completion/publication as there are generally causative reasons ripe for discussion. Unfortunately, the strength of the evidence base within a systematic review was compromised by a small sample size. This review resulted in three studies meeting the eligibility criteria to be included in the analysis. This is relatively low compared to typical systematic reviews conducted in the field of healthcare research. A factor that may be responsible for this is the relative youth of CI research particularly when focused on non-speech sounds. Environmental and non-speech sounds are trending in audiological assessment. Correctly however, the main focus of amplification is still to provide adequate speech discrimination to a patient. Another factor that may be responsible is the strictness of the inclusion and exclusion protocol used in the current review. An extremely limited number of RCTs have been conducted in this very specific field of audiology, leading to many exclusions.

The trend towards bimodal amplification discussed earlier forced many studies to be excluded given that the current study sought to investigate CI users only. It is recommended that future reviews use different criteria allowing those with bimodal amplification to be included. Although the majority of studies conducted in the field are beneficial to CI users (particularly the participants in the study) it is crucial that when conducting future studies in the field, greater effort is put into proper reporting. Minimising flaws such as bias, poor study design, inadequate sample sizes and incomplete reporting will result in more long-term benefits for both current CI users and future CI users. There a several resources available for researchers in order to ensure that studies are being conducted and reported wholly and accurately.

Methods described as assessors for music perception and appreciation in the literature are generally measuring a simplified and specific subcategory of musical content. Pitch, timbre and vocal emotion identification have been the main focus of the field of study. In order to provide CI users with amplification optimised for music perception and appreciation, audiological professionals must understand the complex characteristics of music and develop assessors that encompass them.

The interventions of interest in each study consisted of weekly regimented training with two featuring self-regulated training in addition. The weekly training is proven to be effective in these cases. The self-regulated element of training was only monitored in one of the studies, exposing incompletion of the expected duration of training. Although self-regulated learning can be effective, monitoring and support are vitally important in order to identify and consider variations between completion of interventions for each participant as well as provide the best chance for effective training. Ensuring adequate intervention completion allows us to eliminate variances in the experience of the participants, mitigating the impact of a potential limitation. Limitations were present in all three studies due to small sample sizes. Jiam (2019) was the only study to properly calculate the ideal sample size and was not capable of fulfilling this requirement, the other two studies did not provide a sample calculation so we must assume the sample size was not adequate.

The environment in which the intervention was conducted varied between the studies, one study consisted of social, dynamic training, one consisted of one-on-one tuition and one consisted of online training at home. Given the relative success of each training method, it can be suggested that different types of training environment have the potential to be effective.

Music perception has largely been the focus of previous research with proven results. Music appreciation is a less tangible concept with less attention in previous research. Although assessment of quality-of-life was found to have insignificant changes post-training in Fuller (2018), given the importance of music appreciation for quality of life highlighted in the literature review in this theses, further research is indicated.

Shorter intervention duration resulted in higher completion rates, however long-term training is preferred as effects tend to vary in significance over time. Longer training periods allow a CI user to make the most out of the intervention being provided. Online training utilising the Meludia program (Meludia, Paris, France) showed significant improvement in timbre recognition and should be considered in the rehabilitation of CI users.

One of the main reasons that studies were excluded for the current review was using the wrong study design. Randomised Controlled Trials (RCT) are generally considered in research to be a gold standard offering reliable results and positive outcomes for patients. There are benefits and disadvantages to using a parallel design in comparison to a crossover design. As mentioned earlier, crossover designs tend to yield unreliable results due to the lasting effect of training for those in the group receiving the intervention of interest first. Parallel designs however, require some participants to be allocated to a control group, meaning some of the participants will not be receiving an intervention aimed at improving their music perception and appreciation. For future studies, a parallel design is recommended in order to provide accurate analysis and allow for intervention programs to be proven and implemented in the wider CI population, including the control group (after publication of the study).

Reporting quality in the studies included in the current review was unsatisfactory in some areas i.e., reporting of confidence intervals, mean differences, training dates and omitted significance indicators. Although it may seem unimportant at the time of the study to report some of these factors, all available information should be presented in order to allow proper analysis and interpretation both in future reviews and for audiological professionals wishing to interpret and build an evidence base for best practice. Protocols for proper reporting of RCT studies is outlined in "The Revised CONSORT Statement for Reporting Randomized Trials: Explanation and Elaboration" (Altman et al., 2001), this can be used as a template in order to ensure high quality of reporting, allowing for higher quality subsequent reviews and adding to the growing evidence base for the field of study.

Stimuli in two of the tasks in the studies were presented via Logic X Pro music software. Digitally synthesised instruments may not be able to represent an accurate portrayal of acoustic instruments and as such, recordings of acoustic instruments are preferred for future assessment. This could be argued given that fact that modern music tends to consist of an array of digitally replicated instruments.

No statistically significant correlation was discovered for gender, onset of deafness (pre/post-lingual) or device configuration (unilateral/bilateral CI) in Jiam (2019), providing evidence that musical training has similar effects on various populations and should be considered for all CI recipients.

10 Limitations

The current review is subject to several limitations. The inclusion and exclusion criteria used to undertake the screening process was strict. Although this was intentional in order to analyse studies that were of high quality, a specified population, have highly specified interventions and assessors and in a relatively recent timeframe, it resulted in very few studies being included in the analysis. If relaxation of the inclusion and exclusion protocol was present, more studies would have been available for analysis, providing a more complete and encompassing more aspects of the field by sacrificing relevance, quality and depth of analysis. Each study exhibited biases from varying sources. When bias is present or has the potential to be present, the results of a study become subject to interpretation depending on the severity of the bias. As a result, the evidence base presented in the current study is subject to bias which must be considered in interpreting results. The decision to use a narrative synthesis provided limitations including lack of transparency (Dixon-Woods, Agarwal, Jones, Young, & Sutton, 2005) and formal protocol for conducting the synthesis (Mays, Pope, & Popay, 2005), this may result in reporting bias.

11 Funding

It was reported that no funding or sponsorship was provided for the completion of "A Randomized Controlled Crossover Study of the Impact of Online Music Training on Pitch and Timbre Perception in Cochlear Implant Users" (Jiam, 2019). It was reported support was provided for Deniz Baskent, a collaborator in "Comparison of Two Music Training Approaches on Music and Speech Perception in Cochlear Implant Users" (Fuller, 2018) in the form of a

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12 Conclusion

The findings in this review can be considered additive to the findings from previous reviews mentioned in the literature review. The historical literature shows that inter-professional practice is vital to providing optimal outcomes for CI users. Otolaryngologists must continually educate themselves or be educated in optimal combinations of current CI technology, soft (minimal damage) surgical approaches and apical electrode array insertion. Audiologists involved in the fitting of CI must continually educate themselves or be educated on current CI technology, coding strategies, non-speech assessment and training programs. Creation and development of novel assessments and training methods are necessary in order to address and properly assess the complexities of music perception and appreciation. If training programs are to be developed, audiologists must be aware of them and be willing to provide them for their patients. CI users can advocate for themselves by expressing their preferences in CI setup as well as partaking in training programs and studies in order to optimise their CI experience.

Music is an important part of human life which everyone should have reasonable access to. Hearing impairment greatly degrades music perception and appreciation. Cochlear implant users tend to have an extremely distorted experience of music compared to those with normal hearing. Research is available showing the correlation between musical participation and quality-of-life, this should act as a motivator for improving music perception and appreciation in CI users. Although some aural and musical training programs have proven to be effective, they are generally reliant on active participation (volunteering for studies) and self-motivation. It is the responsibility of audiologists to progress this field of study in order to improve music perception and appreciation for cochlear implant users.

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