

Benefits of music training for children with hearing loss

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“Benefits of music training for children with hearing loss”

Macquarie University

“Music is moral law. It gives soul to the universe, wings to the mind, flight to the imagination, a charm to sadness, and life to everything.” — Plato

“Music is crap, aliens told us so.” — Custard

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Conference Presentations

Portions of this thesis have been presented at the following conferences:

Lo, C. Y., Looi, V., McMahon, C. M., Thompson, W. F. (2018). *Music training for children with prelingual hearing loss: music, speech, and psychosocial enhancement*. Paper presented at the 15th International Conference on Cochlear Implants and Other Implantable Auditory Technology (CI2018), Antwerp, Belgium.

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Abstract

Children with hearing loss report difficulties in a range of challenging listening situations such as music and speech-in-noise perception and have poorer psychosocial outcomes compared to their typical-hearing peers. Music training has been proposed as a suitable form of habilitation; driven primarily by typical-hearing music training studies that indicate a speech-in-noise enhancement for adults and children. The number of studies investigating the benefits of music training for children with hearing loss are modest, though recent studies have shown improvement for some elements of speech perception such as emotional prosody and lexical tone recognition. This thesis aimed to investigate the music, speech, and psychosocial benefits of a 12-week music training program for children with hearing loss.

Eleven children aged between 6.13 and 9.24 years ($M = 7.48$, $SD = 1.07$) with moderate to profound prelingual hearing loss (5 bilateral cochlear implant recipients, 4 bimodal users, 2 bilateral hearing aid users) participated in this study. The design was a pseudo-randomised, longitudinal study (half the cohort was waitlisted, initially serving as a passive control group). Music training was 12 weeks in duration, consisting of weekly face-to-face group-based music therapy sessions with activities such as drumming, singing, dancing, and improvisation; and a suite of online music apps 3 times a week that consisted of activities such as creating compositions, and identification of high, low, fast, or slow sounds. Children were tested at the following timepoints: double baseline (pre-training), mid-training, post-training, and at follow-up (12-weeks after training ceased). The test battery consisted of the Clinical Assessment of Music Perception to assess pitch and timbre perception, a Music Appreciation Questionnaire, the Australian Sentences Test in Noise, the Spectral-temporally Modulated Ripple Test, the Macquarie Battery of Emotional Prosody, a Question/Statement Prosody Test, the Strengths and Difficulties Questionnaire that provides an overview of behaviours, emotion, and relationships, the Paediatric Quality of Life Inventory—a generic measure of health-related quality of life, the Hearing Environments and Reflection on Quality of Life, and the Glasgow Children's Benefit Inventory.

Statistical analyses for the main hypotheses were conducted with linear mixed models, controlling for hearing age, device, and prior formal music training. Double baseline measures (separated by 1-week) were not significantly different, indicating high test-retest validity; additionally, the waitlist group (separated by 12-weeks) were not significantly different, indicating no improvement from natural maturation and development. At the post-training point, statistically significant results

were found for: speech-in-noise perception (speech reception thresholds improved by 1.1 dB ($p = .036$), timbre perception by 8 percentage points ($p = .028$), spectral resolution by 2 rpo ($p < .001$), and question/statement prosody by 14 percentage points ($p = .004$), and various music appreciation measures. Psychosocial outcomes also improved significantly for internalising behavioural problems ($p = .001$), and total scores ($p = .012$). Non-significant results were found for emotional prosody, pitch perception, all domains for the Paediatric Quality of Life Inventory and the Hearing Environments and Reflection on Quality of Life.

The findings suggest even a modest amount of music training has benefits for music, speech and psychosocial outcomes. The results provide further evidence that music training is an excellent complementary means of habilitation to improve the outcomes for children with hearing loss.

Declaration

I hereby declare that this thesis has not been submitted for any degree to any other university or institution. The sources of information used and the extent to which the work of others has been utilised have been indicated in this thesis in the manner conventionally approved in the research field in which the thesis fits.

Ethical and scientific review, guidance, and approval was obtained from:

- Macquarie University Human Research Ethics Committee (Medical Sciences); reference: 5201600081.

I certify that I conceived the original idea and took leadership to conduct this research project, including writing the content of this thesis. My three supervisors (Prof Catherine M. McMahon, Dr Valerie Looi, and Prof William Forde Thompson) assisted in improving the research protocols, analyses, interpretation of the data, as well as the quality of the written components. I conducted the majority of the data collection, with additional support from two research assistants (Denielle Plara and Roisin McDonnell). Statistical support was obtained from Dr Peter Humburg.



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My career began as an audio engineer and I find myself in the unusual situation of having come full circle. Only, instead of providing music for children's entertainment, I have spent the last 3 years investigating the benefits of music for communication and quality of life for children with hearing loss. It has been a lot of fun! But with every ending, there is of course a beginning; and behind every project, a monumental team that holds it all together.

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Acronyms and Abbreviations

ACE	Advanced combination encoder
AFC	Alternative forced choice
4TB	Four-talker babble
AusE	Australian English
AuSTIN	Australian Sentences Test in Noise
bpm	Beats per minute
BTE	Behind-the-ear
CAMP	Clinical Assessment of Music Perception
CI	Cochlear implant
CIS	Continuous interleaved sampling
EMM	Estimated marginal means
Fo	Fundamental frequency
GCBi	Glasgow Children’s Benefit Inventory
GZLM	Generalized linear mixed model
HA	Hearing aid
HEAR-QL	Hearing Environments and Reflection on Quality of Life
HINT	Hearing in Noise Test
HRQoL	Health-related quality of life
ICC	Intraclass correlation coefficients
IDR	Input dynamic range
ITE	In-the-ear
LMM	Linear mixed model
MBEMA	Montreal Battery for Evaluation of Musical Activities
MBEP	Macquarie Battery of Emotional Prosody
MDS	Multidimensional scale
MSMA	Morton Subotnick’s Music Academy
PedsQL	Paediatric Quality of Life
PEPS-C	Profiling Elements of Prosodic Systems—Child version
pps	Pulses per second
QoL	Quality of life
QSPT	Question/Statement Prosody Test
QuickSIN	Quick Speech-in-Noise Test
REML	Restricted Maximum Likelihood

RMFQ	Role of Music in Families Questionnaire
RPO	Ripples per octave
SDQ	Strengths and Difficulties Questionnaire
SIN	Speech-in-noise
SNHL	Sensorineural hearing loss
SMRT	Spectral-temporally Modulated Ripple Test
SNR	Signal to noise ratio
SRT	Speech reception threshold
TH	Typical hearing
UNHS	Universal neonatal hearing screening
WHO	World Health Organisation

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Chapter 1 — General introduction

Background and motivation

Music is ubiquitous—prevalent in everyday lives, found across all cultures, and used in a wide variety of contexts. There is a growing body of evidence that musical skills enhance auditory abilities, and of particular interest are enhancements to speech perception such as speech-in-noise (SIN) and prosody—the suprasegmental patterns of stress and intonation that infer meaning in speech; although such benefits have been derived primarily from cross-sectional studies of typical-hearing (TH) professional musicians (Coffey, Mogilever, & Zatorre, 2017). Furthermore, studies investigating group-based musical activities for TH children have found some positive effects for psychosocial wellbeing and behaviours such as cooperation and prosociality (Dumont, Syurina, Feron, & van Hooren, 2017). Taken together, these perceptual and psychosocial benefits may be particularly useful for children with hearing loss, as they have poorer SIN and psychosocial outcomes than their TH peers (Schafer & Thibodeau, 2006; Stevenson, Kreppner, Pimperton, Worsfold, & Kennedy, 2015).

Remarkably, the intersection between music and deafness has a long history. Likely to be one of the earliest written reports of music for the deaf, Turner (1848 p.2) described their first meeting with Augusta Avery (Darrow & Heller, 1985):

“We were not prepared to hear a young lady, made so deaf when eighteen months old as to be unable to perceive the tones of a piano-forte, playing correctly in point of time and expression, upon that instrument, the simple airs and lessons usually taught to beginners the first year of their instruction. But this we did hear; and we confidently affirm that her performance was fully equal to that of any young person we have met with, who had practised no longer than she had.”

The landscape for deaf individuals has changed dramatically since 1848, with technological breakthroughs such as cochlear implants (CIs) that can provide the sensation of sound to individuals with profound hearing losses. As such, the expectation of typical and possible outcomes has expanded. However, the developmental focus on children with hearing loss is overwhelmingly directed towards improving language and speech perception outcomes, and a holistic understanding of the benefits of music remains less developed. For a range of reasons, music training studies for children with hearing loss are limited in number and scope.

“Designing and implementing music training with children is logistically daunting for a number of reasons, including: recruiting and retaining an appropriate, sufficient sample size; adequate funding to support methodology; feasibility of scheduling the training and testing; maintaining consistency of training parameters over time. All of these pose significant challenges to mounting a well-designed study.” (Gfeller, 2016, p.551)

Nonetheless, recent music training studies are a source of cautious optimism, with results by Chen et al. (2010) indicating the capacity to improve music perception such as pitch—the perceptual ordering of sound from low to high (Oxenham, 2017; Trainor & Unrau, 2012), as well as benefits for speech domains with improvements found in the perception of prosody (Good et al., 2017; Torppa, Faulkner, et al., 2014), and lexical tone in Mandarin (Cheng et al., 2018). While psychosocial benefits from music training remains poorly explored, Yucel, Sennaroglu, and Belgin (2009) provide some evidence that shared musical experiences can lead to closer parent-child relationships; and a study by Innes-Brown, Marozeau, Storey and Blamey (2013) found anecdotal evidence for better prosocial and peer outcomes.

How then, might music training benefit aural communication skills? The core hypothesis is that music skills confer specific benefits for auditory perception, ultimately leading to improved outcomes for speech perception and communication. Referred to as the “musician advantage”, this thesis will consider three broad theories and their corresponding empirical evidence. The first assumption for why transfer effects may be possible between music training and speech perception is based on shared or overlapping cortical processes (Gfeller, 2016; Patel, 2011). There are a number of elements that are common between music and speech domains, such as pitch, rhythm—the grouping and meter of sound (Clarke, 1999), and timbre—the ‘texture’ of sound that allows us to differentiate between different instruments, even when they are played at the same pitch. An obvious example is how these elements are modulated to create emotional prosody (e.g. happy states have a rising pitch, a relatively fast rhythm, and a ‘bright’ timbre; while sad states have a falling pitch, a relatively slow rhythm, and a ‘dull’ timbre).

The second theory for consideration is that music training provides a perceptual advantage for speech perception (Besson, Chobert, & Marie, 2011; Parbery-clark, Strait, Anderson, Hittner, & Kraus, 2011). While the specific mechanisms for the enhancement of perceptual fine-tuning are not completely understood, music training is thought to improve both cognitive and perceptual skills such as auditory-attention and heightened auditory abilities such as the ability to extract acoustic regularities which contributes to better speech perception (Parbery-clark et al., 2011; Parbery-Clark, Strait, & Kraus, 2011).

The third theory is the expanded OPERA hypothesis that proposes music places high demands on cognitive and perceptual processes that overlap with speech perception. When music (and its corresponding demands) are emotionally rewarding, feature repetition, and receive focused attention, then the resulting changes in neural plasticity will lead to the enhancement of speech perception (Patel, 2014).

Finally, despite the lack of a strong body of evidence for the benefits of music training for children with hearing loss; musical interventions are commonly used as a complementary means of habilitation within Australian clinics and early intervention centres; presumably on the bases of enjoyment, compliance, and anecdotal evidence. As such, studies investigating the efficacy of music training for children with hearing loss are warranted—to establish evidence, explore mechanisms, and to ensure that time, efforts, and finances are appropriately managed.

Aims of the thesis

The overall aim of this thesis was to explore the benefits of a music training program for children with prelingual sensorineural hearing loss (SNHL). This was addressed by assessing the benefits of a 12-week music training program that consisted of group-based face-to-face music therapy sessions in conjunction with online music apps. Specific areas investigated were:

- Music outcomes such as pitch and timbre perception, and appreciation.
- Psychoacoustic outcomes such as spectral resolution.
- Potential transfer effects of musical skills to speech domains such as SIN and emotional prosody perception.
- Quality of life and psychosocial wellbeing.

Outline

This thesis presents a longitudinal study which is described within two manuscripts. **Chapter 2** provides a conceptual review of relevant topics within which this thesis is framed. **Chapter 3** provides the shared methodology (experimental design, participants, stimuli and tests, and procedures) that were used across both manuscripts. **Chapter 4** (Manuscript 1) examined the benefits of music training in respect to perceptual abilities. It was hypothesised the children with SNHL would improve in their

ability to perceive music, specifically pitch and timbre perception. In turn, pitch perception would likely yield a transfer effect to speech prosody. Additionally, it was anticipated that their appreciation of music would increase as a result of music participation. After training, statistically significant benefits were found for SIN, timbre perception, spectral resolution, and music appreciation. **Chapter 5** (Manuscript 2) considered the psychosocial and quality of life benefits that music training may provide. It was hypothesised that music training would result in benefits for psychosocial outcomes in which children with SNHL tend to have the poorest outcomes—peer relationships and prosocial measures. After training, statistically significant benefits were shown for internalising behaviours, along with factors associated with emotions and learning. **Chapter 6** provides a summary and conclusion of main findings of this thesis. It also discusses implications and recommendations for music training for children with hearing loss, as well as further research directions. **Appendix A** provides a documentation of ethical approvals and considerations. **Appendix B** provides a week-by-week documentation of the music training curriculum which consisted of face-to-face group-based music therapy and online take-home music apps. **Appendix C and D** provide supplementary statistical results, figures, and tables for Chapters 4 and 5 (Manuscripts 1 and 2), respectively.

*“I can’t understand why people are frightened of new ideas.
I’m frightened of the old ones.” — John Cage*

Chapter 2 — Conceptual review

Hearing loss

Hearing loss is the most common congenital sensory impairment, affecting around 3.3/1000 Australian children (Australian Hearing, 2017). As infants lack the capacity to understand language and alert their caregivers to any hearing difficulties—it can be particularly problematic (Northern & Downs, 2014). Childhood deafness is a multifaceted loss of hearing abilities that has a lifelong effect on the individual and their family beyond sensory perception. Studies report that children with hearing loss have poorer outcomes across a range of domains such as language, behaviour, and psychosocial wellbeing when compared with their TH peers (Flexer & Madell, 2014; Kral & O'Donoghue, 2010). Additionally, outcomes are further affected when the identification of hearing loss is late (after 6 months of age), which can lead to significantly poorer language development (Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998), as well as poorer health and educational outcomes (Wake, Hughes, Poulakis, Collins, & Rickards, 2004). Fortunately, the widespread adoption of universal neonatal hearing screening (UNHS) programs has significantly reduced the likelihood of a late identification, and allows for the facilitation of early intervention (Kral & O'Donoghue, 2010). This has led to improved language and communication outcomes for children with hearing loss; although our understanding of the long-term psychosocial benefits (due to a lack of studies) is inconclusive (Wong et al., 2017).

Hearing loss can arise from any number of dysfunctions at various anatomical structures involved in the auditory system. Hearing loss has traditionally been categorised as: conductive (a mechanical problem at the outer or middle ear, resulting in an obstruction of sound reaching the inner ear), sensorineural (damage or disorder to the inner ear, cochlear or auditory nerve), mixed (a combination of conductive and sensorineural losses), and central (dysfunction involving higher brain centres) (Smith & Gooi, 2018). Hearing loss onset is categorised as congenital or acquired, and aetiologies can be broadly stratified as genetic/hereditary or non-genetic/environmental (Smith, Bale, & White, 2005).

Another important consideration is the degree (or severity) of hearing loss. It should be noted these grades are inconsistent throughout the world, but the World Health Organization (2018) categorises losses for children as: slight/mild (26–40dB), moderate (31–60 dB), severe (61–80dB), profound (> 81 dB). A combination of adjacent terms is often used for losses that overlap or fall near bordering grades; for example, moderately-severe. Historically, the term deaf was applied to individuals

with profound hearing loss, while the term hard-of-hearing was applied for mild to severe losses; though such a demarcation is now generally meaningless as an identifier, with the introduction of effective early intervention leading to successful outcomes irrespective of the degree of hearing loss (Northern & Downs, 2014).

The establishment of a hearing loss is determined by a multi-disciplined team, with various techniques and diagnostics that measure hearing loss, as well as receptive and expressive language skills, and behaviour (Madell & Flexer, 2014). As such, a realistic definition as proposed by Northern and Downs (2014, p.28) emphasises communication and learning outcomes: “a significant hearing loss in a child is any degree of hearing that reduces intelligibility of speech to a degree inadequate for accurate interpretation or as to interfere with learning.”

Hearing devices

There are numerous technologies that may assist in the perception of sound, with each designed for a specific type and degree of hearing loss. As the studies reported in this thesis recruited children with moderate to profound sensorineural hearing loss (SNHL) as participants, a discussion on cochlear implants (CIs) and hearing aids (HAs) follows.

Cochlear implants

CIs are a technological innovation that represent the first example of a neural prosthesis which can effectively simulate a sensory organ (Macherey & Carlyon, 2014). The CI is a surgically implanted biomedical device that converts acoustic stimuli into meaningful patterns of electrical pulses that are interpreted by the central nervous system as the perception of sound (Rubinstein, 2004). As the CI allows the bypassing of missing or damaged hair cells, they are a suitable form of intervention for individuals with a severe-to-profound SNHL.

A CI system consists of the following: 1. A microphone to receive sound; 2. A speech processor that converts the microphone output into a stimuli for an array of electrodes implanted in the cochlea; 3. A connection across the skin that transmits power and stimuli; 4. An implanted receiver/stimulator that will receive, decode, and transfer signal to individual electrodes within an electrode array (Wilson & Dorman, 2008). A CI system can be seen in Figure 1, with numbers indicating the components previously listed.

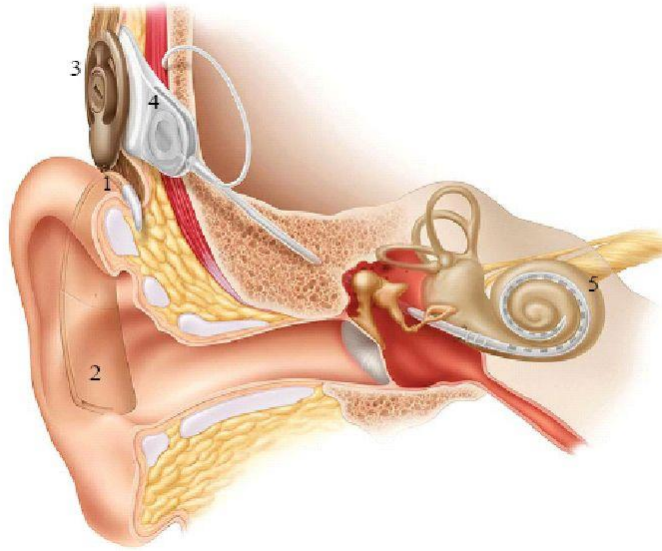


Figure 1. Cochlear implant system (Cowan, 2007)

All sounds can be sub-divided into temporal envelope (slow fluctuating waves) or fine-structure (fast fluctuating waves), as shown in Figure 2. There are numerous sound processing strategies that CIs employ to filter, extract, and encode acoustic information; but the dominant strategy is to use a multi-channel temporal-envelope based system such as continuous interleaved sampling (CIS) or advanced combination encoder (ACE).

CIS is a strategy that filters sound into a series of band-pass filters from approximately 100 to 8000 Hz; these filter-bands are slightly overlapping and the bandwidths also increase with higher frequencies (Wouters, McDermott, & Francart, 2015). The channels (filter outputs) are each assigned to at least one electrode, and thus allows for a crude tonotopical representation (Wouters et al., 2015). Between 4 and 22 pulse trains at 500 to 2000 pulses per second (pps) for each channel are interleaved, resulting in non-simultaneous pulses across electrodes (Wilson & Dorman, 2009).

ACE is the sound-processing scheme developed by Cochlear Ltd, and is fundamentally similar to CIS, but only a subset of channels (typically 8) that have the highest amplitude are selected during any stimulation cycle (Wilson & Dorman, 2009). This results in a reduction of channels presented, a general decrease of masking or noise effects, and an enhancement of spectral features; due primarily to a reduction of cross-talk and distortion between neighbouring electrodes (Friesen, Shannon, Başkent, & Wang, 2001).

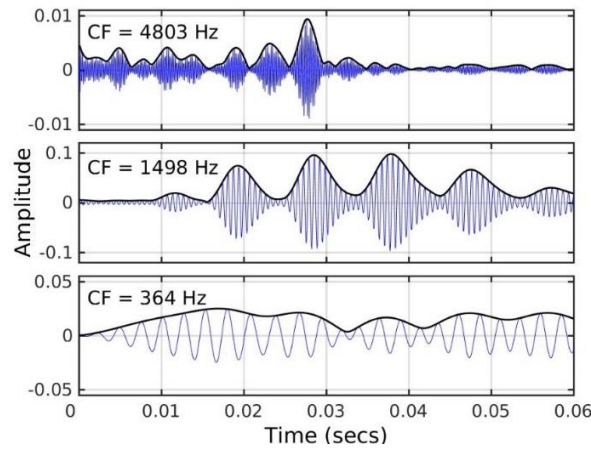


Figure 2. Temporal envelopes (black lines) superimposed on temporal fine structures (blue lines).

Hearing aids

The fundamental function of a HA is to amplify sound signals. In basic form, HAs comprise a microphone, an amplifier, a receiver (or miniature loudspeaker), a system to couple the signal to the ear canal, and a battery to power the amplifier. The microphone receives and converts acoustic signals into an electrical signal, which is then amplified to a designated prescribed formula (e.g. NAL-NL2), determined by the level of hearing loss at designated frequencies. This signal undergoes some digital processing such as compression, noise reduction, or limiting; and finally, the signal is transduced back into acoustic form by the receiver down the ear canal (Dillon, 2012; Northern & Downs, 2014).

Modern HAs have advanced considerably since their early, cumbersome designs that often required the user to wear them on their body. The continual advancement for the miniaturisation of HAs is driven by the demand to create less conspicuous devices, although this often results in a trade-off between processing performance, size, and battery life (Dillon, 2012). There are many HA designs and styles, and a common method of categorising them is in terms of how they are worn. Worldwide, behind-the-ear (BTE) designs are the most popular design which can be seen in Figure 3. They are also the preferred type for children as they can accommodate a larger battery (reducing the need to change batteries), are more durable, and offer tamper-resistant features such as locking battery doors when compared against more discrete designs such as in-the-ear (ITE) HAs (McCreery & Walker, 2017). The majority of HAs in operation are digitally-based, allowing for myriad processing capabilities, offering flexibility for a range of automatic or personal adjustments (Northern & Downs, 2014). Manufacturers tend to price-tier their models based on the number of features available, although there is no evidence of additional benefit (McCreery & Walker, 2017). Irrespective of design and additional features, the basic function of all HAs are the same—amplification.

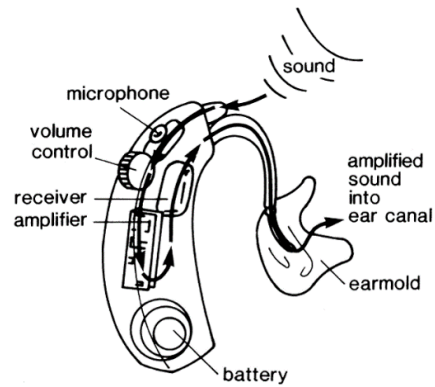


Figure 3. Behind-the-ear hearing aid (Northern & Downs, 2014)

Speech perception

Speech is a complex waveform that features multiple, fluctuating cues such as spectrum, pitch, and amplitude that can operate both simultaneously and independently (Greenberg & Ainsworth, 2004). Speech perception requires the mapping of continuous acoustic changes, and the categorisation of this into discrete perceptual units (Bidelman, Moreno, & Alain, 2013). This categorisation of sounds is guided by learning and language experience, and provides an illustrative example of how perception is modulated by expectation and experience (Greenberg & Ainsworth, 2004). Thus, our ability to make sense of multiple cues is remarkable, and the ability to inhibit non-relevant acoustic stimuli while integrating speech cues allows for meaning to occur when information is transferred from the auditory system to the central nervous system. A difference in any single element essentially leads to a unique phoneme, creating a difference in sound, word and meaning such as *pug* /peg/ and *bug* /beg/.

Australian English (AusE) is a regional dialect of English, and like all languages, is a system of phonemes which can be categorised as being a vowel or consonant. The main articulatory difference is that consonants are produced with a narrow constriction relative to vowels, which leads to the simple perceptual consequence that vowels are louder than consonants (Cox & Fletcher, 2017). Vowel articulations are comparatively simple compared to consonants, and every vowel can be described by its first two formants (the resulting resonant characteristics of the vocal tract and articulators such as the jaw and tongue), as shown in Table 1. Consonants however, require more complex description that include the place of articulation (the location of the major articulator/s), manner of articulation (the degree of constriction, use of an oral or nasal pathway, and a description of the type of airflow), and voicing (the presence or absence of vocal fold vibration) (Cox & Fletcher, 2017); see Figure 4.

While efficient perception of speech tends to require one or two primary cues for identification, speech features highly redundant and robust cues (Mattys, Davis, Bradlow, & Scott, 2012). Some examples of these design features include the importance of formant peaks that are far less susceptible to masking than valleys (Assmann & Summerfield, 2004); the use of periodicity to assist in the grouping of perceptual units (Bird & Darwin, 1998); and the use of fundamental frequency modulations (such as prosody) that assist in separating word boundaries (Friederici & Wessels, 1993).

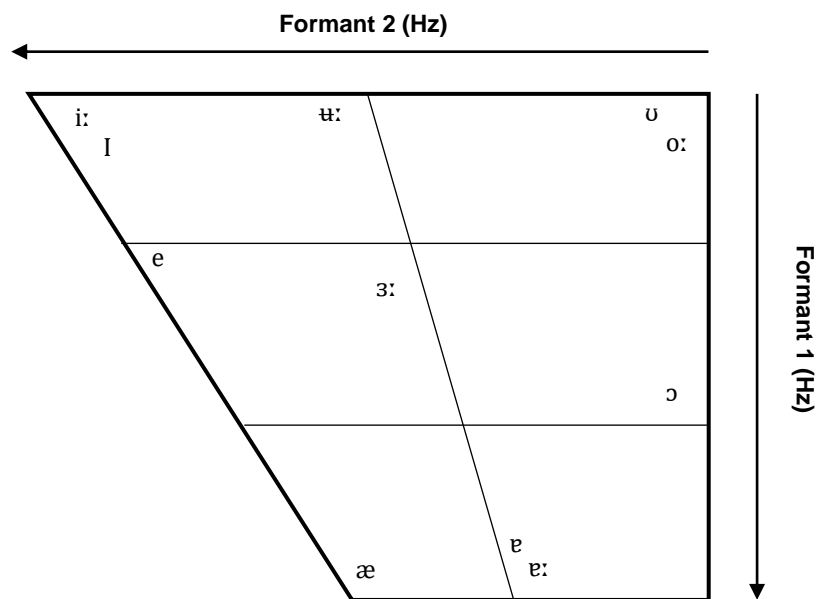


Figure 4. Australian English vowel space schematic.

Table 1. Australian English consonants by place and manner of articulation, and voicing feature.

	Bilabial	Labio-dental	Dental	Alveolar	Post-alveolar	Palatal	Velar	Glottal
Plosive	p b		t d			k g		
Affricate					tʃ dʒ			
Nasal	m			n			ŋ	
Fricative		f v	θ ð	s z	ʃ ʒ			h
Approximant				r		j	w	
Lateral approximant				l				

Prosody is broadly the use of suprasegmental cues such as duration, fundamental frequency and intensity (Wagner & Watson, 2010), for the purposes of modulating meaning. For example, the

distinction between emotions can be described in terms of the relative difference between each of these cues. Relative to happiness, sadness will be longer in duration, with intonation curves moving in a downward direction, with less intensity. Prosodic cues play an additional role in the development of children's language development, as they assist in the separation of boundaries relevant for syntax (Soderstrom, Seidl, Kemler Nelson, & Jusczyk, 2003); and the use of infant-directed speech (which exaggerates prosodic cues) is not only used to assist with language development, but is also preferred by infants (Jusczyk, Cutler, & Redanz, 1993). Derived from the Ancient Greek term *prosōidia* that loosely translates as: "to sing with", it is perhaps unsurprising that musicians are particularly adept with the perception of prosodic cues (Hausen, Torppa, Salmela, Vainio, & Särkämö, 2013).

Music perception

Perceptually, music can be broadly organised into elements of pitch, timbre, rhythm, and intensity. When combined, their interaction forms the foundation that is music. Limb and Roy (Limb & Roy, 2014) emphasized that music is the most complex auditory stimulus in existence. Although such broad-sweeping statements are difficult to determine, it is important to appreciate that both music and speech are complex, and while they share many acoustic cues and features—they are also ultimately different.

Pitch

In many respects, pitch is an abstract spatial concept that has an equally abstract technical definition: "that attribute of auditory sensation by which sounds are ordered on the scale used for melody" (ASA, 2016a). As the perception of pitch depends primarily upon fundamental frequency (Houtsma, 1997), a more useful working definition is perhaps: the perceptual correlate that relates closely to fundamental frequency or periodicity, which can be ordered from low to high (Oxenham, 2017; Trainor & Unrau, 2012). Pitch is a fundamental auditory sensation, and when played sequentially, forms the basis for melody.

There are two broad theoretical accounts of pitch representation: Place and Time theories. Place theories are based on the mechanics of the cochlea—specifically, the tonotopic organisation of the basilar membrane that results in greater sensitivity of low frequency sounds at the apical end, and high frequency sounds at the basal end, due to the passive characteristics of the basilar membrane (i.e. its mass and stiffness is gradient-dependent) (Hartmann, 1996). The displacement of the basilar membrane

results in specific activation of neurons, and crucially, this tonotopic firing of neurons is maintained through ascending levels of the auditory system (Hartmann, 1996). This separation of frequencies is further enhanced by an active component that provides sharp tuning due to the action of the outer hair cells (Dallos et al., 2008). Time theories are based on findings that show that action potentials have a tendency to phase lock—this neural synchrony allows for a precise relationship between action potential firing rate and frequency (Hartmann, 1996; Oxenham, 2017; Rose, Brugge, Anderson, & Hind, 1967).

Physiologically, humans have been noted as having sharp cochlear tuning that allows for the fine separation of sounds on the basis of frequency components (Oxenham, 2017). However, the implications of this are not fully understood. Sharp tuning likely contributes to pitch perception; and while it may assist in communication and speech perception, this is likely in terms of redundancy or of benefit in more complex listening environments, as speech is robust to spectral degradation (Shannon, Zeng, & Kamath, 1995).

Timbre

Timbre remains one of the most poorly understood acoustic features, and there is no current theory or framework that is widely accepted (Town & Bizley, 2013), although multidimensional scale (MDS) models are often used to measure dis/similarities between sounds in reference to a matrix (Grey, 1977). McAdams and Bregman (1979, p.34) go so far as to define what timbre isn't: "timbre tends to be the psychoacoustician's multidimensional waste-basket category for everything that cannot be labelled pitch or loudness."

ASA (2016b) defines timbre as: "that multidimensional attribute of auditory sensation which enables a listener to judge that two non-identical sounds, similarly presented and having the same loudness, pitch, spatial location, and duration, are dissimilar. Timbre is related to sound quality, often specified by qualitative adjectives (e.g., bright or dull)." Broadly, timbre is primarily based on acoustic characteristics such as frequency spectrum and temporal envelope (particularly how they vary over time) (McDermott, 2004). Perceptually, differences in sound quality allow for distinctions to be made when comparing a piano to a violin. While timbre is often understood from a music perspective (in terms of theory and measures), the definition can be generalised to auditory perception (and speech) in general.

Rhythm

Rhythm refers to two broad aspects within music—grouping and meter which consider time spans and time points respectively (Clarke, 1999). Lerdahl and Jackendoff (1983) provide a systematic theoretical account; grouping considers a hierarchical approach to the segmentation of music from small groups of notes, to grouping that would consist of phrases, bars, or sections, to the entirety of the composition. Meter is the regular alternation of stress, such as stressed (strong) or unstressed (weak) elements which are perceived as the underlying periodicity or pulse (Lerdahl & Jackendoff, 1983).

Entrainment is a rhythmic behaviour that refers to how independent systems interact (Clayton, 2012). For example, the coordination of two or more individuals in group-based singing or dancing. Entrainment is not unique to music, and is implicated with motor skills; for example, movement is made more efficient when walking occurs at a consistent rhythm. Interestingly, multiple studies indicate that social engagement of individuals tend to become entrained (Oullier, de Guzman, Jantzen, Lagarde, & Scott Kelso, 2008; Richardson, Marsh, Isenhower, Goodman, & Schmidt, 2007), which likely serves to improve social cohesion and bonding that benefits communication and community (Oullier et al., 2008).

Perceptual outcomes with sensorineural hearing loss

The perceptual impact of SNHL can be broadly categorised as a decrease in: audibility, dynamic range, and frequency resolution (Dillon, 2012; Oxenham, 2017). Thus, relative to their TH peers, children with SNHL receive an auditory signal that is elevated (amplified), compressed, with poorer frequency resolution. The overall effect is that children with SNHL have difficulties perceiving important perceptual cues—particularly in more challenging listening environments.

Audibility

At the basic perceptual level of speech perception, hearing loss results in a reduction of cues (salient and redundant) that help identify and separate phonemes (Boothroyd, 1984). For example, /i:/ can be described as a high front vowel, while /u/ can be described as a high back vowel. The distinguishing feature is the second formant. If we introduce a hypothetical hearing loss around the second formant (approximately 1000 Hz for a male speaker), the distinction between these two vowels is harder to ascertain, leading to perceptual confusions. Additionally, in daily conversation, voiceless consonants

such as /s/ often fall below a hearing-impaired child's hearing threshold. As a commonly recurring phoneme, /s/ is particularly important for various morphological rules such as plural -s and possessive -s (Koehlinger, Van Horne, & Moeller, 2013). Hence, fundamental issues of audibility and perceptual access can lead to morphological language delays (Koehlinger, Van Horne, Oleson, McCreery, & Moeller, 2015; Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007).

Overall, children with SNHL require better signal-to-noise ratios than adults, and their access to sound is tied to their language development (Dillon, Ching, & Golding, 2014). Essentially—sound must reach the brain for auditory-based learning to occur and take shape. For the majority of children with hearing loss that receive effective early intervention, the perception of speech in quiet environments is generally quite good (Blamey et al., 2001), but poor in noisier situations due to the disruption and masking of speech cues (Stickney, Zeng, Litovsky, & Assmann, 2004). This is problematic, as modern industrial society is inherently noisy in many contexts of daily communication; but importantly for children—schools and classrooms frequently exceed noise and reverberation guidelines, thus having a significant impact on learning, development, and educational outcomes (Mealings, Demuth, Buchholz, & Dillon, 2015; Schafer & Thibodeau, 2006).

Dynamic range

Unfortunately, while audibility is the primary problem for the degradation of auditory input, the amount of amplification that can be provided by a HA is limited by a reduction of dynamic range. The increase in hearing threshold is not matched with an increase in threshold of loudness (which remains unchanged) so that low-intensity sounds are inaudible and high-intensity sounds remain the same (Dillon, 2012). An additional consequence is any loudness gain is increased exponentially when compared to TH peers (Steinberg & Gardner, 1937). Thus, the use of compression (the automatic adjustment of amplitude dependent on input) is required to ensure that audibility from a wide range of sources (in terms of frequency and intensity) is maintained at a consistent, comfortable, audible level. It is particularly important for children with hearing loss, as they lack the skill and dexterity to make manual volume adjustments (Northern & Downs, 2014). One of the challenges is to ensure that spectro-temporal changes are maintained for the integrity of the overall signal (Plomp, 1994). For example, plosives such as /p/ or fricative /s/ are disproportionately affected by fast acting compressors than vowels are. Fortunately, the use of multichannel compressors is common on modern HAs, allowing for

the amount of compression to be dependent on frequency specific responses, as well syllabic/phonemic level compression occurring with variable attack times (Dillon, 2012).

The dynamic range fitting goals for CIs are no different to HAs; the processor should be set to provide audibility across a range of listening scenarios; however, dynamic range terms are differentiated. The range of acoustic input level is defined as the input dynamic range (IDR) that is typically set between 30 and 80 dB, whereas the electrical dynamic range is calculated as the difference between T-levels (hearing threshold) and C-levels (maximum comfort threshold). Electrical dynamic range is significantly lower than HA users, and has been reported at 7.4 dB (± 2.3) for 5 year olds (Incerti et al., 2017); in comparison, the IDR for TH adults is approximately 120 dB, although this is reduced to approximately 50 dB for speech input (Zeng et al., 2002). Loizou, Dorman, and Fitzke (2000) reported that a wide dynamic range was required for accurate vowel perception, while only a small dynamic range was required for accurate consonant perception, primarily due to the loss of spectral contrast (i.e. difficulty identifying formants).

Limited dynamic range has a considerable effect on music compositions, that often rely on the dynamic use of intensity to convey expression and emotion, which also affects prosody. Limited dynamic range contributes to decreases in overall music quality appraisals (at least in adult studies), as well as a flattening of the spectral shape that allows for separation of sounds on the basis of timbre perception (Limb & Roy, 2014).

Frequency resolution

A normal functioning auditory system has exceptionally sharp cochlear tuning that allows for the separation of sounds based on frequency components (Oxenham, 2017). Unfortunately, neither HAs or CIs can restore cochlear tuning to this level. In the case of individuals with SNHL, frequency resolution (or selectivity) is much poorer (C. W. Turner, Chi, & Flock, 1999). Important spectral cues such as formants that help determine vowels are significantly degraded due to flatter cochlear tuning, leading to perceptual confusions (Dillon, 2012). Pitch perception is also poorer, again due to issues of frequency selectivity and difficulties resolving harmonics which have cascading consequences on speech perception including SIN and prosodic perception, due to the inability to separate sounds that arrive at a similar frequency region (Bernstein & Oxenham, 2006).

External to problems associated with cochlear tuning; the delivery of pitch-based stimuli via CIs warrants specific discussion. CIs deliver information through a crude tonotopic representation via the

electrode array that consists of between 12 and 24 electrodes that essentially replace 3500 inner hair-cells in a normal functioning cochlea (Oxenham, 2017). As such, this large differential means the electrodes simply cannot provide the fine-grained specificity that is normally delivered (Limb & Roy, 2014). Similar to Place and Time theories in normal pitch perception; pitch is delivered in two ways through a CI. Place pitch uses the tonotopical arrangement of the electrode array with the basal electrodes responding to high frequencies, while electrodes in the apical location respond to lower frequencies. However, most implant arrays do not reach the most apical turns of a cochlea, resulting in poorer representation of pitch at lower frequencies (Limb & Roy, 2014). Additionally, the placement of any electrode relative to its ideal location within the cochlea is prone to being mismatched, although the brain is generally able to make compensations and adapt, within limits (Landsberger, Svrakic, Roland, & Svirsky, 2015). Finally, the stimulation of an electrode can lead to the unintended excitation of adjacent electrodes, and is likely the most significant limitation for frequency resolution (Abbas, Hughes, Brown, Miller, & South, 2004). Known as channel or spatial interaction, the spread of excitation leads to a general reduction perceptual cues, and findings by Crew and Galvin (2012) indicate that greater channel interactions lead to poorer pitch perception due to a disruption of the tonotopic specificity.

Paediatric perspectives

The vast majority of evidence in the literature for the perceptual outcomes of SNHL is in relation to adults with post-lingual hearing loss. The most significant difference to consider in children with SNHL is that they are reliant on auditory input and stimulation to shape and develop their auditory system. Importantly, Kral, Dorman and Wilson (2019) suggest that passive exposure to auditory stimuli is not as effective, and possibly not sufficient for the development of language competence. On the other hand, active participation and social feedback help facilitate language development (Goldstein & Schwade, 2008). Children have maximum neuroplasticity between 3.5–4.0 years of age (underscoring the importance of effective early intervention) which gradually decreases a function of age, with the period of greatest sensitivity ending at around 6.5–7.0 years of age (Kral & Sharma, 2012).

The developing auditory system can be interpreted as a naïve system, with reduce sensitivity to a variety of auditory cues and features (Kral & Sharma, 2012). There are broadly two milestones for the effective development of the auditory system. The first stage is the ability to recognise sounds, such as phonemes, the bark of a dog, the ring of a bell, and so on; the second stage goes beyond the level of

simple recognition, in which the sounds are now conceptualised as auditory objects (Kral & O'Donoghue, 2010). Importantly, this early stage of auditory development is influenced by other non-auditory regions such as the motor cortex (Murakami, Kell, Restle, Ugawa, & Ziemann, 2015). In the context of the “connectome model of deafness” by Kral, Kronenberger, Pisoni, & O'Donoghue (2016) that posits the importance of neural networks and multiple levels of representation; a complete understanding of auditory development is thus reliant on the development of all motor, sensory and cognitive functions.

Music training

Music training is a multisensory activity that typically involves the broad activation of auditory, visual, cognitive, and motor domains. For example, consider the act of playing a traditional solo composition on a violin. The musician begins by reading a notation system which is translated into a manual and dexterous sequence of actions that require the asynchronous movement of one hand (and its corresponding fingers to determine the pitch) while the other hand generates sound by drawing the bow. While this is happening, it is the auditory system that provides feedback as to whether or not the correct sequence has been played. In the context of learning, music places specific demands such as fine-grained perception and motor control that is atypical to other everyday activities (Herholz & Zatorre, 2012). Additionally, the social aspect to music performance should not be overlooked. Group-based musical activities can help facilitate group cohesion, cooperative behaviour, and foster pro-social attitudes with peers and family (Kirschner Sebastian, Tomasello, Kirschner, & Tomasello, 2010; Rickard et al., 2013; Williams, Barrett, Welch, Abad, & Broughton, 2015). The topic of music is immense and complex. Given the nature of this thesis is the (re)habilitative aspects of music training for children with hearing loss; this review will focus on transfer effects—how music training can enhance speech perception and psychosocial skills, with theoretical and empirical considerations from the literature with a distinct focus on SIN.

The musician advantage

The musician advantage (also referred to as the musician effect/edge) suggests that music training confers benefits for auditory processing and perception, particularly in more challenging situations such as SIN (Fuller, Galvin, Maat, Free, & Başkent, 2014). There are inconsistencies regarding the classification of a musician, though most studies use the following criteria: having at least 8–10 years of

continuous training on their principal instrument, having commenced their training as a child prior to the age of 7–9, and ongoing regular practice with their instrument. There is a general bias that emphasises instrumental training (as opposed to singing), and general consensus that the classification of a musician requires early childhood experience, and ongoing consistency with their practice. Unusually, most studies do not make a distinction between a musician that trains in a pitched instrument (e.g. a guitar or piano) or a percussive instrument (e.g. drums), if they are a soloist or part of an ensemble. At face-value, it would seem such differences would yield a wide difference in skills and advantages. Although Slater and Kraus (2016) found evidence that percussionists, vocalists, and non-musicians were more likely to have their SIN perception correlating with their rhythmic ability, most studies tend to conceptualise musicians as a homogenous population.

The range of SIN tasks is also inconsistent between studies, with no gold-standard measure for SIN. Differences in stimuli with the use of words, sentences, prosody, natural or synthetic utterances; as well as differences in delivery through headphones or loudspeaker arrangement (single or spatial configurations); along with differences in masker type are prevalent. Nonetheless, in spite of such variability, a review by Coffey et al. (2017) found that 18 out of the 20 studies that met their inclusion criteria supported SIN benefits for musicians. As such, the musician advantage is likely the result of multiple mechanisms associated with music training (Coffey et al., 2017). As children with SNHL perform significantly poorer than their typical hearing peers for SIN tasks (Caldwell & Nittrouer, 2013), the exploration of the music training for this population is of great interest for (re)habilitation purposes.

The majority of studies exploring the musician advantage have been cross-sectional in design, comparing the performance of musicians to non-musicians. The cross-sectional approach allows for 3 broad conclusions: 1. Individuals with natural advantages in auditory and cognitive processes relevant for both speech and music tasks are likely to be attracted to music training; 2. Music training leads to neuroplastic changes that benefits speech processing, or 3. A combination of both considerations (Swaminathan et al., 2015). Parbery-Clark et al. (2012) found that musicians had better performance in discriminating synthesised plosives such as /ba/ from /ga/ that were differentiated on the basis of the second formant. Hence, musicians had some advantage for the representation in formant frequencies, and this was demonstrated at both a behavioural and subcortical level (with greater neural differentiation as measured by auditory brainstem responses). Beyond the phoneme level, they also found a statistically significant difference on the Quick Speech-in-Noise Test (QuickSIN). However, the actual benefit as measured by participants SRT or SNR was not reported. Swaminathan et al., (2015) explored the classic “cocktail party problem” by manipulating both the spatial location and intelligibility

of the masking sources. The advantage the musician's had was substantial—an improvement of ~6 dB SNR compared to their non-musician counterparts, indicating a particularly enhanced ability for SIN when presented in spatialized conditions. A study by Başkent and Gaudrain (2016) manipulated the vocal configurations (fundamental frequency— F_0 and vocal tract length) between target and masker speech. It was hypothesised that musicians would have an advantage over non-musicians for this task, due primarily to the better perception of pitch cues. However, while the musicians performed better than the non-musicians as expected, but the advantage was not associated with pitch performance. Başkent and Gaudrain (2016) suggested that a broader range of abilities such as better speech segregation, rhythm, or cognition was likely the mechanism for this advantage. Similarly, a study by Madeson, Whiteford and Oxenham (2017) showed that while musician's pitch discrimination abilities were superior to non-musicians, there was no correlation between their pitch perception and 2-talker babble SIN, and concluded pitch discrimination did not confer any benefit for speech perception. Well-designed and with a relatively large sample size of 30 musicians and 30 non-musicians, unlike the aforementioned studies, the authors ultimately found there was no evidence that musical skills conferred any speech perception benefits for young adult musicians.

Cross-sectional designs do not allow for inferences of causality, and the potential effect that individuals with exceptional auditory abilities are predisposed towards musical experiences is a significant limitation. These designs are also inappropriate for children, given the classification of a musician requires the criterion of extensive long-term training. As such, randomised, longitudinal paradigms with long-term follow-up are ideal for developing evidence of causality. A recent study by Slater et al. (2015) provided the most compelling evidence that the musician advantage is the result of music training, and not any prior disposition or abilities. This study followed a cohort of elementary school children over the course of 2-years, and assessed their abilities to perceive SIN before and after participation in the Harmony Project—a non-profit organisation that provides free music education to children in the gang reduction zones of Los Angeles. Using a waitlisted design, their cohort consisted of 38 children that were split into two groups. Group 1 was waitlisted, and acted as the control group during the first year, while Group 2 completed 1 year of training; thus when Group 1 completed training, Group 2 had completed 2-years of training. This design allowed for a within and between-subjects analysis. Importantly, it also provided a baseline measure of maturational changes which was important considering the 2-year length of training, as well as changes resulting from the music intervention. The music training commenced with introductory musicianship classes, followed by instrumental classes. The musicianship classes met for 1-hour, twice a week, and the learning objectives and activities were

broadly based around: rhythm, pitch, performance, improvisation and composition, musical awareness, musical terminology, and orchestral instrumentation. Students would generally spend 1-year in the musicianship class, dependent upon their ability and instrument availability, before progressing onto instrumental classes that consisted of at least 4 hours of training per week with an ensemble. Slater et al. (2015) found that SIN improved significantly, with a mean improvement to participant's SNRs of -2.1 dB. Importantly, this study helped validate the musician advantage beyond cross-sectional design methodologies, with an ecologically-valid community-based training program.

The evidence of the musician advantage for individuals with hearing loss in the context of SIN is non-existent, but a small number of studies have explored transfer effects for other speech perception tasks. Lo, McMahon, Looi, and Thompson (2015) trained 16 adult CI recipients in a computer-based melodic contour training program over a 6-week duration. Post-training results indicated that participants improved in their ability to perceive prosodic cues in question/statement contexts, as well as consonant perception, though no benefit was shown when these were disrupted with the addition of four-talker babble (4TB), suggesting that these embedded pitch cues are particularly susceptible to degradation by noise. In another melodic contour training study for Mandarin-speaking CI recipients aged between 5–9 years, improvements from an 8-week melodic contour training program led to improved lexical tone recognition (Cheng et al., 2018). It should be noted that while melodic contour training certainly contains aspects of music training, the demands are significantly different to broader music training such as instrumental or community-based training. These two studies utilised a very similar paradigm, which involved the use of 9, 5-note melodic contour configurations. On the other hand, instrumental training features a much greater variety of sequences, and thus broader demands. Good et al. (2017) conducted a 6-month, weekly piano training program for 18 CI recipients aged between 6–15 years of age. Half the children were assigned to music training and the other half to visual art training. Improvements were noted for both musical and emotional prosody perception, but only for the musically trained children. Overall, these studies provide evidence for an encouraging transfer effect between music-based training and prosodic/intonation perception, with the likely mechanism being an improvement in pitch-based abilities.

Mechanisms for the musician advantage

There are three broad mechanisms that suggest why music training may transfer to benefits for speech perception. The first mechanism suggests overlap as its key component and that there are common links between music and speech. This overlap was initially proposed in the context of acoustic overlap. For example, Patel (2011) argued that periodicity is an important cue for both speech and music, but the neural representation of pitch will be realised differently, depending on its context (i.e. linguistic or musical). Additionally, Patel (2011) argued that a distinction needs to be made between the actual processing of a perceptual attribute (such as pitch)—which may show a hemispheric bias (Zatorre & Gandour, 2008), and to its acoustic features (such as periodicity)—which is likely to be encoded similarly, irrespective of context. As such, the basic underlying assumption is that the basic acoustic features present in both speech and music lead to overlapping neural networks.

Peretz, Vuvan, Lagrois, and Armony (2015) argued for a clarification of the terms neural overlap and neural sharing. They suggested that while most brain regions are involved in shared processes that are interpreted as overlapping, the actual neural processes that occur within, or adjacent to regions of interest may be neurally distinct and separate. A review of various studies utilising a range of neuroimaging techniques and found both speech and music had considerable overlapping activation patterns, but there were also unique patterns that were distinct for both domains. As such, the interpretation of neural overlap and/or sharing is simultaneously one of shared overlap and distinct separation, and remains an ongoing area of investigation.

The second mechanism is that music training fine-tunes the auditory system to provide perceptual advantages for speech perception. Parbery-Clark et al. (2011) hypothesised that musicians would have better SIN perception, due to a greater sensitivity to acoustic regularities (such as periodicity). The results indicated that musicians had better SIN perception than non-musicians, and this was driven by a greater representation of FO—an accessible cue in the target speech, which was not available in the more variable and irregular speech-shaped background noise. Additionally, this sensitivity to regularity could also be fine-tuned and developed through experience, such that increased subcortical responses will occur for relevant signals, while irrelevant ones are suppressed. Parbery-Clark et al. (2011) further argue that another advantage of improved sensitivity to acoustic regularity is that it enables the formation of templates, based upon a system of pattern recognition. This in turn allows for more rapid sensory processing (Haenschel, Vernon, Dwivedi, Gruzlier, & Baldeweg, 2005). Although these findings were not explicitly referred to as statistical learning, this conclusion was also supported

by a study that indicated musicians have superior performance in auditory statistical learning than non-musicians, using an embedded triplet task (Mandikal Vasuki, Sharma, Demuth, & Arciuli, 2016).

The third theory attempts to reconcile these two mechanisms, by broadly considering the sensory/acoustic and cognitive processes to provide a framework for the musician advantage. The original OPERA hypothesis posits that 5 factors are required for music training to drive neuro-plastic benefits for speech perception (Patel, 2011). These are: Overlap, there are shared acoustic features between speech and music that also overlap in neural networks; Precision, the processing demands of music are greater than that of speech; Emotion, music elicits a strong positive emotion that promotes the development of these neural pathways; Repetition, the musical activities are performed frequently; Attention, the musical activities are associated with focused attention. The expanded OPERA hypothesis makes only one significant addition, in that the emphasis is moved away from a focus on acoustic and sensory processing, and also considers the importance of cognition in each of the 5 factors (Patel, 2014).

Summary

In summary, reduced audibility, dynamic range, and frequency resolution contribute to difficulties with intelligibility of speech and music. SNHL is ultimately the combination of each of these factors that results in poorer perceptual outcomes for children with hearing loss compared to their TH peers. A good outcome is challenging, as it must account for the intelligibility of soft sounds, a tolerance for loud sounds, the comfort of day-to-day conversation, accuracy of pitch representation, and in a wide range of listening environments and contexts. Recent attention has been turned towards encouraging findings from music training studies. Though considerable efforts need to be undertaken to build upon the body of evidence, results from a number of independent studies support the musician advantage as a compelling and cautious source of optimism within the field of auditory (re)habilitation.

Chapter 3 — Methodology

Two manuscripts (to be submitted) form the basis of this thesis, and the following section describes the shared methodology. Manuscript 1: “Music training for children with hearing loss improves music and speech outcomes”, explored perceptual benefits; while Manuscript 2: “Music training for children with hearing loss: quality of life and psychosocial outcomes”, examined psychosocial improvements. Note that while the design and participants were shared, the materials were manuscript specific (Manuscript 1: perceptual materials; Manuscript 2: questionnaires). Supplementary materials consisting of additional figures and tables for statistical analyses in Manuscript 1 and 2 can be found in Appendix C and D respectively.

Participants

Two groups of participants were tested in the study, stratified by hearing status (children with SNHL and TH). One group consisted of 14 children (7 female, 7 male) with prelingual bilateral moderate-to-profound SNHL (8 bilateral CI, 4 bimodal, 2 bilateral HA) that ranged in age from 6.1–9.2 years ($M = 7.5$, $SD = 1.1$) when measured at baseline 2. Inclusion criteria for children with SNHL included prelingual (aiding or implantation < 3.5 years), bilateral SNHL with moderate-to-profound thresholds. Most children with SNHL (9/14) were enrolled in mainstream school settings. Relevant demographic data for children with SNHL can be found in Table 2.

For comparative purposes, 16 TH children (7 female, 9 male) that ranged in age from 6.3–8.7 years ($M = 7.6$, $SD = 0.8$) were also included. There was no significant difference in chronological age between children with SNHL and TH, $t(25) = 0.86$, $p = .400$. At the start of each session, the TH children underwent pure tone audiometric testing to confirm hearing thresholds (0.25 to 8 kHz ≤ 20 dB HL). All participants were native Australian English speakers. Exclusion criteria for all participants included any diagnosed psychological or developmental disorder. Relevant demographic data for TH children can be found in Table 3.

Participant recruitment was multi-faceted to encourage a broad sample of participants and reduce sampling bias. Direct invitations were sent via Australian Hearing clinics and the Sydney Cochlear Implant Centre (SCIC) to families within NSW fitting the inclusion criteria, and flyers were distributed to clinics and hearing/deafness groups for distribution in newsletters and social media

outlets. Parental written consent and participant assent were obtained prior to commencement of testing, and approval for this study was granted by the Macquarie University Human Research Ethics Committee (Medical Sciences); reference: 5201600081.

Experimental design

Data collection spanned approximately 9 months, using a longitudinal waitlist design. After an initial test session (baseline 1), children with SNHL were pseudo-randomly assigned to commence music training in March 2017 (Group 1) or placed in the waitlisted group (Group 2) that commenced music training 12 weeks later. Pseudo-random assignment was due to the lengthy time-commitment this study placed on families (i.e. if specific dates were not suitable for participation they could opt for the other group). For all perceptual measures, double baseline testing occurred, separated by 1-week for Group 1, and separated by 12-weeks for Group 2. The advantage of this experimental design is that it allowed for an assessment of test-retest reliability, a baseline measure of natural development and maturation over a 12-week period for the waitlisted group, and had the additional benefit of maximising statistical power by not having to split the cohort into a training and control group. After the completion of double-baselines; participants were tested after 6-weeks of music training (mid); after completion of the full 12-weeks of music training (post); and finally, 12-weeks after training was completed to measure retention (follow-up). From the group of 14 children with SNHL, 11 commenced the music training, while the remaining 3 only completed the 12-week double baseline measures. Of the 11 children with SNHL that commenced music training, 9 completed all testing sessions, 1 withdrew after the mid-point due to a surgical operation, and 1 family left the country at the follow-up stage.

As the time course for psychosocial benefits was expected to be longer-term and not subject to learning effects, questionnaires were not disseminated at the mid test-point; additionally, Group 1 did not complete questionnaires at baseline 1. An additional cohort of age-matched TH children was included as a comparison group; they did not receive music training and did not complete questionnaires at baseline 1 as they were only utilised to indicate the broad difference between children with SNHL and TH children. While there are published normative data for the majority of questionnaires used; the decision to utilise a comparison group was to have a more robust comparison that would account for age, cultural and linguistic differences, and testing protocol. An overview of this design can be seen in Figure 5.

Table 2. Demographic information for children with SNHL.

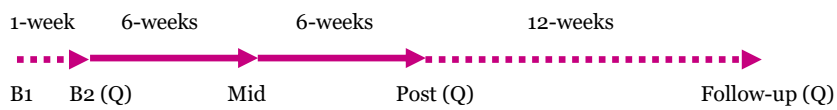
ID	Group	Age/ Hearing age (baseline 2)	Age at first fitting/ implantation	Sex	Formal music experience*	Degree of hearing loss	Device configuration	Device	Processor	Strategy	Active electrodes	Aetiology	Schooling
HL1	1	6.3/6.0	0.3	F	0	L: Profound R: Profound	CI	L: CI422 (SRA) R: CI522 (SRA)	L: CP910 R: CP910	L: ACE R: ACE	L: 22 R: 22	Unknown	Specialised
HL3	1	8.3/7	1.3	M	3.7	L: Profound R: Profound	CI	L: CI24RE (ST) R: CI522 (ST)	L: CP810 R: CP810	L: ACE R: ACE	L: 7 R: 22	Pneumococcal meningitis	Mainstream
HL5	1	6.1/3.1	3.0	F	0.7	L: Profound R: Moderate	Bimodal	L: CI24RE (ST) R: Siemens Motion M	L: CP910	L: ACE	L: 22	Enlarged vestibular aqueduct	Specialised
HL6	1	7.8/7.5	0.3	M	1.3	L: Moderately-severe R: Severe	Bimodal	L: Phonak BTE R: CI512 (CA)	R: CP910	R: ACE	R: 22	Unknown	Mainstream
HL8	1	8.5/7.7	0.8	F	4.2	L: Moderately-severe R: Moderately-severe	HA	L: Siemens Motion P R: Siemens Motion P				Usher syndrome	Mainstream
HL11	2	6.7/6.2	0.5	F	0	L: Moderately-severe R: Profound	Bimodal	L: Siemens BTE R: CI24RE (CA)	R: CP910	R: ACE	R: 22	Hypoplasia of the auditory nerve	Mainstream
HL12	2	7.8/5.8	2.0	M	4.3	L: Profound R: Profound	CI	L: CI24RE (CA) R: CI24RE (CA)	L: CP920 R: CP920	L: ACE R: ACE	L: 22 R: 22	Unknown	Mainstream
HL14	2	6.7/4.9	1.8	F	0.2	L: Profound R: Profound	CI	L: CI24RE (CA) R: CI24RE (CA)	L: CP920 R: CP920	L: ACE R: ACE	L: 22 R: 21	Waardenburg syndrome type 2	Specialised
HL15	2	6.3/6.0	0.3	M	1.3	L: Profound R: Profound	CI	L: CI512 (unknown) R: CI422 (unknown)	L: CP920 R: CP920	L: ACE R: ACE	L: 22 R: 22	Unknown	Mainstream
HL16	2	8.6/8.5	0.1	M	4.8	L: Moderately-severe R: Moderately-severe	HA	L: Phonak BTE R: Phonak BTE				Genetic	Mainstream
HL17	2	6.8/6.7	0.1	F	4.5	L: Profound R: Severe	Bimodal	L: Concerto FLEX28 R: Siemens BTE	L: Sonnet	L: FS4	L: 12	Connexin 26	Mainstream
HL18	2	9.2/7.2	2.0	F	0	L: Profound R: Profound	CI	L: CI24RE (ST) R: CI24RE (ST)	L: CP910 R: CP910	L: ACE R: ACE	L: 22 R: 19	Unknown	Specialised
HL19	2	6.8/6.3	0.5	M	0	L: Profound R: Profound	CI	L: CI24RE (CA) R: CI24RE (CA)	L: CP910 R: CP910	L: ACE R: ACE	L: 22 R: 22	Genetic	Specialised
HL20	2	8.8/8.4	0.4	M	3.6	L: Profound R: Profound	CI	L: CI512 (CA) R: CI512 (CA)	L: CP910 R: CP910	L: ACE R: ACE	L: 22 R: 20	Connexin 26	Mainstream

*Formal music experience was calculated as the duration (in years) of the musical activity, multiplied by its frequency, divided by the number of categories (n = 6). The musical activity categories were: music lessons, singing groups, instrumental groups, dance classes, and group-based classes. As an example, 1-year of weekly piano lessons = 0.7.

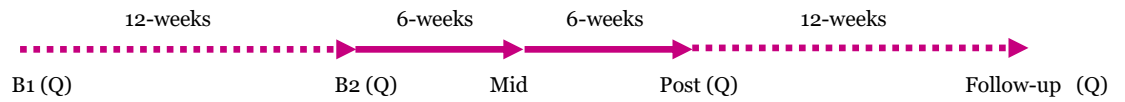
Table 3. Demographic information for TH children.

ID	Age	Sex	Formal music experience
TH1	8.0	F	2.7
TH2	6.3	M	0.7
TH3	7.8	F	10.8
TH4	6.3	F	4.0
TH5	8.2	M	3.3
TH6	8.3	M	2.7
TH7	6.6	F	5.3
TH8	8.6	F	2.5
TH9	6.3	M	0.0
TH10	7.5	M	2.0
TH11	7.2	F	0.3
TH12	8.7	M	1.0
TH13	7.6	M	1.3
TH14	7.3	M	0.0
TH15	8.4	F	7.7
TH16	7.7	M	1.5

Group 1



Group 2 (waitlisted)



Typical-hearing comparison



Figure 5. Overview of study design. The two groups were essentially the same, apart from a difference in retest duration between the baselines (B1 and B2). Questionnaire testing is denoted with “Q”.

Stimuli and tests

Australian Sentences Test in Noise (AuSTIN)

SIN was measured with the AuSTIN, an adaptive SIN test that has the unique advantage of being specifically designed for AusE CI recipients (Dawson, Hersbach, & Swanson, 2013). The complexity of the speech materials was suitable for children, as the sentences were developed with audiologists and speech-pathologists familiar with the linguistic capabilities of Australian children with hearing loss (Dawson et al., 2013). The AuSTIN features an adult female as the target speaker in the presence of 4TB featuring two adult female and male speakers. Twenty sentences (each comprising between four and six words, or six to eight syllables) were randomly selected without replacement and presented. Participants were asked to repeat the sentence as best they heard; and were morphemically scored (e.g. singing consists of two morphemes: sing and -ing). If the participant scored $\geq 50\%$ morphemes correct the competing noise level was increased, and if the participant scored $< 50\%$ morphemes correct the competing noise level was decreased. The AuSTIN adaptive rules and speech reception threshold (SRT—defined as the signal-to-noise ratio at which 50% of words were correctly perceived) calculation rules were selected. The initial signal-to-noise (SNR) ratio was 12 dB, with 4 dB step sizes for the first four sentences, followed by 2 dB step sizes for the remaining sentences. SRTs were calculated as the average of the SNRs for sentences 5 to 20, and the SNR of sentence 21 (which was not presented), based on the participant's response to sentence 20. AuSTIN has been well validated, and using these parameters, has a test-retest reliability of 0.99 dB. Thus, it is a suitable and appropriate SIN test for a longitudinal study with AusE children with SNHL.

Spectral-temporally Modulated Ripple Test (SMRT)

Spectro-temporal modulation detection performance was measured with SMRT version 1.1 (Aronoff & Landsberger, 2013). The SMRT has been used effectively in child studies (Kirby, Browning, Brennan, Spratford, & McCreery, 2015; Landsberger, Padilla, Martinez, & Eisenberg, 2017). Stimuli were non-harmonic tone-complexes with 202 equal amplitude pure-tone frequency components spaced every $1/33.33$ of an octave from 100 to 6400 Hz. Stimuli were 500 ms in duration, with 100 ms onset/offset linear ramps generated with a 44.1 kHz sampling rate. Participants were presented with a three-alternative forced choice task (3-AFC) in which two choices were reference stimuli at 20 rpo. The third choice was the target stimulus at an initial 0.5 rpo, modulated with a 1-up, 1-down adaptive procedure with a step size of 0.2 rpo. After ten reversals, a threshold was calculated based on the last six reversals.

Macquarie Battery of Emotional Prosody (MBEP)

Emotional prosody was measured with the MBEP (Thompson, Marin, & Stewart, 2012) that consisted of sentences that varied in emotional prosody. The sentences were semantically neutral such as “the girl and boy went to the fridge, to get some milk for lunch”, and were recorded by 4 female and 4 male speakers. Each sentence was 14 syllables in length and spoken with the emotional state of: happy, sad, angry, and scared. For this study, the MBEP was configured as a 2AFC task with two conditions: happy/sad, and angry/scared. The happy/sad sentences were representative of an easier task as their acoustic features were more perceptually distinct than angry/scared. Scores were averaged between the two conditions and calculated as percentage correct. While this specific test has not been previously used with children, the sentences and paradigms are not dissimilar to comparable test materials that have been used effectively for children with hearing loss (e.g. Chatterjee et al., 2015).

Question/Statement Prosody Test (QSPT)

The QSPT was developed for the present study to measure performance in differentiating questions from statements through a rising or falling terminal pitch. Two native adult speakers of Australian English recorded eight simple bi-syllabic words (e.g. carrot, garlic, orange; typical fruit and vegetable items) uttered naturally in question form with a rising pitch, and in statement form with a flat or falling pitch. Speakers maintained a consistent vocal effort, tempo, and level of intonation. The tokens were recorded in a sound-proof room with an AKG (Vienna, Austria) C535 EB microphone connected to a PreSonus StudioLive 16.4.2 mixing console with ProTools 11. High pass filtering was set on the mixing console at 75 Hz. Each token was saved as an individual .wav file, and the RMS level was adjusted to -25 dBFS. Participants were presented with 32 words in random order (2 speakers x 2 intonations x 8 words) and results were scored as percentage correct. Participants were instructed they would hear one word and had to decide if it sounded like the person speaking was asking the participant if they wanted the item (question utterance); or if it sounded like the person speaking was telling the participant they were simply pointing out an item (statement utterance). Pitch intonation curves were extracted using the Praat Vocal Toolkit (Corrette, 2018) and are presented in Figure 42 in Appendix C. On average, the pitch extraction for both male and female speakers for the question utterance was approximately one octave (or 12 semitones) when measured from lowest to highest frequency. The tokens developed for this test were similar to those in the turn-end receptive subtest of the Profiling Elements of Prosodic Systems—Child version (PEPS-C), which is appropriate for both adults and children (Peppé & McCann, 2003).

Clinical Assessment of Music Perception (CAMP)

The CAMP test was developed as a measure of music perception for adult CI recipients (Kang et al., 2009), but has been successfully administered for child CI recipients (Jung et al., 2012). It consists of 3 subtests: pitch direction discrimination, melody recognition, and timbre recognition. In the present study, 2 subtests were used: pitch direction discrimination and timbre recognition. Prior to each subtest, participants were provided brief practice sessions.

The pitch direction discrimination task used a two-alternative forced choice (2-AFC), 1-up 1-down adaptive testing method. The stimuli consisted of digitally synthesized, complex piano tones at three base frequencies: 262 Hz (C4), 330 Hz (E4), and 392 Hz (G4). Two tones were presented consecutively, a base frequency, and an initial interval presented at 12 semitones (1 octave), in random order. Participants were instructed to select the tone that was higher in pitch (i.e. the first or second tone). A correct response would yield a smaller subsequent pitch interval, whereas an incorrect response would yield a larger subsequent pitch interval (at that base frequency). The largest interval size was 12 semitones, the lowest interval size was 1 semitone, and the step-size was 1 semitone. Participant's pitch discrimination thresholds were calculated using the last 6 of 8 reversals at each base frequency, and their final pitch discrimination threshold was calculated as an average of all three base frequencies.

The timbre recognition task was an eight-alternative forced choice (8-AFC) task. The stimuli comprised eight live-recorded musical instruments that spanned four major classes: strings (violin and cello), brass (saxophone and trumpet), woodwinds (flute and clarinet), and percussion (guitar and piano). All instruments played an identical five note melody (C4-A4-F4-G4-C5) at 82 bpm, which were level-matched, and played with the same articulation and phrasing. Each instrument was played 3 times in random order, and participants were tasked with selecting the instrument they heard. Scores were calculated as percent correct.

Formal music experience

The Role of Music in Families Questionnaire (RMFQ) was developed to evaluate the role of music in families of children with hearing loss, and their general attitudes and level of engagement with music (Tuckerman, 2017; Tuckerman, McMahon, Looi, Lo, & Prvan, 2018). The RMFQ consists of 7 broad sections: General Demographic Information, Childhood Music Participation and Experiences, Attitudes and Reactions to Music, Resources for Child Regarding Music, Overall Importance of Music in your Household and Family, Child's Music Listening Preferences, and Future Perspective. One section of the RMFQ (Childhood Music Participation and Experiences) was used in the present study to appraise the level of formal music participation and experience each participant had received prior to commencement of the present study. A score was calculated on the basis of duration (in terms of years), multiplied by its frequency (1 = less often than monthly, 2 = once a month, 3 = two to three times a month, 4 = once a week, 5 = four to six times a week, 6 = two to three times a week, and 7 = daily), divided by the total number of categories ($n = 6$) that assessed activities: music lessons, singing groups, instrumental groups, special children's programs, dance classes, and group-based music classes. As an example, 1-year of weekly piano lessons = 0.7.

Music appreciation

A music appreciation questionnaire developed by Looi, King, & Kelly-Campbell (2012) for adults with hearing loss was adapted for use in the present study. Changes in music appreciation were measured after music training was completed. Questionnaires were child and parent-reported, requiring a response (depending on context) of "much better/more", "a little better/more", "no change", "a little worse/more", or "much worse/more", and assigned a value of +2, +1, 0, -1, and -2 respectively. Scores were averaged across parent and child. The questions asked were: Has the music program... 1. changed your enjoyment of music? 2. made music sound more pleasant? 3. made music sound more natural? 4. changed your ability to identify instruments? 5. changed your ability to recognise melodies? 6. changed your ability to learn new songs? 7. changed how much music you listen to? 8. changed how much you want to continue learning/exploring music? 9. changed your overall interest in music? 10. changed how much you want to learn an instrument/continue learning an instrument?

Strengths and Difficulties Questionnaire (SDQ)

The SDQ was developed by Goodman (1997) as a brief behavioural screening questionnaire that provides an overview of children's behaviour, emotion, and relationships. It consists of 25-items equally subdivided into five hypothesised subscales: Hyperactivity, Emotional symptoms, Conduct problems, Peer problems, and Prosocial. Example items of each respective subscale include: "Easily distracted, concentration wanders", "Many worries or often seems worried", "Often fights with other children or bullies them", "Has at least one good friend", "Considerate of other people's feelings". These were scored as: "not true", "somewhat true", or "certainly true", and assigned a value of 0, 1, and 2 respectively. Values were converted into subscales using SPSS syntax ("Scoring the SDQ," 2016). Based on the age of present study's cohort, parent-reported versions (recommended for children up to 10 years) were used. Due to the small sample size, the SDQ results were examined on the broader Internalising (Emotional + Peer), Externalising (Conduct + Hyperactivity), Prosocial, and Total (Emotional + Peer + Conduct + Hyperactivity) subscales as recommended by Goodman, Lamping, & Ploubidis (2010). The additional advantage of this technique is a reduction of measurement error (A. Goodman et al., 2010) .

Paediatric Quality of Life (PedsQL) Inventory

The PedsQL Inventory was developed by Varni, Seid, and Rode (1999) as a generic measure of health-related quality of life (HRQoL) that consists of a Generic Core Scale (GCS) and various condition-specific modules. The 23-item PedsQL GCS consists of four domains: Physical functioning (8 items), Emotional functioning (5 items), Social functioning (5 items), and School functioning (5 items). The following subscales were used for analyses: Physical Health Summary score consisting of the Physical functioning scale; a Psychosocial Health Summary score consisting of the Emotional, Social, and School functioning subscales; and a Total summary score. Both a parent-reports and child-reports were used. The self-reports for children aged 8–12, and parent-reported items were scored on a 5-point scale: "never", "almost never", "sometimes", "often", and "almost always", and assigned a value of 100, 75, 50, 25, and 0 respectively. The self-report for children aged 5–7 years were simplified pictorially with happy/sad faces and used a 3-point scale: "never", "sometimes", and "always", corresponding to 100, 50, and 0. Thus, for all PedsQL scales, a higher score is indicative of a better HRQoL.

Hearing Environments and Reflection on Quality of Life (HEAR-QL-26)

The HEAR-QL is a quality of life (QoL) assessment tool designed specifically for children with hearing loss (Umansky, Jeffe, & Lieu, 2011). The 26-item HEAR-QL-26 is designed for self-report in children aged between 7–12 years and comprises 3 domains: Environments (13-items), Activities (6-items), and Feelings (7-items). Items were scored on a 5-point scale: “never”, “almost never”, “sometimes”, “often”, and “almost always”, and assigned a value of 100, 75, 50, 25, and 0 respectively. Thus, a higher score on a HEAR-QL-26 subscale indicates a better HRQoL.

The Glasgow Children's Benefit Inventory (GCBI)

Unlike the majority of questionnaires that make an assessment at a single point of time, the GCBI was designed as a post-intervention health-related benefit measure (Kubba, Swan, & Gatehouse, 2004). As such, the GCBI is potentially a more sensitive measure of change resulting from an intervention than the SDQ, PedsQL GCS or HEAR-QL-26. The 24-item GCBI is a flexible, parent-reported questionnaire, that broadly considers factors of emotion, physical health, learning, and vitality. Although the GCBI was designed primarily for surgical/medical intervention; it is designed to be modified such that any intervention can be reworded into the items. For example, “Has your child’s (participation in the music program) affected their learning?”. Items were scored as “much better”, “a little better”, “no change”, “a little worse”, and “much worse”, and assigned a value of +2, +1, 0, -1, and -2 respectively. A total score was calculated by adding all numerical scores, dividing by 24 (number of questions) and multiplying by 50 to produce a total score on a scale of -100 (maximum harm) to +100 (maximum benefit).

Procedures

Testing

All testing occurred in an acoustically treated sound booth. The test battery was administered using a laptop computer with the following peripheral connections: audio output was through a loudspeaker (Genelec 8020C; Iisalmi, Finland) connected to an external soundcard (Yamaha AUDIOGRAM 3; Hamamatsu, Japan). Test battery responses were displayed and inputted by the child on a touchscreen monitor. Presentation level of test materials was calibrated to 65 dBA with a Digitech QM1592 sound level meter measured at the participants position, located 1 metre directly in front of the loudspeaker. The exception was the MBEP, as each emotion varied with intensity; as such, the happy sentences were used for level calibration.

The test battery took approximately one hour to complete. All perceptual test materials were presented in randomised order, followed by questionnaires in fixed order (parents: SDQ, PedsQL GCS, GCBI; children: PedsQL GCS, HEAR-QL-26). Questionnaires were paper-based, and the experimenter read aloud each questionnaire item to the child who could ask for clarification at any time. Children responded either verbally, or by pointing to their selection. Honest responses were emphasised at each session, and children were not allowed to consult or discuss their responses with their parents, who completed the questionnaires independently. All questionnaire reports were in relation to a 1-month recall period. All testing was shared between three experimenters (the first author and two research assistants); as such, approximately half of all test sessions were blinded. Participants could have a break at any time and were prompted by the experimenter if they would like a break half-way through the test session. Feedback and encouragement was provided for the first three tokens of each perceptual test, or for the duration of the practice trials. A token gift such as a sticker was provided half-way through the testing to maintain motivation, and at the end of the test session.

Music training

Music training was provided over 12-weeks, with a focus on maximising access to a broad range of musical skills and activities. The curriculum consisted of weekly, 40-minute, face-to-face group-based (4 to 5 children per class) music therapy sessions facilitated by a registered music therapist in the Speech and Hearing Clinic at Macquarie University on a Saturday morning. The activities were based on the Nordoff-Robbins approach (Nordoff, Robbins, & Marcus, 2007) with input to cater for a hearing impaired population. Participants were also expected to complete a series of activities 3 times a week (approximately 15 to 30 minutes depending on ability) with MusicFirst Junior (Music Sales Group, 2018)—an online-based suite of music apps designed for children aged between 6 and 12 years that is compatible for PC/Mac/smart devices that included: Morton Subotnick's Music Academy and Groovy Music. The app curriculum was developed by the first author, with input from the music therapist to match the goals at each week. Parents were encouraged to set aside a regular time for app use, which was regarded as homework. MusicFirst Junior allows for a rudimentary logging of activity (not completed, partially completed, or completed activity), and app-use and compliance was discussed at each Saturday morning session with the parents. Examples of music therapy activities include drumming, singing, dancing, and improvisation. Examples of the music apps include “drawing” and creating compositions, and identification of high, low, fast, or slow sounds. The use of group-based face-to-face music therapy sessions is in alignment with what is offered at many early intervention centres

within the greater Sydney region, while the use of computer-based programs is a common approach for research purposes. While group-based activities are ecologically valid and have the advantage of social engagement, they lack the level of control that computer-based approaches allow for, which also have the additional benefit of data-logging the activities. Thus, this hybrid approach of face-to-face group-based activities, supplemented by online-based apps, bridges the gap between research and practice, and encouraged maximum participation in a broad range of musical activities during a limited timeframe. The full curriculum can be seen in Appendix B.

Attendance at the music therapy sessions was generally high, ranging from 67% to 100% attendance rate ($M = 83\%$, $SD = 10\%$) with most absences due to illness or family obligations. Use of apps was more variable, with one participant not using the app at all (the parent reported time constraints). With the removal of this outlier, music-app compliance ranged from 39% to 83% ($M = 64\%$, $SD = 13\%$). Additionally, one participant also left the study in week 8 due to a surgical procedure.

“The beautiful thing about learning is nobody can take it from you.” — B. B. King

Chapter 4 — Music training for children with hearing loss improves music and speech outcomes

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Keywords: Paediatrics, Hearing loss, Music, Speech perception, Training

Abstract

A growing body of evidence suggests that long-term music training provides benefits to auditory abilities for typical-hearing adults and children. Of particular interest are speech perception enhancements such as speech-in-noise (SIN) and prosody. In this study, children aged 6–9 years with prelingual sensorineural hearing loss participated in a 12-week music training program. Activities included weekly group-based music therapy and take-home music apps 3-times a week. The design was a pseudo-randomised, longitudinal study (half the cohort was waitlisted, initially serving as a passive control group). There were no changes for any outcomes for the passive control group. After music training, perception of SIN, question/statement prosody, musical-timbre, and spectral resolution improved significantly, as did measures of music appreciation. There were no benefits for emotional prosody or pitch perception. The findings suggest even a modest amount of music training has benefits for music and speech outcomes. The results provide further evidence that music training is an excellent complementary means of habilitation to improve the outcomes for children with hearing loss.

Introduction

The continual advancement and confluence of effective early intervention, hearing technologies, clinical practice, and community engagement have resulted in better outcomes for children with hearing loss, and the majority achieve suitable proficiency when perceiving speech in quiet environments (Blamey et al., 2001). Poorer and more variable outcomes are observed in challenging listening situations such as SIN (Davies, Yellon, & Purdy, 2001; Schafer & Thibodeau, 2006), spectral resolution (Landsberger et al., 2017), and prosodic tasks (Chin, Bergeson, & Phan, 2012; Volkova, Trehub, Schellenberg, Papsin, & Gordon, 2013). The perception and appreciation of music and musical features such as pitch and timbre may also present perceptual challenges for many individuals (Gfeller et al., 2011; Jung et al., 2012; Petersen et al., 2015; Trehub, Vongpaisal, & Nakata, 2009). Modern industrial society is inherently noisy, and the primary concern for children with hearing loss is that they have access to adequate audibility and intelligibility in the context of their learning, education, and social communication.

Studies have investigated the use of music training as a means of improving auditory skills in a wide range of adult and paediatric populations. Music training may be especially effective at refining auditory skills because it requires sensitivity to rapidly changing, fine-grained spectral and temporal cues (Kraus & Chandrasekaran, 2010). Such benefits may be especially useful for hearing impaired populations, as supported by research on TH professional musicians. A recent review by Coffey, Mogilever, and Zatorre (2017) found that 18 of the 20 reviewed studies found support for a “musician advantage”—an enhancement of SIN perception. However, such benefits are difficult to interpret, because musical skills and activities are highly variable among musicians, and SIN can be measured with varying types of noise in a variety of speaker configurations. As such, the mechanisms by which musical skills lead to SIN enhancement have yet to be fully understood. Additionally, these 20 reviewed studies were cross-sectional in design, and as such, it is plausible that individuals with better-than-average auditory skills may be predisposed into pursuing musicianship. On the other hand, a paediatric study by Slater et al. (2015) provided the first longitudinal evidence for a causal SIN benefit from music training. In this study, 38 TH children were equally distributed between Harmony Project’s standard curriculum (introductory musicianship followed by group instrumental training) or to a waitlisted (control) group. Randomisation removed the risk of sampling bias and pre-existing differences, and there were no significant differences on age, sex, gender, IQ, maternal education, SIN, or age of English acquisition. After 2 years of music training, 19 TH children showed a mean improvement of -2.1 dB SNR on the Hearing in Noise Test (HINT), demonstrating the efficacy of a community-based music program for improving speech perception outcomes in children.

Music and speech share many acoustic similarities, and the broad principle underlying the mechanism for the musician advantage is generally conceptualised as overlapping (or shared) perceptual or neural processes (Patel, 2014). However, evidence for cognitive transfer functions from music to speech perception is far from established, and there is also conflicting evidence for the functional specialisation of brain structures with preference for specific sound categories (Angulo-Perkins et al., 2014; Peretz et al., 2015). Irrespective of these discrepancies, a consolidating perspective is that musicality is a multimodal experience that activates a wide range of brain regions associated with arousal, emotion, cognition, memory, and motor coordination (Brown & Palmer, 2012; Rauscher & Hinton, 2011; Thompson, Schellenberg, & Husain, 2001; Wan & Schlaug, 2010)—all of which may contribute to more effective auditory learning (Herholz & Zatorre, 2012; Shams & Seitz, 2008).

Benefits of multimodal activities have also been reported in paediatric hearing loss studies; Vongpaisal, Caruso, and Yuan (2016) demonstrated that even in a short-term song learning task, training that combined auditory and motor components was more beneficial than in auditory training alone for 9 CI recipients aged between 4–12 years. Another study by Innes-Brown, Marozeau, Storey, and Blamey (2013) investigated the benefits of a year-long participation in “Music Club”—45 minute musical activities centred around play for 11 children with hearing loss aged between 9–12 years. While participation did not confer any perceptual advantages, the children and teachers reported a wide range of benefits such as increased engagement and interest in music, and increased levels of socialisation with peers. Taken together, these findings promote physical engagement with music as an effective means of habilitation that may provide benefits beyond the auditory domain. Additionally, while the enjoyment of music is highly variable among children with hearing loss (Gfeller et al., 2011), it is a generally engaging activity that may assist in maintaining motivation and compliance—critical for longitudinal training studies (Gfeller, 2016; Patel, 2011; Trehub et al., 2009).

The number of studies that have investigated the benefits of music training for children with hearing loss is modest, with a wide range of music training protocols, age ranges, and outcomes of interest; for a review of music training for children with CIs see Gfeller (2016). The majority have been concerned primarily with music outcomes; Chen et al. (2010) tested 27 CI recipients aged between 5–14 years on a same/different pitch task. Half the participants were provided Yamaha Music School classes that involved listening, singing, score reading and instrument playing over varying durations (2–36 months). A significant correlation was found between the duration of training and pitch perception, suggesting possible tonotopical reorganisation and finer frequency tuning. These findings were further supported by Fu, Galvin, Wang, and Wu (2015), with 14 CI recipients aged between 5–9 years improving

in melodic pitch perception after 10-weeks of computer-based training; and in a study by Torppa et al. (2014) that found 8 (from a total of 21) unilaterally implanted CI recipients aged between 4–13 years with music experience (primarily singing) performed significantly better than those without music experience in auditory perception and attention.

Other investigations have considered potential transfer effects to other domains with a focus on speech perception. Good et al. (2017) compared the effects of 6-months of music training to visual art training for CI recipients aged between 6–15 years, which led to an enhancement of musical skills and emotional prosody processing for the musically trained children, but not for the visual art trained children. In a melodic contour training study for native Mandarin-speaking CI recipients aged between 5–9 years, significant improvements were observed for melodic contour identification and lexical tone recognition after 8-weeks of training (Cheng et al., 2018). These studies suggest a transfer effect between music and prosodic/intonation tasks, which is well supported by findings in TH studies (Hausen et al., 2013; Thompson, Schellenberg, & Husain, 2004), as well as adult CI studies (Lo et al., 2015), all of which implicate the use of pitch and rhythm as primary cues for intonation perception.

Broadly, spectral resolution is the ability to perceive and resolve fluctuations in the spectral domain and plays a key role in speech and music perception, which rely on various spectral cues and contrasts. A common method of measuring spectral resolution is with spectral ripple tests that have the advantage of avoiding confounds of language due to its non-linguistic stimuli. Several studies have shown reduced spectral resolution for adults with SNHL (C. W. Turner et al., 1999) and children with CIs (Landsberger et al., 2017), when compared to their TH peers. Interestingly, in cross-sectional studies, spectral resolution has also been found to correlate with SIN and music performance in postlingually implanted adults (Won, Drennan, Rubinstein, & Surgery, 2007); and SIN in prelingually implanted children (Jung et al., 2012).

Separate to perceptual accuracy, music appreciation considers the role of enjoyment and qualitative appraisal as an important, yet often overlooked outcome measure; for a review see Looi, Gfeller, and Driscoll (2012). For example, listeners do not need to identify instruments or specific notes within a composition to derive enjoyment from a musical piece. Music training studies in adult CI populations have shown that music appreciation can be learned and improved (Looi, King, et al., 2012). Furthermore, the lack of correlation between perceptual outcomes and appreciation as noted by Gfeller et al. (2008) and Wright and Uchanski (2012), highlights the importance in evaluating appreciation separately. The enjoyment of music in paediatric populations with hearing loss shows individual

variability with a general trend towards engagement and enjoyment (Chen-Hafteck & Schraer-Joiner, 2011; Gfeller et al., 2011).

Multiple studies have recommended the use of music training as a complementary means of habilitation for children with hearing loss (Abdi, Khalessi, Khorsandi, & Gholami, 2001; Chen et al., 2010; Petersen et al., 2015). However, the current body of evidence that music training is effective, or more effective than a standard habilitation program are limited, although recent findings for speech transfer effects are promising (Cheng et al., 2018; Good et al., 2017). Finally, Fuller, Galvin, Maat, Başkent and Free (2018) suggested extensive and intensive programs that combine face-to-face lessons along with computer-based pitch training may yield the greatest benefit, while Chen-Hafteck and Schraer-Joiner (2011) suggest best-practice may be the utilisation of a wide-range of activities to encourage the development of diverse skills.

The purpose of the present study was to investigate the benefits of a 12-week music training program, consisting of group-based face-to-face music therapy, supplemented by online music-apps for children with prelingual SNHL. Outcome measures included: SIN, speech prosody (specifically emotional and question/statement prosody), spectral resolution, pitch and timbre perception, and music appreciation. Based on previous findings by Chen et al. (2010) and Good et al. (2017), we hypothesized music outcomes would improve, and pitch perception would likely transfer to speech prosody. Irrespective of any change in perceptual accuracy, it was also hypothesised that participants would report higher levels of music appreciation after training. Additionally, given TH studies indicate a SIN benefit for adults and children with music training (Coffey et al., 2017; Slater et al., 2015), a SIN enhancement may also be possible for children with hearing loss. A measure of spectral resolution was included as there is evidence that better spectral resolution is associated with better SIN performance in prelingual children with CIs (Jung et al., 2012), as well as music perception in adult CI recipients (Won, Drennan, Kang, & Rubinstein, 2010). Finally, compared to the children with TH, it was expected children with SNHL would have poorer outcomes on all perceptual measures.

Results

Statistical analyses

IBM SPSS Statistics (version 22) was used to perform main hypothesis testing using linear mixed models (LMM) with Restricted Maximum Likelihood (REML). A significant advantage to LLM models is that it

can take into account missing data, hence all data from participants can be used for analysis even for those that did not complete the entirety of the music training ($n = 2$). An independent samples t -test was used for comparisons between children with SNHL and TH; concordance between parent and child responses on music appreciation was examined using Cohen's kappa statistical test, and a non-parametric Wilcoxon signed-rank test for the appreciation questionnaire responses. Criterion for statistical significance was fixed at $p = .05$.

For the double baseline analyses of the children with SNHL ($n = 14$), the following fixed effects were entered: time (baseline 1 and baseline 2), group (1-week retest and 12-weeks retest/waitlisted cohort), time * group (interaction term), device (CI, Bimodal, and HA), formal music experience, and hearing age (chronological age – age at fitting/implantation). It should be noted that hearing age was used to simplify the model and avoid over-parametrisation (due to the small sample size) by accounting for both chronological age and age at fitting/implantation as one variable. Accounting for formal music experience and hearing age in analyses is recommended by Gfeller (2016) for music training studies. For the double baseline analyses of the TH children ($n = 16$), the following fixed effects were entered: time (baseline 1 and baseline 2), formal music experience, and chronological age. For the music training analyses of the children with SNHL ($n = 11$), the following fixed effects were entered: time (pre—their baseline 2 scores, mid, post, and follow-up); device (CI, Bimodal, HA); formal music experience; and hearing age.

For the training analyses of the children with SNHL, participants were entered as random effects with random intercepts (random slopes were of interest, but they failed to converge); however, due to a lack of variability primarily from ceiling effects, the TH children were entered as random effects without random intercepts. Visual inspection of Q-Q plots did not reveal any obvious deviations from expected normal distributions. These models were used to predict: SIN, spectral resolution, pitch, timbre, emotional prosody, and question/statement prosody performance over time; controlling for device, hearing age/chronological age (for TH children), and formal music experience.

Double baseline measures

For the children with SNHL, no statistically significant differences were found for the main effect of time, or the interaction between time and group (i.e. either 1-week or 12-weeks retest) for any measure, with the exception of emotional prosody that improved significantly by 6.7% from baseline 1 to baseline 2, $F(1,13) = -2.746$, $p = .017$; driven primarily by the waitlisted group. Hearing age was a statistically

significant factor for pitch, timbre, emotional prosody, and question/statement prosody [$F(1,8) = -4.75$, $p = .001$; $F(1,8) = 4.41$, $p = .002$; $F(1,8) = 4.83$, $p = .001$; $F(1,8) = 2.33$, $p = .048$, respectively]—underscoring the importance of hearing age as a parameter of interest. Device was only statistically significant for the spectral resolution task; HA users’ spectral resolution ($M = 5.0$ rpo) were significantly better than CI recipients, $M = 2.68$ rpo, $F(2,8) = -2.68$, $p = .029$, and bimodal users, $M = 2.5$ rpo, $F(2,7) = -2.69$, $p = .031$. Formal music experience was trending towards significance for pitch perception $F(1,8) = -2.22$, $p = .057$.

For the TH children, no statistically significant differences were found for the effect of time. Chronological age was a statistically significant factor for pitch, spectral resolution, emotional prosody, and trending towards significance for SIN, [$F(1,28) = -2.78$, $p = .010$; $F(1,26) = 2.93$, $p = .007$; $F(1,19) = 2.21$, $p = .040$; $F(1,27) = -2.04$, $p = .052$, respectively]. Formal music experience was a statistically significant factor for question/statement prosody $F(1,27) = 2.36$, $p = .026$. All double baseline tables and figures for children with SNHL and TH can be found in Appendix C.

Perceptual measures

A table summarising outcome measures across time points can be found in Table 4. Mean estimates of each outcome measure across time with a TH comparison can be observed in Figure 6. The following results are estimated marginal means relative to performance at the pre-training measurement; comparisons to TH children are made in respect to raw baseline 2 measures (as the models to calculate each group’s estimated marginal means are not equivalent).

Speech-in-noise

A statistically significant improvement was observed for SIN at the post-training point with a mean SRT decrease of 1.1 dB, $F(3,11) = -2.40$, $p = .036$, which was essentially retained at the follow-up point with a decrease of 1 dB $F(3,15) = -2.17$, $p = .046$. On average, TH children's SRTs were 3.8 dB lower, 95% CI [-5.6, -2.0] than children with SNHL, $t(12) = -4.55$, $p < .001$.

Spectral resolution

A statistically significant improvement was observed for spectral resolution at the post-training point with a mean increase of 2 rpo, $F(3,12) = 4.89$, $p \leq .001$, and this was retained at the follow-up point with an improvement of 1.7 rpo, $F(3,9) = 3.76$, $p = .005$. On average, TH children's spectral resolution were 4.5 rpo higher than children with SNHL, $t(25) = 6.66$, 95% CI [3.1, 5.8], $p < .001$.

Emotional prosody

No statistically significant improvement for emotional prosody was observed for any time point. However, performance was generally excellent at pre-training (82% correct) suggesting a task that was too easy, with 4 participants scoring above 95% at the pre-training time-point, indicating a ceiling effect. On average, TH children's perception of emotional prosody were 13 percentage points higher than children with SNHL, $t(13) = 2.95$, 95% CI [4, 23], $p = .012$.

Question/Statement prosody

A statistically significant improvement was observed for question/statement prosody at the post-training point with a mean increase of 14 percentage points, $F(3,12) = 3.61$, $p = .004$, although this benefit was not fully retained at the follow-up point with an improvement of 8 percentage points, $F(3,13) = 1.99$, $p = .069$. On average, TH children's perception of question/statement prosody were 10 percentage points higher than children with SNHL, however this was not statistically different, $t(25) = 1.32$, 95% CI [-6, 26], $p = .197$.

Pitch

No statistically significant improvement for pitch threshold was observed over any time point. Surprisingly, TH children's pitch thresholds were not significantly different to the children with SNHL. On average, mean thresholds were 2.1 semitones lower, $t(25) = 1.80$, 95% CI [-4.4, 0.3], $p = .083$.

Timbre

A statistically significant improvement was observed at the mid-training point with timbre perception increasing by 6 percentage points $F(3,12) = 2.46$, $p = .028$, and at the post-training point with an increase of 8 percentage points $F(3,12) = 2.44$, $p = .032$, although this was not retained at the follow-up point with an improvement of 5 percentage points $F(3,4) = 1.41$, $p = .227$. On average, TH children's timbre perception were 31 percentage points higher than children with SNHL, $t(23) = 5.56$, 95% CI [19, 42], $p < .001$.

Device, hearing age, and formal music experience

Generally, device, hearing age, and formal music experience were not significant factors for most outcomes measures in the statistical model. Considering that hearing age was a significant factor for pitch, timbre and prosodic tasks; and device was a significant factor for spectral resolution at baseline measures; it suggests that the effect of training was more significant than the effect of hearing age or device. Device and formal music experience were not significant factors in any of the measures with music training. However, hearing age was a significant factor for emotional prosody and pitch perception [$F(1,6) = 6.2$, $p < .001$; $F(1,5) = 3.2$, $p = .022$, respectively]—reiterating the importance of including hearing age as a parameter of interest, particularly for pitch-based tasks. A scatter plot of hearing age with pitch and emotional prosody (averaged across all time points) can be observed in Figure 7.

Mechanisms for SIN enhancement

Post-hoc analyses explored possible mechanisms for SIN enhancement. As both spectral resolution and timbre perception improved significantly, bivariate correlations between these and SIN were analysed (measures were averaged over all time points). As shown in Figure 8, a moderate correlation was found between timbre perception and SIN, Pearson's $r = 0.611$, $p = .046$, although correlation does not equate

to causation, this finding provides evidence to further explore this relationship as a potential mechanism. No correlation was found between spectral resolution and SIN, Pearson's $r = -0.149$, $p = .662$.

Table 4. Results from the LMM for perceptual measures across time points.

Parameter	Estimate (<i>M</i> , <i>SE</i>)	<i>t</i>	<i>p</i>	95% CI	
				Lower	Upper
Speech-in-noise (SRT, dB)					
Pre	3.4 (0.6)	.	.	2.1	4.8
Mid	2.9 (0.6)	-1.04	.314	1.5	4.2
Post	2.3 (0.6)	-2.40	.036*	1.0	3.6
Follow-up	2.4 (0.6)	-2.17	.046*	0.9	3.8
Spectral resolution (rpo)					
Pre	3.6 (0.5)	.	.	2.5	4.7
Mid	4.7 (0.6)	1.94	.076	3.3	6.1
Post	5.6 (0.5)	4.89	< .001*	4.5	6.8
Follow-up	5.3 (0.5)	3.76	.005*	4.1	6.6
Emotional prosody (%)					
Pre	82.2 (2.0)	.	.	77.7	86.6
Mid	85.2 (2.1)	1.23	.239	80.6	89.7
Post	85.3 (1.8)	1.40	.191	81.0	89.6
Follow-up	85.3 (1.6)	1.58	.138	81.1	89.5
Question/Statement prosody (%)					
Pre	70.8 (5.8)	.	.	57.9	83.7
Mid	77.8 (5.9)	1.40	.181	64.8	90.9
Post	84.4 (4.9)	3.61	.004*	72.1	96.8
Follow-up	79.1 (5.3)	1.99	.069	65.7	92.5
Pitch (threshold, semitones)					
Pre	4.3 (0.5)	.	.	3.1	5.4
Mid	3.6 (0.5)	-1.30	.216	2.3	5.0
Post	3.8 (0.9)	-0.59	.571	1.7	5.8
Follow-up	4.0 (0.6)	-0.61	.566	2.6	5.3
Timbre (%)					
Pre	24.3 (3.1)	.	.	16.2	32.4
Mid	30.6 (3.2)	2.46	.028*	23.1	38.1
Post	32.4 (3.8)	2.44	.032*	23.9	40.8
Follow-up	29.6 (4.3)	1.41	.227	19.2	40.0

* $p \leq .05$, relative to pre-training measurement.

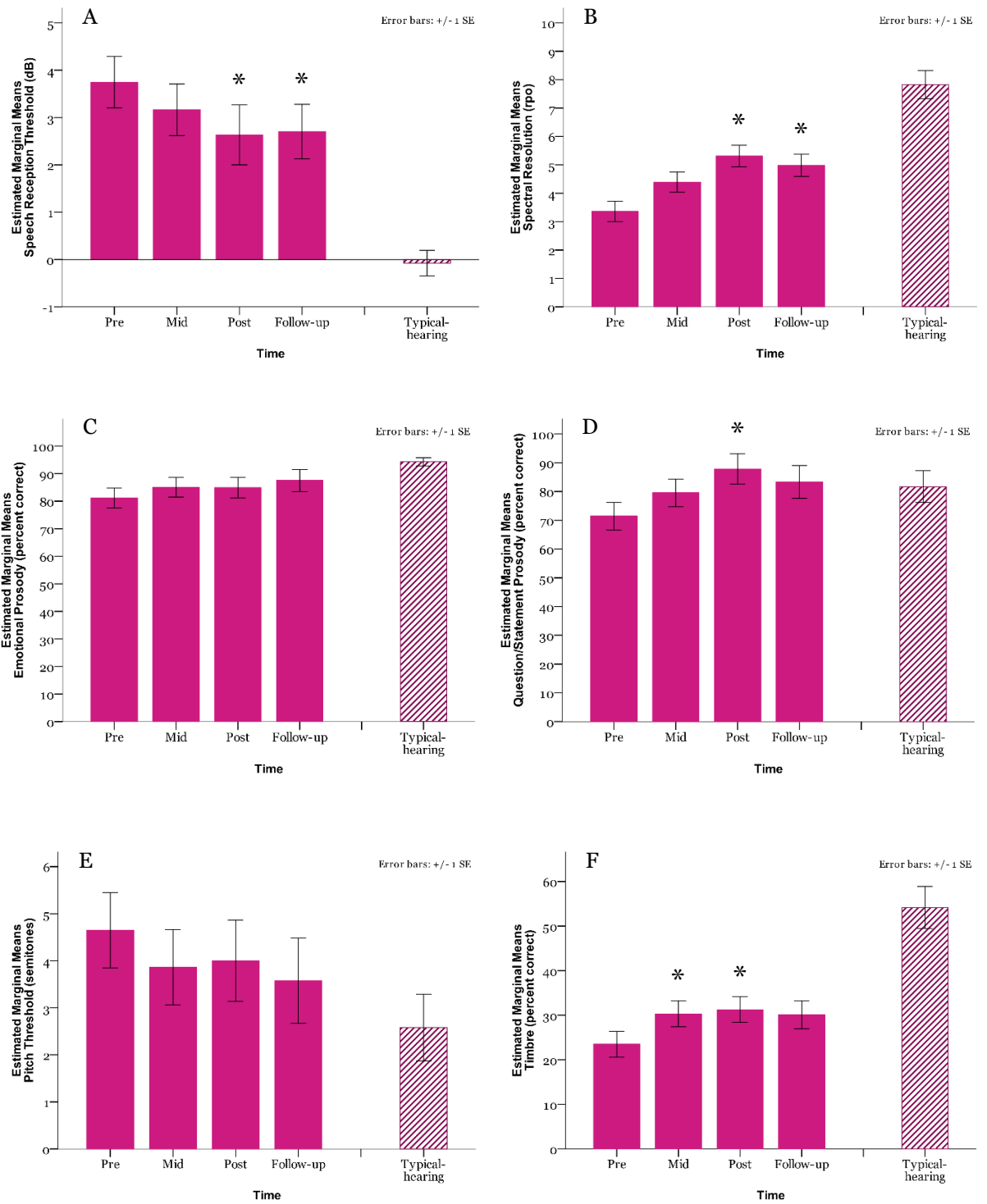


Figure 6. Bar graphs of estimated marginal means across time with a comparison of TH children's performance: (A) SIN, (B) spectral resolution, (C) emotional prosody, (D) question/statement prosody, (E) pitch, and (F) timbre. * $p \leq .05$ compared to pre time point.

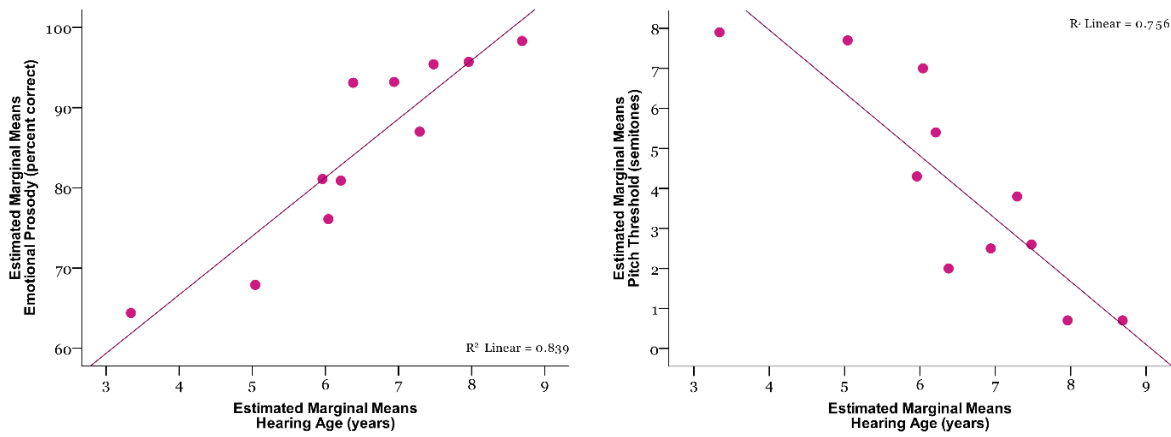


Figure 7. Scatter plot of estimated marginal means for hearing age with emotional prosody (L), and hearing age with pitch (R).

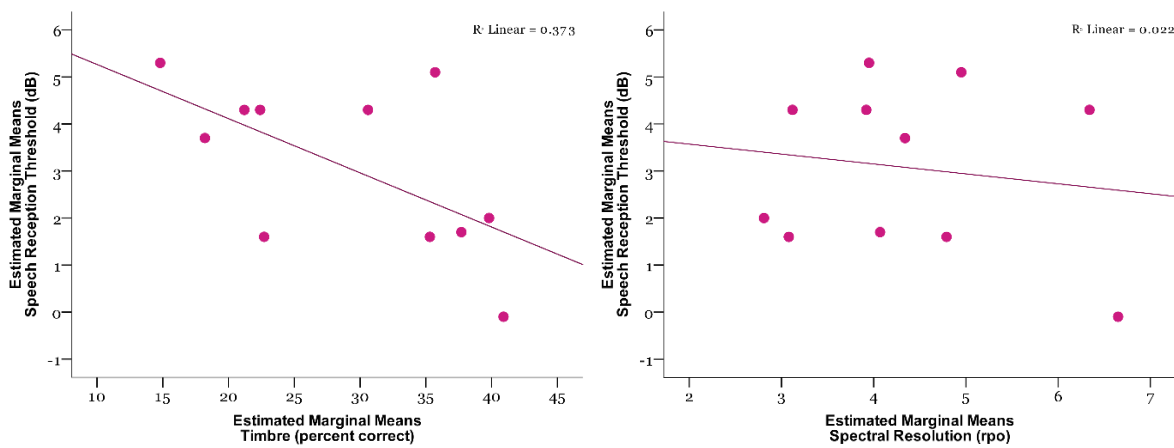


Figure 8. Scatter plot of estimated marginal means for timbre with SIN (L), and spectral resolution with SIN (R).

Music appreciation

Interrater reliability was examined between parent and child responses for music appreciation. For most measures, Cohen's kappa agreement was poor (-0.67 to 0.17), except questions asking if the music program had, 1. affected the child's ability to identify instruments, and 2. affected the child's motivation to learn or continue learning an instrument (0.44 = moderate agreement). Music appreciation was evaluated with a Wilcoxon signed-rank test, with a hypothesised median of interest set to 0 = no change. Table 5 indicates that after music training, a statistically significant improvement was observed for the vast majority of parent reported observations; while children reported music sounded more pleasant, that it improved their ability to identify instruments, and that they wanted to learn, or continue to learn an instrument.

Table 5. Wilcoxon signed-rank test of music appreciation and interrater reliability.

Has the music training program...	Parent report			Child report			kappa
	Z (SE)	Median	<i>p</i>	Z (SE)	Median	<i>p</i>	
1. changed your enjoyment of music?	45 (8.2)	1	.006*	6 (1.8)	0	.102	0.09
2. made music sound more pleasant?	10 (2.5)	0	.046*	10 (2.5)	1	.046*	0.09
3. made music sound more natural?	10 (2.6)	0	.059	7 (2.7)	0	.458	0.00
4. changed your ability to identify instruments?	28 (5.8)	1	.015*	15 (3.6)	1	.038*	0.44
5. changed your ability to recognise melodies?	45 (8.2)	2	.006*	10 (2.6)	1	.059	0.17
6. changed your ability to learn new songs?	28 (5.8)	1	.015*	6 (1.8)	0	.102	-0.67
7. changed how much music you listen to?	28 (5.8)	1	.015*	9 (2.6)	0	.131	0.15
8. changed how much you want to continue learning/exploring music?	28 (5.8)	1	.015*	6 (1.7)	0	.083	0.00
9. changed your overall interest in music?	28 (5.7)	2	.014*	13 (3.6)	1	.129	-0.04
10. changed how much you want to learn an instrument/continue learning an instrument?	28 (5.8)	1	.015*	15 (3.5)	2	.034*	0.44

**p* ≤ .05, relative to a hypothesised median = 0 (no change).

Discussion

After a 12-week music training program, outcomes for SIN, spectral resolution, timbre, and question/statement prosody were improved for children with prelingual, moderate-to-profound SNHL. While improvement to question/statement results broadly corroborate prosodic benefits (Good et al., 2017; Lo et al., 2015); the enhancement of SIN, spectral resolution, and timbre perception are novel, and to the authors' best knowledge, this is the first time such an effect has been observed after a music-based intervention for children with hearing loss. The trajectory of benefit was specific to each outcome variable, with question/statement prosody only improving at the post time point; timbre perception improving at mid and post time points; and SIN and spectral resolution improving at the post and follow-up points. It is difficult to ascertain whether these differences of trajectory are due to auditory development or the specificity of the curriculum provided. Additionally, it should be noted that by the follow-up time point, the number of participants was reduced by two, leading to a reduction of power for all measures at this point. Despite this, the observation that SIN and spectral resolution benefits were maintained at follow-up indicates a fairly robust effect. As expected, children with SNHL performed more poorly than their TH peers in the majority of measures, with the exception of pitch and emotional prosody perception, which will be discussed at a later stage.

Double baseline results for all groups (children with SNHL over 1-week or 12-week retests; and a TH comparison) were non-significant, except for emotional prosody for the waitlisted group which improved significantly. Collectively, the results indicate that all tests had suitable test-retest reliability, and that natural development and maturation over a 12-week period is insufficient to generate a significant change for the vast majority of outcomes measured. Additionally, the near-ceiling

performance on most tasks for the TH group indicated measures were developmentally appropriate for the age range of the children in the current study. While it was possible that some of the children with SNHL had a slight developmental delay—particularly in language, this likelihood was reduced by the widespread adoption of effective early intervention principles (Ching, 2015), and that the children in this study were prelingually implanted/aided. Nonetheless, any differences between the children with SNHL and TH were likely perceptual in nature. Hence, any change in outcome is likely attributable to the music intervention itself.

SIN enhancement is of considerable interest for habilitation, as children with hearing loss require a greater SNR than TH children, and SIN remains a commonly reported problem (Davies et al., 2001; Schafer & Thibodeau, 2006). It is also assumed that speech perception affects overall quality of life—although this is not well established empirically (Schorr, Roth, & Fox, 2009). SIN can be conceptualised as a higher-order auditory task, which is likely supported by top-down processes; which are in turn activated, developed, and organised through auditory input (Kral & Eggermont, 2007). The results of the present study are encouraging and suggest that SIN enhancement is potentially attainable for children with hearing loss, likely driven by perceptual fine-tuning—even with a short duration of music training. However, while the overall benefit for SIN was statistically significant, the effect size is relatively small with a mean improvement of 1.1 dB for SRTs. This value is close to the test-retest reliability of AuSTIN for adult CI recipients (Dawson et al., 2013); but the longitudinal nature of the study, the use of a double-baseline, in conjunction with the maintenance of SIN improvement at the follow-up point, supports the assertion that this is a reliable effect.

Timbre perception significantly improved at the mid- and post-training time points but was not retained when measured at the follow-up time point, due in part to a reduction of statistical power with 2 participants absent at this time point. This perceptual finding was supported by a music appreciation question that directly probed whether participants believed they were better able to identify instruments after training. Unlike previous adult hearing loss studies that have found qualitative/subjective appraisals do not necessarily correlate with perceptual outcomes (Gfeller et al., 2008; Looi, McDermott, McKay, & Hickson, 2007), the present study provides some evidence that both children and parents are fairly accurate self-reporters. Improved identification of instruments was also one of the few music appreciation measures that had a moderate level of agreement between parent and child-reports. Interestingly, post-hoc analyses showed a moderate correlation between timbre and SIN perception, and no correlation between spectral resolution and SIN. The relationship between timbre and SIN in CI adult studies is mixed, with Kang et al. (2009) finding a positive association, while Gfeller, Knutson,

Woodworth, Witt and DeBus (1998)—albeit from a much older CI study, did not. Furthermore, the relationship between speech and musical timbre in paediatric populations with hearing loss has not been previously explored. While our findings are correlational and unable to account for any causal effect, it suggests two interpretations for future consideration. Firstly, while SIN and timbre are often described in terms of discrete spectral or temporal cues, it is important to note that spectro-temporal modulations are more representative of natural speech (Santoro et al., 2014), and timbre dynamics (Patil, Pressnitzer, Shamma, & Elhilali, 2012). As such, the results suggest the possibility that an underlying shared process (such as enhancement of temporal cues, or spectro-temporal modulations) was improved, improving performance for both SIN and timbre perception. Secondly, the benefit may be conceptualised as a direct consequence of better timbre perception skills improving the perceptual organisation of auditory objects relevant for auditory scene analysis (Bregman, 1994; Ding & Simon, 2012; Kraus & Chandrasekaran, 2010). That is, the timbre task required the identification of instruments; improvement may have transferred specifically to the SIN task, in terms of better identification of the target (single female speaker) from masker signals (4TB) that differ in spectro-temporal modulations.

The large improvement in spectral resolution is noteworthy. Previous investigations have found that TH adults and children improve their spectral resolution as a function of age, whereas children with CIs mature at around 7 years old (Horn et al., 2017), and do not seem to improve as a function of age (Landsberger et al., 2017). Better spectral resolution is also associated with better SIN performance in postlingually implanted adults (Lawler, Yu, & Aronoff, 2017; Won et al., 2007) and prelingually implanted children (Jung et al., 2012). However, post-hoc analyses from the present study investigating correlations between spectral resolution and SIN find no evidence to support this relationship. This is in line with suggestions by Horn et al. (2017) and Landsberger et al. (2017) who argue that the auditory development of prelingually implanted children is fundamentally different to that of postlingual adults, with a greater weighting of temporal cues over spectral cues. The discrepancy between SIN and spectral resolution correlations in prelingually implanted children as reported by Jung et al. (2012) could be due to the small sample size, difference in age (8–16 years), and the difference in test material. Frequency discrimination assessed in spectral ripple tests is often confounded by factors such as loudness, spectral centroid, and changes to spectral edges—and these are exacerbated by CIs (Azadpour & McKay, 2012). The SMRT (which was used in the present study) was designed to avoid these confounding factors, and may be a more accurate measure of spectral resolution (Aronoff & Landsberger, 2013).

Additionally, a study by Nittrouer, Caldwell-Tarr, Moberly, and Lowenstein (2014) investigated perceptual weighting strategies in 8 year old children with and without CIs. Based on their findings, they

proposed that limited access to spectral cues diminished the development of language and perceptual weighting strategies. However, key to their argument was that this was independent to auditory sensitivity, and that enhancing sensitivity in and of itself was not optimal for phonemic (i.e. language-based) learning. As such, improvement to spectral resolution is likely to yield benefits, but in the longer-term context of language development. A final consideration is that learning effects have been noted in tests of spectral resolution. Tested at multiple time points, de Jong, Briare, and Frijns (2017) found the maximum mean improvement of 1.6 rpo was noted after 4-weeks. However, as suggested by de Jong et al. (2017), the use of a double-baseline, as well as test time points that are beyond a “carry-over” period—an effect or ability that carries over from one test to another, are recommended. As there was no significant difference for any baseline measures, our results are likely indicative of actual improvement resulting from the intervention that avoids a carry-over effect. Taken together, the results from the present study are both novel and encouraging; and open opportunities to the utility of music as a means of enhancing spectral resolution that otherwise does not appear to develop over time. However, benefits for music and speech outcomes as a result of improved spectral resolution for prelingual children with hearing loss would likely require a longer timeframe to develop (White-Schwoch, Carr, Anderson, Strait, & Kraus, 2013).

Contrary to findings by Chen et al. (2010), pitch perception did not improve in the present study. This was likely due to differences in training protocol, study design, and age of cohort; Chen et al. (2010) provided 13 CI recipients with Yamaha Music School classes that likely had a greater focus on traditional music pedagogy that involved score reading and instrument playing. Additionally, their findings were based on a study design that provided 2–36 ($M = 13.2$) months of music training to participants, as opposed to the present study in which all participants essentially received the same amount of training. As such, their results are based on a correlation between duration of training and pitch perception, as opposed to whether perceptual abilities were significantly different to baseline or control performance. Additionally, the curriculum of the present study had a broad range of musical activities that initially focussed on rhythm-, then timbre-, and then pitch-related tasks accordingly. As such, the amount of pitch-based training may not have been sufficient for changes to occur. Interestingly, in the present study, pitch perception performance was not significantly different between children with SNHL and those with TH. This is likely due to the age of the cohort, as the development of pitch has been estimated to not be fully matured until 11 years in TH children (Lamont, 1998), or early adolescence for more complex harmonic stimuli (Trainor & Unrau, 2012). This interpretation is also supported by the significant factor of hearing age for pitch perception and emotional prosody in statistical modelling,

which is shown in Figure 7. While there is a lack of data regarding the perceptual development of pitch perception as a function of age in children with SNHL, it is reasonable to expect that maturation would be delayed, given delayed access to auditory input, as well as early intervention programs focussing extensively on speech and language development.

Two prosodic tasks were used in the present study. While the sentence-based emotional prosody tasks did not significantly improve, the single-word question/statement task did. Unlike the study by Good et al. (2017) that used a 4AFC task and did find a significant benefit to emotional prosody after music training; the present study used a 2AFC task differentiated by difficulty, with happy/sad (easier condition) and angry/scared (harder condition). As such, the emotional prosody task was likely hampered by ceiling effects with 5 participants scoring above 95% ($M = 96\%$) at the pre-test session with both conditions averaged; and with 4 participants scoring above 90% ($M = 95\%$) at the pre-test even for the harder condition. On the other hand, question/statement prosody improved significantly. As pitch intonation is the primary cue for both prosodic tasks, upon initial inspection, this finding was not expected. However, the intonation curves were approximately 12 semitones (or an octave) in width, which is well within the participant's pitch thresholds; and these naturalistic utterances are also within the expected range for rising intonation utterances in studies with more controlled stimuli that extend as high as 15 semitones (Chatterjee & Peng, 2008; Holt & McDermott, 2013). Additionally, pitch was only tested using a discrete pitch direction task, and it is possible that broader measures of continuous pitch changes such as in the Montreal Battery for Evaluation of Musical Activities (MBEMA), or a melodic contour identification task as developed by Galvin, Fu, and Nogaki (2007) may be more suitable for measuring pitch-based improvements for children with hearing loss.

While the music appreciation results showed little concordance between parent and child responses; results indicated an overall positive change to music appreciation. The lack of concordance between the parents and children is not entirely surprising, given the vast difference in perceptual abilities and expectations from the study. Borrowing from quality of life literature that have long examined inter-rater reliability between child and parent-proxy reports—without consistent evidence as to which is more reliable, it is preferable to consider that each rater provides a contribution from a different perspective (Eiser & Morse, 2001; Jokovic et al., 2004). Overall, the parents reported widespread benefits across the vast majority of music appreciation measures, while responses from the children were more conservative. After training, children reported music as sounding more pleasant, and had an improved ability to identify instruments, which corresponds to the measured improvement in timbre perception. Interestingly, while there was no significant change in general interest towards

music, likely as the children had a high level of engagement and interest in music to begin with (Chen-Hafteck & Schraer-Joiner, 2011), they specifically wanted to learn, or continue learning an instrument. A study by MacKenzie (1991) investigated the motivations for wanting to learn an instrument in 48 TH children aged between 7–11 years. Their findings suggest that they are primarily self-motivated, followed by the influence of a teacher. As even the children who were learning an instrument wanted to continue, it is highly likely the music therapist was also influential in the present study. Anecdotally, many parents discussed instrumental training with the music therapist at the end of the 12-week music training session.

Compared to findings in postlingually implanted adults that found music more natural sounding after training (Looi, King, et al., 2012), this was not the case in the present study, which makes sense in the context of prelingually implanted/aided children who have no point of reference as to what “natural” should be, other than their own subjective experience. There was no change in how much music (more/less) participants wanted to listen to. While we did not explicitly ask how much music they were already listening to prior to training, it was likely sufficient, and not different to their TH peers. This is supported by findings that the hours spent listening to music for children between 2 and 5 years is similar, irrespective of hearing loss (Tuckerman, 2017; Tuckerman et al., 2018).

Compliance and general enjoyment of the group-based face-to-face music therapy sessions was high. A Wilcoxon signed-rank test $Z = 21, p = .023$ indicated high levels of enjoyment as reported by the children. However, the use of apps was, at times, hampered by technical issues. Difficulties arose primarily as the app required an online connection, and a few parents expressed frustration at the slow load times, and some compatibility issues on a range of devices. For the children that did not have technical problems, anecdotal evidence suggested overall enjoyment of the apps was high, but a Wilcoxon signed-rank test $Z = 19, p = .068$ was not significant, indicating a neutral appraisal of the apps in general. As stated by the parents:

“The app concept was great but let down by delivery over the internet. Too slow and lots of waiting. The loading time made it hard to engage with the activities and had problems with logging in on a few occasions.” [Parent of HL12]

As an off-the-shelf product with a purported wide range of hardware compatibility, one limitation is that participants’ engagement with the apps was flexible and not controlled. Parents reported greatest levels of success on tablet-like devices (compared to desktop computers), which was likely preferred as they are mobile and allow for tactile engagement. Overall, the hybrid approach of

face-to-face classes complimented by online apps was a fairly effective means of maximising the amount of music training provided in a limited period of time. Parents were also asked to provide feedback after the music training program:

“We would like to continue with the music program, as our son has made significant progress in the 12 weeks, and we would love for him to go further again! We have noticed that he has become quicker to identify songs on the radio, and even more astounding is that he has suddenly developed some intonation and tune to his singing along, which was previously non-existent. In addition, his music teacher at school has commented on his improvement, as have his clarinet teacher and band leader.” [Parent of HL3]

The present study was limited by a small sample size, lack of an active control group, and an unbalanced number of children using CI/bimodal/HA configurations. It should be noted that no analysis has been made to make any distinction between these configurations, instead treating the cohort as a broader group of children with moderate-to-profound SNHL. Furthermore, hearing device type was a factor in the statistical model, and the use of a repeated-measures design helps mitigate any potential differences this may entail. Nonetheless, the strengths of this study include the use of double baselines; a relatively well-constrained age range compared to most studies of this nature; additional controlling for age effects by including hearing age as a factor in statistical modelling—which was a significant factor for pitch and prosodic tasks; the use of a follow-up test point to measure retention. The use of a non-linguistic spectral resolution task was also novel, as was the measurement of both music appreciation and perceptual accuracy. Musical activities and benefits were maximised by using a multi-modal training protocol that combined group-based music therapy with the flexible use of apps. While this naturalistic pedagogical approach makes implementation of music therapy for children with hearing loss viable with minimum modification to a standard curriculum, it potentially makes generalisation and replication more variable and difficult than a highly structured computer-based approach (Gfeller, 2016). Replication of the present findings with larger sample sizes, across a range of ages will be required to reinforce the efficacy of music training for children with hearing loss. Furthermore, highly structured training protocols targeting specific areas of music perception will be required to understand various auditory processes and speech transfer mechanisms. Longer term training studies are also likely to generate greater outcomes.

Overall, the findings from the present study provide evidence that music training benefits tasks beyond music skills, such as SIN, timbre, spectral resolution, and question/statement prosody during

and after a 12-week music training program for children with SNHL. Much of the efficacy is likely derived from the multi-modal approach of the music training in conjunction with high levels of enjoyment that music provides. This study considered mechanisms and benefit primarily from a perceptual basis; nonetheless there are a great many possible mechanisms and areas of enquiry that are worthwhile considerations for future studies, including: statistical learning (Mandikal Vasuki, Sharma, Ibrahim, & Arciuli, 2017), cognitive factors such as working memory and attention (George & Coch, 2011; Torppa, Huotilainen, et al., 2014), language (Linnavalli, Putkinen, Lipsanen, Huotilainen, & Tervaniemi, 2018), and the development of musical production skills (Xu et al., 2009). In conclusion, the findings lend support to previous studies indicating transfer effects to speech perception; and adds to a growing body of evidence that supports the use of music as an effective and complementary means of habilitation.

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“For children, music is a natural inclination, and it often appears to be as essential to their well-being as it is for them to be warm, fed, and well-rested.”—Patricia Shehan Campbell

Chapter 5 — Music training for children with hearing loss: Quality of life and psychosocial outcomes

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Abstract

A small number of studies for typical-hearing children have suggested that group-based music activities are beneficial for pro-social outcomes and develop a sense of belonging. Children with hearing loss tend to have poorer psychosocial and quality of life (QoL) outcomes than their TH peers—particularly in areas with peers and school functioning. In this study, children aged 6–9 years with prelingual sensorineural hearing loss participated in a 12-week music training program. Activities included weekly group-based music therapy and take-home music apps 3-times a week. The design was a pseudo-randomised, longitudinal study (half the cohort was waitlisted, initially serving as a passive control group). There were no changes for any outcomes for the passive control group. Questionnaires utilised the Strengths and Difficulty Questionnaire (SDQ), the Peds QL, the HEAR-QL, and the Glasgow Children's Benefit Inventory (GCBI). After music training, SDQ internalising problems such as peer and emotional problems were significantly reduced. Additional benefits were noted for emotional and learning factors on the GCBI. However, there were no significant changes for the PedsQL or HEAR-QL instruments. This study provides some initial evidence that suggests music training has benefits for psychosocial and QoL outcomes for children with hearing loss.

Introduction

The main goal of early intervention for children with hearing loss is the provision of audibility for the primary purpose of maximising speech and language development (Joint Committee on Infant Hearing, 2007). Indeed, the majority of research investigating outcomes for children with hearing loss have been focussed towards improving and understanding language and speech perception (Blamey et al., 2001; Ching et al., 2017, 2018; Schorr, Roth, & Fox, 2008), with far fewer studies exploring psychosocial capabilities (Wong et al., 2017). The emphasis on language outcomes is warranted given the evidence that poorer language outcomes are correlated with a range of behavioural problems (Hoffman, Quittner, & Cejas, 2015; Stevenson, Mccann, Watkin, Worsfold, & Kennedy, 2010). However, it is important to consider that children's needs extend far beyond language. As Hargreaves, Marshall, and North (2003) assert, "most musical activity is carried out with and for other people—it is fundamentally social—and so can lay an important part in promoting interpersonal skills, teamwork, and cooperation". Considering the needs of children with hearing loss are complex, an examination of the relationship between music and psychosocial function is warranted, to provide a holistic perspective, as well as explore strategies that may be employed to improve habilitation not only for perceptual outcomes, but ultimately—quality of life.

The World Health Organisation (WHO) (1998, p.551) defines QoL as, "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns. It is a broad ranging concept affected in a complex way by the person's physical health, psychological state, personal beliefs, social relationships and their relationship to salient features of their environment. This definition reflects the view that quality of life refers to a subjective evaluation that is embedded in a cultural, social and environmental context." However, there are numerous definitions, often determined by the specific requirements and subjectivity of the enquiring field (Post, 2014; Wallander & Koot, 2016). Within paediatric audiology, QoL is often hypothesised as a resulting cascade of consequences that is directly influenced by the presence of prelingual deafness, subsequent intervention, and auditory and linguistic outcomes (Lin & Niparko, 2006; Stacey, Fortnum, Barton, & Summerfield, 2006; Summerfield & Marshall, 1999).

Health-related QoL (HRQoL) considers a subset of health-related terms that broadly encapsulate physical health, psychosocial health, and social interaction, and has become a dominant QoL measure (Wallander & Koot, 2016). HRQoL instruments are broadly stratified as either generic or specific. The advantage of generic measures are their wide applicability and use across many

populations, and they are typically well-validated with normative data, which allows for direct comparisons between groups of interest (Varni, Burwinkle, Seid, & Skarr, 2003). While less generalisable, specific instruments are designed for populations with a given disease or symptom and may be more sensitive for specific target outcomes (Umansky et al., 2011). A combination of both generic and specific measures are recommended to fully comprehend the broadness of HRQoL (Solans et al., 2008; Warner-Czyz, Loy, Tobey, Nakonezny, & Roland, 2011).

There is high variability in QoL outcomes for children with hearing loss, and a meta-analysis (albeit of only 4 studies using the PedsQL inventory for children aged between 6 to 18), by Roland et al. (Roland et al., 2016) found statistically and clinically poorer outcomes on the domains of school and social functioning in comparison to TH peers, whereas physical and emotional domains were not. It has also been reported in multiple studies that children with hearing loss are at greater risk for poorer psychosocial outcomes than their TH peers (Fellinger, Holzinger, Sattel, & Laucht, 2008; Kant & Adhyaru, 2009; Moeller, 2007). A range of psychosocial problems are associated with hearing loss; overt behaviours such as aggression and problems around conduct which are categorised as externalising behavioural problems, while behaviours such as depression and anxiety are categorised as internalising problems (American Psychiatric Association, 2013; Theunissen et al., 2014). A recent review by Stevenson et al. (Stevenson et al., 2015) examined emotional and behavioural difficulties for children and adolescents with hearing loss aged between 6 and 21 years of age and found the area at most risk and concern was in peer relationships. A study by Nunes, Pretzlik, and Olsson (2001) found that while children with hearing loss in a mainstream primary school setting were not disliked; TH peers often reported communication difficulties as a barrier to friendship—resulting in an isolating experience. However, a more recent study of social integration in an inclusive primary school setting that provided itinerant teacher support, speech and language therapy, sign language for all students, and use of FM systems found the children with hearing loss were not different to their TH peers on outcomes examining peer acceptance, social status, and number of mutual friendships (Wauters & Knoors, 2008).

Language and communication are well established factors associated with psychosocial development, as they are the primary means of establishing and maintaining social interactions (Barker et al., 2009; Stevenson et al., 2010). Better speech intelligibility scores are also associated with better adjustment and social competence (Hoffman et al., 2015; Polat, 2003). This is likely due to the general behaviour that from about 4 years of age, children tend to play in larger groups (Benenson, Apostoleris, & Parnass, 1997), increasing SIN, which leads to challenges in group-based interactions (Punch & Hyde, 2011). Surprisingly, when language abilities are controlled, degree of hearing loss does not appear to be

a significant factor (Stevenson et al., 2010; Theunissen et al., 2014). In effect, while hearing loss has a large psychosocial effect; there is a minimal difference due to *degree* of hearing loss, and this is likely attributable to effective early intervention and support (Fellinger et al., 2008).

According to a literature review investigating peer interactions for children with hearing loss in inclusive settings between the years 2000–2013, only one study has explored the efficacy of a social skills training program (Xie, Potměšil, & Peters, 2014). Suárez (2000) investigated an intervention for 18 children with hearing loss aged between 9 and 13 years that had the basic objective of improving interpersonal skills. The program consisted of 20 1-hour sessions twice a week that dealt with cognitive and interpersonal problem solving, followed by 6 1-hour social skills programs that was taught in conjunction with TH peers. However, the total duration of the study was not clearly reported. Overall, psychosocial factors such as emotional and social adjustment, as well as self-image were improved. More than half of the children showed improvements of assertive behaviour, inhibition, and thinking; these positive findings were supported by teachers of the deaf and self-reports.

More recently, Jeddi, Jafari, Motasaddi Zarandy, and Kassani (2014) investigated the efficacy of an 8-month longitudinal auditory-verbal rehabilitation program for 15 CI recipients with a mean age of 3.7 (± 1.2) years. Outcomes were measured every 2-months with the Newsha Development Scale—a scale evaluating the development of children with Persian language up to 6-years of age (Jafari & Asad-Malayeri, 2012). Social communication skills such as self-confidence, appropriate tone of voice, initiation of conversation, ability to follow topic changes within conversation, and expression of needs and feelings showed improvement at each time point. However, at the end of the 8-month intervention, despite the benefit, the children did not demonstrate an age-appropriate skill level, consistent with a generalised delay in language.

A small number of music training studies for TH children have also explored social skills and psychosocial wellbeing with mixed, but mostly positive findings. Kirschner and Tomasello (2010) found that joint music making tasks enhanced the prosocial behaviour of 4-year old children, which was not apparent in the control group that involved the same joint task and activities, but in a non-musical condition. Rickard et al. (2013) conducted a 2.5-year longitudinal study comparing the effects of participation in an embedded music curriculum compared to a standard curriculum across 9 schools. As such, the participants were not randomised, as their school enrolment determined their grouping, although baseline results indicated no significant differences. A total of 359 children from grades 1–3 participated, and while there was no significant benefit to social skills (potentially because they were above average at baseline), benefits were noted for self-esteem. Another longitudinal study by Williams,

Barrett, Welch, Abad, and Broughton (2015) explored the contributions of early book reading and music activities between parent and child in the home environment. Data was collected from 3031 children when they were 2–3 years, and again at 4–5 years. A large number of outcomes were investigated such as vocabulary, numeracy, school readiness, attentional and emotional regulation, and prosocial skills. Interestingly, shared music activities (and not shared book reading) were associated with better prosocial outcomes. Williams et al. (2015) suggested that activities such as dancing, singing, and instrument playing contribute to more face-to-face time between children and parents; and the additional benefit of music as non-linguistic activity may make it more accessible and interactive than shared reading.

The psychosocial benefits of musical activities are also apparent in older TH children. A study by Schellenberg, Corrigan, Dys, and Malti (2015) investigated a 10-month music program for 8–9 year old children ($n = 38$) in comparison to a control group that did not receive music training ($n = 46$). The music program focussed on the use of the ukulele and students were encouraged to “show your neighbour” and share their knowledge and skills, actively encouraging cooperative behaviour. There was some evidence of benefit, but prosocial skills only improved for children that were already poor performers from baseline. A large study by Welch, Himonides, Saunders, Papageorgi, and Sarazin (2014) evaluated the benefits of *Sing Up* (2007–2011), a national singing program in the United Kingdom. Paired data ($n = 6087$) between singing assessment scores and their mean questionnaire responses of social inclusion, showed that children with more developed singing ability had a more positive sense of self and were better socially integrated. For a review of music interventions and TH child development across a range of domains, see Dumont et al. (2017).

The psychosocial and QoL benefits from music-based interventions are mostly unexplored for children with hearing loss. A 2-year pilot study by Yucel et al. (2009) explored a music training program for 18 (9 active, 9 control) paediatric unilateral CI recipients and bimodal users that focussed on the use of a take-home electronic keyboard. Unfortunately, the ages were not clearly reported. Activities were centred on parents playing prescribed intervals and songs, and parents were encouraged to dance and play finger games. After training, benefits were shown for pitch and melodic perception, and parent-child relationships were noted as closer. Another study by Innes-Brown et al. (2013) investigated the benefits of a year-long participation in “Music Club”—45 minute musical activities centred around play for 11 children with hearing loss aged between 9–12 years. While participation did not confer any perceptual advantages, anecdotal reports from a debriefing session with the teachers suggested a wide

range of benefits such as increased engagement and interest in music, increased levels of socialisation with peers, and a sense of belonging.

The purpose of the current study was to explore the effect of a 12-week music training program on psychosocial and HRQoL outcomes for children with hearing loss. To the authors' best knowledge, no study has explored this with standardised generic or specific HRQoL questionnaires. It was hypothesized that music training would result in better outcomes for domains in which peer relationships and prosocial measures are central. Given all participants were physically healthy, we did not expect any benefit for physical domains. On the balance of previous findings, it was expected that children with hearing loss would have poorer outcomes than their TH peers on psychosocial and peer domains, and all measures of the HEAR-QL-26 that were directly related to hearing-specific problems.

Results

Statistical analyses

IBM SPSS Statistics (version 22) was used to perform main hypothesis testing using generalized linear mixed (GZLM) models. One advantage for using the GZLM model was to account for differences in the distribution of responses. For example, the majority of SDQ responses were not normally-distributed and were better suited to an analysis with a gamma-distribution. Asymmetric responses in the SDQ have also been noted in the longitudinal Millennium Cohort Study ($n = 11972$ observations) (Tzavidis, Salvati, Schmid, Flouri, & Midouhas, 2016). Another advantage to GZLM models is that it can take into account missing data, hence all data from participants can be used for analysis even for those that did not complete the entirety of the music training ($n=2$). Concordance between parent and child responses on the PedsQL was examined using intraclass correlation coefficients (ICC). ICC estimates and their 95% CIs were calculated on a mean-rating, absolute agreement, 2-way mixed effect model based on guidelines recommended by Koo & Li (2016). Criterion for statistical significance was fixed at $p = .05$.

For the training analyses of the children with SNHL ($n = 14$), the following fixed effects were entered: time (baseline 1, baseline 2, post, and follow-up), device (CI, Bimodal, and HA), and hearing age (chronological age – age at fitting/implantation). It should be noted that hearing age was used to simplify the model and avoid over-parametrisation (due to the small sample size) by accounting for both chronological age and age at fitting/implantation as one variable. For all analyses of the children with SNHL, participants were entered as random effects with random intercepts (random slopes were of

interest, but they failed to converge). Visual inspection of Q-Q plots indicated that SDQ measures for Internalising, Externalising, and Total scores were gamma-distributed (and thus analysed as such), while all other measures did not show any obvious deviations from expected normal (linear) distributions. All results are presented as estimated marginal means with respect to baseline 2 as a reference point, except for the GCBI which is only measured post-intervention. The use of baseline 2 as the reference time point allowed for comparisons with natural maturation and development (baseline 1), as well as any benefit from music training (post), and the retention of any benefit (post).

These models were used to predict measures over time for: SDQ (Internalising problems, Externalising problems, Prosocial, Total), PedsQL GCS (Physical Health, Psychosocial Health, Total), HEAR-QL-26 (Environment, Activities, Feelings), and GCBI over time; controlling for device, and hearing age. The following results are presented as estimated marginal means relative to performance at the pre-training measurement; comparisons to TH children are made in respect to raw baseline 2 measures (as the models to calculate each group's estimated marginal means were not equivalent).

SDQ

A summary of results for the SDQ scales can be found in Table 6 and Figure 9. A statistically significant improvement was observed at the post-training time point with SDQ Internalising problems decreasing by 3.5 points, $F(3,13) = 17.7$, $p = .001$, and this was retained at the follow-up time point with a decrease of 2.5 points, $F(3,16) = 5.4$, $p = .036$. A statistically significant improvement was observed at the post-training time point with a decrease of 4.8 points for SDQ Total difficulties, $F(3,13) = 8.2$, $p = .012$; however, this improvement was not maintained at follow-up, $F(3,12) = 2.4$, $p = .148$. There was no change across time for Externalising problems or the Prosocial scale. Device and hearing age were not significant factors in this model. On average, children with SNHL had SDQ Internalising problems that were 3.0 points higher, 95% CI [1.1, 4.9] than their TH peers, $t(26) = 3.21$, $p = .003$. Additionally, compared to TH peers, Externalising problems were 1.2 points higher, 95% CI [-2.3, 4.7], Prosocial scores were 0.6 points lower, 95% CI [-2.1, 0.9], and Total difficulties were 4.2 points higher, 95% CI [-0.6, 9.0], but these were not significantly different [$t(26) = 0.70$, $p = .488$; $t(26) = -0.85$, $p = .402$; $t(26) = 1.79$, $p = .085$, respectively].

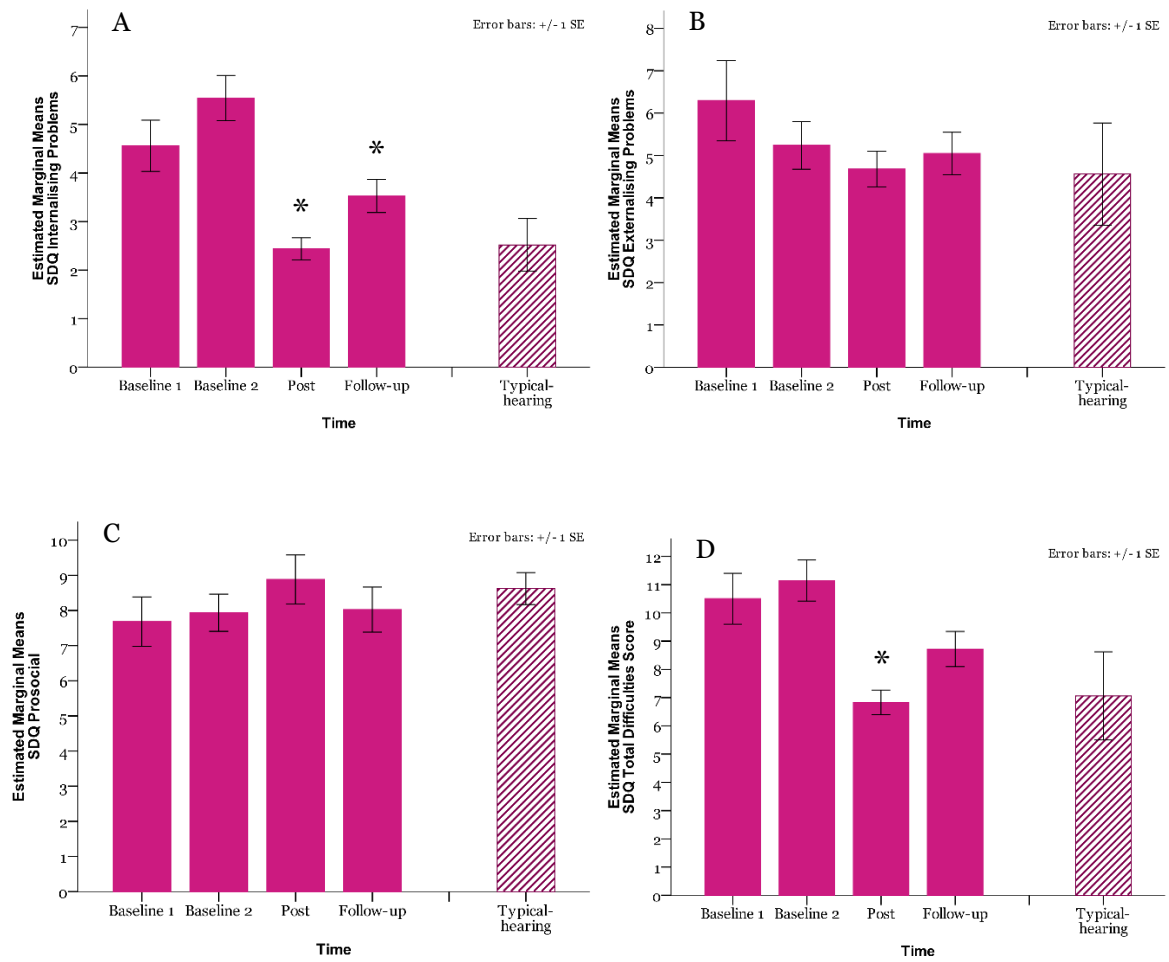


Figure 9. Bar graphs of estimated marginal means for SDQ subscales across time with a comparison of TH children's performance: (A) internalising problems, (B) externalising problems, (C) prosocial, (D) total difficulties.
 $*p \leq .05$ compared to baseline 2.

Table 6. Results from the GZLM for SDQ Scales across time points.

SDQ Scales	Parent-report (<i>M</i> , <i>SE</i>)	<i>t</i>	<i>p</i>	95% CI	
				Lower	Upper
Internalising Problems					
Baseline 1	5.1 (1.3)	-0.62	.559	2.7	9.5
Baseline 2	6.0 (1.0)	.	.	4.2	8.7
Post	2.5 (0.4)	-4.20	.001*	1.7	3.7
Follow-up	3.5 (0.7)	-2.32	.036*	2.2	5.5
Externalising Problems					
Baseline 1	6.5 (1.3)	1.07	.306	4.0	10.4
Baseline 2	5.2 (1.2)	.	.	3.1	8.6
Post	4.4 (0.9)	-0.73	.476	2.7	7.3
Follow-up	5.0 (0.9)	-0.24	.815	3.2	7.7
Prosocial					
Baseline 1	7.8 (0.9)	-0.71	.531	6.0	10.0
Baseline 2	8.1 (0.9)	.	.	6.3	10.4
Post	9.1 (1.1)	1.65	.126	6.9	12.2
Follow-up	8.0 (1.0)	-0.19	.853	5.9	10.8
Total Difficulties					
Baseline 1	11.3 (1.9)	-0.16	.874	7.7	16.6
Baseline 2	11.7 (2.1)	.	.	8.0	17.2
Post	6.9 (0.9)	-2.87	.012*	5.0	9.5
Follow-up	8.8 (1.1)	-1.54	.148	6.5	11.9

* $p \leq .05$, relative to measurement at baseline 2

PedsQL GCS

Interrater reliability was examined between parent and child responses across all time points. For the psychosocial health measure, ICC = 0.37, with a 95% CI [-0.27, 0.69]; for the physical health measure, ICC = 0.01, with a 95% CI [-0.82, 0.48]; and for the total score, ICC = 0.31, with a 95% CI [-0.34, 0.65]—all of which suggest poor reliability on average (Cicchetti, 1994; Koo & Li, 2016). A summary of results for the PedsQL GCS across time points for children with SNHL can be seen in Table 7. In terms of the effect of training, there was no change across time for any PedsQL measure whether parent, or child-reported. Device and hearing age were not significant factors in this model. On average, children with SNHL had slightly lower scores than their TH peers in general, though there was no statistically significant difference between them for any PedsQL measure which can be observed in Table 8.

Table 7. Results from the GZLM for PedsQL GCS across time points.

PedsQL Scale	Parent-report (<i>M</i> , <i>SE</i>)	<i>t</i>	<i>p</i>	95% CI		Child-report (<i>M</i> , <i>SE</i>)	<i>t</i>	<i>p</i>	95% CI		
				Lower	Upper				Lower	Upper	
Psychosocial Health											
Baseline 1	61.1 (4.1)	-1.26	.230	50.5	71.6	65.1 (5.9)	-0.27	.979	50.4	79.7	
Baseline 2	68.4 (4.6)	.	.	58.2	78.5	65.2 (4.5)	.	.	55.0	75.4	
Post	74.9 (2.5)	1.34	.203	69.0	80.7	60.3 (5.1)	-1.07	.333	49.1	71.6	
Follow-up	69.2 (3.2)	0.16	.872	62.0	76.5	63.1 (6.0)	-0.38	.714	47.8	78.5	
Physical Health											
Baseline 1	75.5 (5.3)	-0.66	.525	63.1	87.9	66.0 (6.0)	0.20	.853	52.1	79.9	
Baseline 2	79.2 (5.1)	.	.	68.2	90.2	65.3 (5.8)	.	.	51.8	78.8	
Post	79.5 (4.2)	0.05	.960	69.9	89.0	62.5 (7.7)	-0.47	.651	45.4	79.6	
Follow-up	74.0 (8.7)	-0.59	.569	54.4	93.6	66.4 (10.3)	0.12	.909	43.3	89.5	
Total Score											
Baseline 1	65.8 (4.0)	-1.14	.293	45.6	86.1	65.3 (5.9)	-0.74	.944	51.3	79.3	
Baseline 2	71.9 (4.4)	.	.	62.1	81.7	65.7 (4.9)	.	.	54.6	76.8	
Post	76.1 (2.6)	0.94	.372	70.3	81.9	61.3 (5.2)	-1.00	.345	49.7	72.9	
Follow-up	70.4 (3.8)	-0.30	.769	61.7	79.0	64.6 (6.1)	-0.21	.839	50.3	78.8	

Table 8. Independent *t*-tests of PedsQL between children with SNHL and TH.

SDQ Scales	M, SE Difference	<i>t</i>	<i>p</i>	95% CI	
				Lower	Upper
PedsQL Psychosocial Health (Parent reported)	-7.8 (5.3)	-1.46	.158	-18.8	3.2
PedsQL Psychosocial Health (Child reported)	-7.9 (5.3)	-1.49	.150	-18.9	3.1
PedsQL Physical Health (Parent reported)	-1.0 (8.2)	-0.12	.907	-17.7	15.8
PedsQL Physical Health (Child reported)	-5.6 (6.1)	-0.92	.365	-18.1	6.9
PedsQL Total (Parent reported)	-5.4 (5.9)	-0.92	.368	-17.5	6.7
PedsQL Total (Child reported)	-7.1 (5.0)	-1.43	.166	-17.4	3.2

HEAR-QL-26

A summary of results for all HEAR-QL-26 domains across all time points for children with SNHL can be seen in Table 9. There was no significant change in any domain as a function of music training. Device and hearing age were not significant factors in this model. Compared to TH peers, children with SNHL reported lower outcomes for all HEAR-QL-26 domains. The domains of Environments were 21.2 points lower, 95% CI [-33.1, -9.3], Activities were 14.5 points lower, 95% CI [-27.3, -1.8], Feelings were 25.7 points lower, 95% CI [-42.7, -8.7] and Totals were 20.4 points lower, 95% CI [-30.1, -10.8], all of which were significantly different [$t(24) = -3.68, p < .001$; $t(10) = -2.54, p = .030$; $t(12) = -3.29, p = .007$; and $t(24) = -4.36, p < .001$ respectively].

Table 9. Results from the GZLM for HEAR-QL-26 across time points.

HEAR-QL Domains	Parent-report (<i>M</i> , <i>SE</i>)	<i>t</i>	<i>p</i>	95% CI		
				Lower	Upper	
Environments						
Baseline 1	63.0 (7.7)	0.17	.871	44.3	81.7	
Baseline 2	61.6 (4.6)	.	.	50.8	72.4	
Post	57.0 (3.6)	-1.01	.332	48.1	65.8	
Follow-up	62.5 (6.5)	0.13	.900	47.6	77.4	
Activities						
Baseline 1	79.5 (7.7)	0.30	.770	61.6	97.5	
Baseline 2	77.3 (5.2)	.	.	65.9	88.8	
Post	74.4 (4.8)	-0.76	.473	63.1	85.7	
Follow-up	82.8 (7.1)	0.86	.414	66.4	99.2	
Feelings						
Baseline 1	67.3 (7.2)	0.27	.790	46.0	88.5	
Baseline 2	64.5 (7.8)	.	.	46.8	82.2	
Post	58.9 (8.5)	-0.50	.630	38.8	79.1	
Follow-up	46.7 (7.9)	-1.67	.124	26.3	67.0	
Total						
Baseline 1	70.6 (7.3)	0.32	.757	52.7	88.5	
Baseline 2	68.0 (5.0)	.	.	56.4	79.7	
Post	63.5 (3.5)	-0.95	.365	54.9	72.0	
Follow-up	65.7 (5.0)	-0.39	.706	54.6	76.9	

Glasgow Children's Benefit Inventory (GCBI)

The GCBI was evaluated with a Wilcoxon signed-rank test, with a hypothesised median of interest set to 0 = no change. Table 10 indicates that after music training, a statistically significant improvement was observed for overall life, $p = .015$; the capacity to do things, $p = .014$; better behaviour, $p = .020$; progress and development, $p = .009$; learning, $p = .005$; concentration, $p = .020$; happiness and contentment, $p = .046$; and confidence, $p = .025$, which indicated benefits primarily for emotion and learning factors, but not physical health and vitality. Total scores ranged from 0 to 48, $M = 20$, 95% CI [8, 31], in which -100 = maximum harm, and +100 = maximum benefit.

Table 10. GCBI results after music training.

Has your child's participation in the music program...	<i>p</i>	Observed median
1. made their overall life better or worse?	.015*	1 = a little better
2. affected the things they do?	.014*	1 = a little better
3. made their behaviour better or worse?	.020*	1 = a little better
4. affected their progress and development?	.009*	1 = a little better
5. affected how lively they are during the day?	.059	0 = no change
6. affected how well they sleep at night?	.317	0 = no change
7. affected their enjoyment of food?	1.000	0 = no change
8. affected how self-conscious they are with people?	.317	0 = no change
9. affected how well they get on with the rest of the family?	.157	0 = no change
10. affected their ability to spend time and have fun with friends?	.102	0 = no change
11. affected how embarrassed they are with other people?	.317	0 = no change
12. affected how easily distracted they have been?	.059	0 = no change
13. affected their learning?	.005*	1 = a little better
14. affected the amount of time they have had off school?	1.000	0 = no change
15. affected their ability to concentrate on a task?	.020*	1 = a little better
16. affected how irritable they are?	.180	0 = no change
17. affected how they feel about themselves?	.059	0 = no change
18. affected how happy and content they are?	.046*	0 = no change
19. affected their confidence?	.025*	1 = a little better
20. affected their ability to take for their self as well as you think they should, such as washing, dressing, and using the toilet?	.157	0 = no change
21. affected their ability to enjoy leisure activities such as swimming and sports, and general play?	.083	0 = no change
22. affected how prone they are to catch colds or infections?	1.000	0 = no change
23. affected how often they need to visit a doctor?	.317	0 = no change
24. affected how much medication they need to take?	1.000	0 = no change

**p* ≤ .05, relative to a hypothesised median = 0 (no change)

Discussion

The aim of the current study was to evaluate psychosocial and HRQoL outcomes for children with hearing loss after participation in a 12-week music training program. A combination of generic and specific, parent- and child-reported questionnaires were used to evaluate internalising and externalising problems, psychosocial and physical health, as well as hearing-specific questions targeting environments, activities and feelings. The primary finding was that internalising problems were significantly reduced at the post-training point which were also retained at follow-up. Somewhat surprisingly, there was no benefit for prosocial outcomes. Additionally, responses from the GCBI suggest a generally positive effect of training, with benefits primarily around emotional and learning factors. Compared to TH children, children with hearing loss had poorer outcomes for internalising problems, and all measures of the hearing-specific questionnaire; there were no differences for general psychosocial health, and as predicted—physical health. Finally, there were no differences between any

of the double baseline results, which suggests that any post-training benefit was likely due to the effect of the music intervention.

Parent-reports of children with hearing loss generally indicate greater internalising and peer problems than parent-reports of TH children (Barker et al., 2009; Hoffman et al., 2015; van Eldik, Treffers, Veerman, & Verhulst, 2004). The results from the present study are encouraging, as internalising problems (measured by the SDQ as the sum of peer and emotional problems) were improved after training and maintained at the follow-up time point. The prosocial scale showed no change across time, though the strongest evidence that music training may support prosocial behaviours in children with hearing loss, comes from one study that relied on anecdotal reports (Innes-Brown et al., 2013). In TH children, the evidence is mixed, with a review by Dumont et al. (2017) finding partially positive findings in 3 studies (one of which was designated as high quality), and another study reporting no benefits. In the current study, total scores on the SDQ were also significantly improved at the post-training time point, but not maintained by follow-up. However, this was likely driven by internalising factors rather than externalising ones which did not significantly differ over time. Given the small sample size and nature of the study, the decision to use the broader internalising and externalising factors was warranted, as findings suggest that not all of the individual SDQ subscales are necessarily distinct (A. Goodman et al., 2010). As such, the broader factors are more suitable, particularly for interpretation.

Measuring QoL in children presents a number of challenges. While best practice is to use parent and self-reported measures in tandem to provide a comprehensive understanding of a child's QoL, many measures (and studies) are designed to rely exclusively on parent reports; particularly if children may not have the capacity (due to age, illness, disability) to reliably self-report (Umansky et al., 2011; Upton, Lawford, & Eiser, 2008). The SDQ is one such case, which is parent-reported for the age range of the present study's cohort. Internalising problems have been noted as being easy to miss by parents, as these behaviours are less visible and obvious than externalised ones (Clarke-Stewart, Allhusen, McDowell, Thelen, & Call, 2003). Nonetheless, irrespective of the sensitivity to observing problems, internalised behaviours were notably improved for up to a 6-month period in this study (as measured from baseline 2 to the follow-up time point).

There was no significant improvement for any PedsQL GCS measure across time. One likely factor was that on average, the parents and the children with hearing loss did not report any difficulties for psychosocial or physical health when compared to their TH peers. Interrater reliability between parents and children for the PedsQL GCS was poor. This was unsurprising, as QoL concordance between

parent and child ratings is highly inconsistent from poor to good (Upton et al., 2008), and these differences reflect separate perspectives, all of which are relevant and valuable (Jozefiak, Larsson, Wichstrøm, Matthejat, & Ravens-Sieberer, 2008; Upton et al., 2008). While studies have often noted that some domains are in better concordance than others (Barker et al., 2009); a review by Upton et al. (2008) found no systematic pattern, or evidence as to why. In the present study, concordance on both psychosocial and physical health domains were similarly poor. However, on the basis of previous findings and given the small sample size, this is not an unexpected finding. Additionally, Looi, Lee, and Loo (2016) used the PedsQL inventory for a cross-sectional study of children with hearing loss and noted that the age-related guidelines for self-reporting may not be applicable for children with hearing loss—given potential language delays. Interestingly, while the children’s scores did not significantly change at any time point in the present study, there was a general negative trajectory across all measures from baseline 1, to the post time point. A possible explanation is that the music program was generally aligned with the start (and end) of the school term, and a potential effect could have been general school fatigue, as children tend to evaluate their QoL in respect to the present moment (Silvey et al., 2014), despite the time reference of the entirety of the previous month being stated to the children.

The HEAR-QL-26 was specifically used, as it is a validated self-reported hearing-related QoL instrument that directly probed: their capacity to hear in a range of daily environments; the effect of hearing on social activity and participation; and how their hearing loss made them feel (environments, activities, and feelings, respectively). There was no significant change across time after training. Given there were some perceptual benefits resulting from the music training¹, it was surprising this did not transfer to better hearing-related QoL. However, while perceptual benefits such as SIN were statistically significant, this may not have had a significant real-world effect or may require a longer time scale to emerge. Additionally, at face-value, questions on the ability to hear in daily environments would likely correlate with SIN perception. However, questions within the environments subscale are all framed in terms of “Is it hard to hear in... your classroom, the cafeteria, etc”, which may be more representative of listening effort as opposed to perception. Additionally, a study by Klatte, Lachmann, and Meis (2010) investigated the effect of noise and reverberation in classroom-like settings with first and third grade TH children. While noise and reverberation had a clear negative effect on their speech perception performance, their subjective appraisals were low, suggesting the children were unable to estimate the

¹ See Manuscript 1: Music for children with hearing loss improves music and speech outcomes.

effect of disruption. However, the generalisability of this to children with hearing loss, and in a range of environments outside the classroom, is unknown. Nonetheless, the large and significant differences between children with hearing loss and TH children reiterates its sensitivity and utility as a hearing-specific QoL measure.

GCBI outcomes were generally positive, with a total average score of 20, 95% CI [8, 31], out of a maximum benefit/harm scale of ± 100 . However, there are no reporting guidelines as to a clinically significant score for the GCBI. As a comparison, Roland et al. (2016) performed a meta-analysis of GCBI outcomes after children received bone-anchored hearing aids (BAHA), with total scores of 43, 95% CI [25, 56]. Overall, we can conclude there is evidence for benefit, and no evidence of any harm. More specifically, the GCBI has factors that broadly consider emotion (e.g. self-confidence, and self-esteem), physical health (e.g. school colds, and doctor visits), learning (e.g. progress and development, concentration), and vitality (e.g. liveliness, and fun with friends). After music training, parents reported that both emotion and learning factors improved, which is partially supported by the improvement for SDQ internalising behaviours, although evidence that music may improve attention and executive functioning (that are broadly associated with learning) in TH children is mixed (Dumont et al., 2017).

There are three broad considerations as to why the children with hearing loss may have improved in internalising behaviours (as measured by the SDQ), as well as learning and emotion factors (as measured by the GCBI). Firstly, participation in the face-to-face group-based music therapy sessions required engagement in activities geared towards turn-taking, coordinated re/action, peer interaction, imitation, and emotional expression. Each of these was key for collective musical success, which (Kirschner Sebastian et al., 2010) argue may encourage shared and cooperative behaviours that are centred around sharing with others—thus improving peer interactions and creating a sense of belonging. Additionally, children were provided the opportunity to develop skills over a variety of musical tasks such as singing, dancing, and playing instruments. This, in turn with opportunities to lead various activities, likely facilitated feeling of competence, and a sense of achievement (Hallam et al., 2016). Secondly, there may have been a specific benefit, primarily for the children who were mainstream schooled. Anecdotally, the majority of mainstream-educated children were the only child with hearing loss at their school, and many studies have established that experiences of loneliness (particularly in mainstream settings) are common (Most, Ingber, & Heled-ariam, 2012; Schorr, 2006). The “hearing aid effect” is noted as a generalised stigma associated with wearing HAs (and CIs) (Cameron et al., 2008), that may result in anxieties around acceptance within peer groups (Punch & Hyde, 2011). As the cohort consisted exclusively of children with hearing loss, this may have been alleviated—resulting in a sense

of belonging. Finally, the improvements may have been a result of better communication skills. A number of perceptual enhancements were noted in this study¹, of most relevance for communication were benefits for SIN perception and question/statement prosody. These results are compatible with the hypothesis that Summerfield and Marshall (1999) put forward, suggesting that short-term outcomes such as communication have a cascading effect on longer-term outcomes such as QoL. However, the time scale that constitutes short-term and long-term effects are not well-defined. Additionally, while SIN and prosodic perception are an important perceptual aspect of communication, this study did not directly assess broader communication ability which consists of myriad elements such as: receptive and expressive language, turn-taking, sustained attention, initiation, pragmatics, and comprehension (Bishop, 1998; Stevenson et al., 2015).

There are several limitations to be discussed in the present study. Without an active control group, it is plausible that improvements were not music-specific and could be attained from any group-based social activity. The small sample size is not unusual for longitudinal paediatric studies of hearing loss investigating interventions, but nonetheless reduced statistical power and generalisability. The benefits were also only noted from the perspective of the parents; as such, it is plausible that parents were biased, based on an expectation that participation in the music training program would yield benefits for their children. On the other hand, the children themselves were likely naïve to the overarching aims of the study. Ultimately, on the balance of evidence, we can only assume the status quo—that all perspectives are valid (Upton et al., 2008).

There is a clear paucity of evidence linking music training to psychosocial and QoL benefits for children with hearing loss. To date, the most compelling evidence was from a study by (Innes-Brown et al. (2013) that relied on anecdotal reports from the children’s music teachers, which are inherently difficult to interpret. The present study provides a valuable contribution, by utilising validated questionnaires that suggest music training can improve psychosocial and HRQoL outcomes for children with hearing loss. The primary areas of improvement were based on internalising behaviours, along with factors associated with emotions and learning. These findings provide the first evidence that the general psychosocial and HRQoL benefits noted in music studies for TH children may be applicable to paediatric populations with hearing loss. While the mechanisms are likely different and at present not clear; overall, the findings suggest a positive effect of group-based musical activity for children with hearing loss. This is encouraging, as they are at greater risk of poorer psychosocial and QoL outcomes (Stevenson et al., 2015; Theunissen et al., 2014). An open question that remains is the time course that one would expect QoL changes to occur. The total duration to observe the effects of music training in the present

study was relatively short, with a maximum time of 6-months to establish significant change from baseline 2 to the follow-up time point; that some improvement was noted is remarkable. Longitudinal studies confirming the reliability of these findings, with larger samples, and across a range of ages are warranted.

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Chapter 6 — Summary and conclusion

The research described in this thesis explored the benefits of music training for children with prelingual SNHL aged between 6 and 9 years. The provision of music involved a 12-week training program that utilised weekly face-to-face group-based music therapy sessions, as well as take-home online apps to be completed 3-times a week. Data was collected from 14 children with SNHL, and 16 TH children and analysed in two manuscripts. Manuscript 1 (Chapter 4) evaluated the perceptual benefits of participation in the program, while Manuscript 2 (Chapter 5) evaluated the benefits from a psychosocial and QoL perspective. Together, these manuscripts present findings to support the initial areas of investigation as described in Chapter 1: Would music training... 1. improve music outcomes such as pitch and timbre perception, and appreciation? 2. enhance psychoacoustic outcomes such as spectral resolution? 3. transfer musical skills to speech domains such as SIN and emotional prosody perception? 4. provide benefits for QoL and psychosocial wellbeing?

1. Music outcomes: pitch, timbre, and appreciation

Pitch is one of the most important and salient cues relevant for music and melody. The results presented in Chapter 4 showed there was no significant improvement to pitch perception, but a large number of factors likely contributed. Firstly, the focus of the music training was to provide a wide variety of musical activities that broadly began with simpler tasks of rhythm, followed by intermediate tasks of timbre, followed by more complex tasks of pitch. As such, the time dedicated to pitch-related activities may not have been sufficient for change. This is supported by findings by Chen et al. (2010) in which a significant correlation was found between the duration spent enrolled in Yamaha Music School classes (that focussed on instrumental training) and pitch perception. Given some students were provided as much as 36 months of music training ($M=13.2$), it is likely that pitch-related changes for children with SNHL may require a lengthier, more pitch-focused instruction for such gains to be made apparent. Chapter 4 also demonstrated that pitch perception performance was not significantly different between children with SNHL and those with TH. This was likely due to age and developmental issues, as pitch is not matured until 11 years in TH children (Lamont, 1998), or early adolescence for more complex harmonic stimuli (Trainor & Unrau, 2012). This interpretation is also supported by the statistical models that indicated that hearing age was associated with pitch perception.

Timbre is a multidimensional attribute that relies on spectro-temporal cues to identify and separate different sounds (the focus of which were instruments in the present study). One of the key benefits shown in Chapter 4 was for timbre perception that improved significantly in behavioural measures by 8 percentage points. This was also supported by participants' subjective appraisals that they believed they were better at identifying instruments after training. Interestingly, post-hoc analyses showed a moderate correlation between timbre and SIN perception, providing some insight as to a potential mechanism for SIN enhancement. Considering most adult hearing loss studies focus on pitch-based training and associated benefits,

Finally, Chapter 4 also considered measures of music appreciation that consider the role of subjective appraisals that are not necessarily correlated with perceptual accuracy. This consideration of appreciation is important as children with SNHL are unlikely to achieve the same level of perceptual accuracy that TH children do, but this may not diminish the level of enjoyment derived from music (Chen-Hafteck & Schraer-Joiner, 2011; Gfeller et al., 2011). Overall, music appreciation measures improved significantly. Children with SNHL reported music as sounding more pleasant, noted an improved ability to identify instruments, and encouragingly, were keen to learn, or continue learning an instrument.

2. Psychoacoustic outcomes: spectral resolution

Spectral resolution is often used as a non-linguistic proxy measure of speech perception and the removal of linguistic confounds is particularly useful when testing children. However, while there appears to be a robust association with speech outcomes for postlingually implanted adult CI recipients, its association for prelingual children with hearing loss is less established. Interestingly the findings from Chapter 4 showed children with hearing loss improved their spectral resolution abilities by a significant margin (2 rpo) and the benefits were retained at follow-up. However, post-hoc analyses indicated that spectral resolution was not significantly correlated with any other perceptual measure. As such, better spectral resolution provided no evidence it was associated with other perceptual or communication outcomes. This warrants further investigation, as it is likely that children with prelingual SNHL have developed cue-weighting strategies that utilise and bias temporal envelope more than spectral cues. This is supported by findings by Landsberger et al. (2017) that indicated that spectral resolution did not improve as a function of age in children with hearing loss, as it did in TH children. As such, better spectral resolution may lead to benefits, but changes to cue-weighting strategies are likely to be

developed over a much longer-time course (Nitttrouer et al., 2014). The choice of test for spectral resolution was the SMRT, given it was designed to avoid potential confounding cues such as loudness, and had previously been used in child studies. However, it is also plausible that the SMRT may also be subject to a number of unknown confounds.

3. Transfer effects: SIN and prosody outcomes

One of the motivations to utilise music training is its effectiveness in developing spectral and temporal sensitivity (Kraus & Chandrasekaran, 2010). In turn, this may have a generalisable effect that benefits broader auditory abilities, and more specifically—speech perception. Benefits to SIN perception have been observed in TH adults (Coffey et al., 2017) and children (Slater et al., 2015). Encouragingly, the results from Chapter 4 indicated a significant improvement to SIN performance of 1.1 dB SRT that was also retained at the follow-up time point. Considering SIN difficulties are a primary concern for children with hearing loss (Davies et al., 2001; Schafer & Thibodeau, 2006), these findings highlight the potential benefits and utility of music training. The significant correlations between SIN and timbre perception suggest a shared or underlying mechanism; or a benefit for SIN performance in terms of auditory scene analysis due to better separation of target from masker signals. Future studies exploring the specific mechanisms of music that enhance SIN in more controlled studies may be particularly informative.

Chapter 4 also explored prosody perception, which plays an important function for emotive communication in social contexts. It was hypothesised that prosodic tasks would likely benefit from music training, as there is a growing body of evidence from studies for adult CI recipients (Lo et al., 2015) and children with CIs (Good et al., 2017). Prosody was measured in two ways; emotional prosody through semantically neutral sentences with 2-AFC happy/sad and angry/scared contrasts, and question/statement prosody that utilised single, bisyllabic words. While emotional prosody did not show improvement—likely because many participants were at ceiling performance at baseline—question/statement prosody showed a large improvement of 14 percentage points. Prosody relies primarily on pitch and although there was no benefit for pitch threshold (when measured as discrete pitch intervals), the improvement to question/statement prosody suggests other pitch-based abilities such as melodic contour recognition may have improved.

4. Quality of life and psychosocial wellbeing

QoL and psychosocial wellbeing are highly variable among children with hearing loss, but outcomes are generally poorer when compared to TH peers (Stevenson et al., 2015; Theunissen et al., 2014). There are a small number of studies that indicate potential benefits for TH children—primarily for prosocial skills and cooperative behaviours (Dumont et al., 2017). However, the evidence that music training has a similar benefit for children with hearing loss is almost non-existent. Thus, the findings from Chapter 5 provide some of the first evidence for QoL and psychosocial benefits from music training for children with hearing loss.

The primary finding was that parents reported internalising problems were significantly reduced after music training and retained at follow-up, with additional benefits for behavioural changes relating to emotional and learning factors. There was no improvement for any of the PedsQL GCS measures, and most surprisingly, no improvement for the HEAR-QL. As the HEAR-QL was designed to specifically measure the QoL for children with hearing loss, the expectation was that this questionnaire would be the most sensitive to measuring change. Given there were perceptual benefits, a positive change was expected in the HEAR-QL measures. However, a plausible explanation is that the statistically significant results with generally small effect sizes do not lead to significant real world benefit. Overall, these results are a positive first step. Three broad considerations were proposed theorising why such benefits occurred. Firstly, face-to-face group-based activities actively encouraged cooperation, peer interaction, and social cohesion that may directly transfer into behaviours outside the music classes (Kirschner Sebastian et al., 2010). Secondly, these interactions may have been particularly beneficial as most participants were anecdotally reported as being the only child with hearing loss in their mainstream school—which may lead to feelings of loneliness, particularly within their school environment (Most et al., 2012; Schorr, 2006). Musical activities that brought together children with hearing loss may have created a sense of belonging. Finally, considering better outcomes of speech perception such as SIN and question/statement prosody were noted in Chapter 4, improvements may be attributable to generalised enhancement of communication skills. This is in line with the general theory that communication is the gateway to social competence and subsequent social activity (Hoffman et al., 2015).

Limitations

The lack of an active control group and small sample size are obvious limitations for both studies as presented in Chapters 4 and 5. These two limitations are related. Randomised, double-blinded studies with active control groups remain the “gold standard” for experimental design. One of the more robust designs regarding music interventions and hearing loss, was a study by Fuller et al. (2018) that compared the effects of two music training programs and an active control group. However, the study was not blinded, had adult participants, was completed over a shorter 8-week duration, and was underpowered due to the division of the cohort into 3 groups. Due to practical reasons regarding their family’s commitments, the children with SNHL were not completely randomised to the music or waitlisted group. Nonetheless, as the trajectory of results was similar, the pseudo-random assignment was not thought to have contributed to any significant difference between the groups. The heterogeneity of hearing devices across children in conjunction with the unbalanced number of devices within the cohort was also a limitation, though some efforts were made to reduce such effects by accounting for hearing device as a factor in the statistical modelling. Difficulties in recruiting children with hearing loss for participation in a longitudinal training study (up to 36-weeks total duration from baseline 1 to follow-up for the waitlisted group) necessitated an alternative approach, as equivalent outcomes between small groups were unlikely to occur due to high variability. As such, the use of a waitlisted design, with the use of double-baseline measures was in part pragmatic, but also provided additional data. Measures of natural development and maturation are not typically reported, but are an important consideration for paediatric studies, and the robust factoring of hearing-age across all statistical models was an important factor to control for.

Future directions

This thesis provides novel contributions that indicate significant benefits of music training for children with hearing loss. It is significant that the music training program was only modestly adapted for children with hearing loss; that is, this study has evaluated the benefits of participation in a music training program that is already readily available. Based on the findings presented in Chapters 4, the enhancement of SIN and corresponding mechanisms are prime areas for future investigation. The findings on spectral resolution are also noteworthy, and future studies clarifying its role in perceptual tasks, specifically for children with prelingual hearing loss are likely to yield interesting results.

The development of a test battery that is both comprehensive of all the questions a researcher wants addressed and suitable for children, can seemingly be at odds. Overall, the test battery was mostly successful with the notable exception being the length of the pitch task, which on average, took each participant between 6 and 10 minutes to finish, excluding any breaks in-between. The pitch subtest from the CAMP test used an adaptive 2AFC paradigm to determine participants' thresholds. Halving the length (and thus number of tokens) would be ideal and make the test more suitable for children, although pilot testing would be required to ensure the standard deviations are not unreliable. Alternatively, using a 3AFC paradigm with only one choice being unique and correct, such as in the SMRT (which was a generally enjoyable task) would likely be a suitable test paradigm. Finally, the decision to use a touchscreen for participants' inputs likely resulted in a much faster and easier option for the children to operate as opposed to with a mouse.

Given the broadness of music, additional studies with different perspectives will be required to further our understanding. This is particularly true for evaluating different types of music training and corresponding causal mechanisms. This thesis made broad considerations on the basis that music perception may lead to better auditory skills, which in turn may improve speech perception. Additionally, psychosocial and QoL benefits may have resulted due to better speech perception ability, or due to a generalised improvement to cooperative behaviours and feelings of connectedness. However, future studies may consider: the role of language, given it remains a central concern; measures of statistical learning ability, given the evidence that musicians have superior performance and its potential contribution to activities such as language (Arciuli & von Koss Torkildsen, 2012); and cognitive factors such as working memory and attention that are mediators of language and speech perception (Pisoni & Cleary, 2003). Music training may also have additional and specific benefits for tonal-language development; and the relationship between music perception and production is another area for future investigations. Finally, considering benefits for perception and psychosocial wellbeing were noted in Chapters 4 and 5, an obvious area to consider is the relationship between perceptual abilities and psychosocial wellbeing—especially as this area has been mostly unexplored (Schorr et al., 2009).

Conclusion

The beautiful thing about music is the incredible depth as to what music constitutes, and what music does. Music is all-encompassing and packaged in a manner that is accessible, enjoyable, and beneficial across a wide range of outcomes for children with hearing loss. As Turner (1848 p.5–6) put forward; “*Cui bono?* What possible benefit can result from teaching music to the deaf or from exercising them in musical performance when learned?” Some 170-years later, I can respond with some confidence—the benefits are wide-ranging and profound! Children with hearing loss engage and enjoy music to the same extent as any child, with potential benefits for music perception, speech perception, music appreciation, psychosocial wellbeing, and QoL. Remarkably, despite a relatively short training period of 12-weeks, the SIN benefits are similar to those reported in longer training paradigms for children with TH, and the prospect of investigating the long-term benefits of music training for children with hearing loss are both necessary and appealing. The findings reported in this thesis suggest music is an excellent complementary form of habilitation, and children with hearing loss should be encouraged to participate in musical activities.

Anecdotally, many parents in this study cited that representatives at their children’s school were actively discouraging their child’s participation in musical programs. This was often on the basis of poor singing ability in the choir, or difficulties when performing in an ensemble due to noise. This is unacceptable, and provisions must be made to ensure participation in musical activities is equitable. I would argue that the vast majority of children in a choir have sub-optimal singing ability—but this perspective fails to acknowledge the benefits beyond perceptual skills such as music appreciation and social participation. Additionally, strategies such as breaking the ensemble into smaller groups may be a simple solution. On a more positive note, I was encouraged by the considerable interest in this study throughout Australia (we were of course, only able to accommodate those who could travel to Macquarie University). There is clearly a want in the community for activities of this nature. I was also impressed by the number of parents that were actively enquiring about further music training and what instrument may be suitable for their child.

In conclusion, this thesis provides robust evidence that active participation in music training is not just an enjoyable activity, but one that may significantly improve many aspects of life for children with hearing loss.

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Appendix A — Ethics

Office of the Deputy Vice-Chancellor
(Research)

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MACQUARIE
University
SYDNEY • AUSTRALIA

15 July 2016

Dear Associate Professor McMahon

Reference No: 5201600081

Title: *Benefits of a music training program for children with hearing loss*

Thank you for submitting the above application for ethical and scientific review. Your application was considered by the Macquarie University Human Research Ethics Committee (HREC (Medical Sciences)).

I am pleased to advise that ethical and scientific approval has been granted for this project to be conducted at:

- Macquarie University

This research meets the requirements set out in the *National Statement on Ethical Conduct in Human Research* (2007 – Updated May 2015) (the *National Statement*).

Standard Conditions of Approval:

1. Continuing compliance with the requirements of the *National Statement*, which is available at the following website:

<http://www.nhmrc.gov.au/book/national-statement-ethical-conduct-human-research>

2. This approval is valid for five (5) years, subject to the submission of annual reports. Please submit your reports on the anniversary of the approval for this protocol.

3. All adverse events, including events which might affect the continued ethical and scientific acceptability of the project, must be reported to the HREC within 72 hours.

4. Proposed changes to the protocol and associated documents must be submitted to the Committee for approval before implementation.

It is the responsibility of the Chief investigator to retain a copy of all documentation related to this project and to forward a copy of this approval letter to all personnel listed on the project.

Should you have any queries regarding your project, please contact the Ethics Secretariat on 9850 4194 or by email ethics.secretariat@mq.edu.au

The HREC (Medical Sciences) Terms of Reference and Standard Operating Procedures are available from the Research Office website at:

http://www.research.mq.edu.au/for/researchers/how_to_obtain_ethics_approval/human_research_ethics

The HREC (Medical Sciences) wishes you every success in your research.

Yours sincerely



Professor Tony Eyers

Chair, Macquarie University Human Research Ethics Committee (Medical Sciences)

This HREC is constituted and operates in accordance with the National Health and Medical Research Council's (NHMRC) *National Statement on Ethical Conduct in Human Research* (2007) and the *CPMP/ICH Note for Guidance on Good Clinical Practice*.

Details of this approval are as follows:

Approval Date: 12 July 2016

The following documentation has been reviewed and approved by the HREC (Medical Sciences):

Documents reviewed	Version no.	Date
Macquarie University Ethics Application Form		Received 10/02/2016
Correspondence responding to the issues raised by the HREC (Medical Sciences)		Received 06/07/2016
MQ Participant Information and Consent Form (PICF)	2	06/07/2016
Parent/Guardian Contact Letter	1	06/07/2016
Recruitment Flyer	1	06/07/2016
Summary of Recruitment Assistance Across Organisations	1	06/07/2016
Participant Questionnaire	1	10/02/2016

***If the document has no version date listed one will be created for you. Please ensure the footer of these documents are updated to include this version date to ensure ongoing version control.**

Phone: +61 (2) 9850 8106

Fax: +61 (2) 9850 9199

Email: chi.lo@mq.edu.au

RE: Benefits of music training for children with hearing loss

Dear Parent/Guardian,

You are invited to participate in a study of music training. The purpose of the study is to determine the benefits that music may provide for children aged 6 to 11 years with hearing loss. We anticipate benefits for music perception, music appreciation, speech perception, and social wellbeing.

If you decide to participate, your child will be provided with free music lessons using apps, as well as group-based music therapy sessions. The duration of the training is 3 months, in which your child will engage with musical tasks such as: matching melodies, learning about instruments, and creating music. The commencement is around March and July 2017.

The study is being conducted by Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au) to meet the requirements of a PhD thesis, under the supervision of A/Prof Catherine M. McMahon (ph: 9850 8775; cath.mcmahon@mq.edu.au) from the Department of Linguistics, Prof Bill Thompson (ph: 9850 4083; bill.thompson@mq.edu.au) from the Department of Psychology, and Dr Valerie Looi (valerie.looi@scic.org.au), from the SCIC Cochlear Implant Program, an RIDBC Service.

Participation in this study is entirely voluntary: you are not obliged to participate and if you decide to participate, you are free to withdraw at any time without having to give a reason and without consequence. If you choose not to participate or withdraw at any time, this will in no way affect your care at Australian Hearing, SCIC, RIDBC, The Shepherd Centre, Macquarie University, or any other place of treatment.

If you are interested in participating in this study, or have further questions, please contact Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au).

Thank you for your consideration, it is greatly appreciated.



Chi Yhun Lo

Phone: +61 (2) 9850 8106

Fax: +61 (2) 9850 9199

Email: chi.lo@mq.edu.au

RE: Benefits of music training for children with hearing loss

Dear Parent/Guardian,

You are invited to participate in a study of music training. The purpose of the study is to determine the benefits that music may provide for children aged 6 to 11 years with hearing loss. This may include benefits for music perception, music appreciation, speech perception, and/or social wellbeing.

If you decide to participate, your child will be provided with free music lessons using apps, as well as group-based music therapy sessions. The duration of the training is 3 months, in which your child will engage with musical tasks such as: matching melodies, learning about instruments, and creating music. A computer or smart device with internet access is required. The commencement is around March and July 2017, and you will also receive \$30 to assist with travel costs

You may have already received an invitation to this study in 2016. However, we have made several changes to the project, hence the reason for re-contacting you. The study is now supported by Nordoff-Robbins who will provide the music therapy sessions free of charge to you, the age criteria has been expanded, and the duration reduced to 3 months.

The study is being conducted by Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au) to meet the requirements of a PhD thesis, under the supervision of A/Prof Catherine M. McMahon (ph: 9850 8775; cath.mcmahon@mq.edu.au) from the Department of Linguistics, Prof Bill Thompson (ph: 9850 4083; bill.thompson@mq.edu.au) from the Department of Psychology, and Dr Valerie Looi (valerie.looi@scic.org.au), from the SCIC Cochlear Implant Program, an RIDBC Service.

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If you are interested in participating in this study, or have further questions, please contact Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au).

Thank you for your consideration, it is greatly appreciated.



Chi Yhun Lo

Phone: +61 (0)2 9850 8775
Fax: +61 (0)2 9850 9199
Email: cath.mcmahon@mq.edu.au

Supervisor: A/Prof Catherine M. McMahon

Participant Information and Consent Form

Name of Project: Benefits of music training for children with hearing loss

You and your child are invited to participate in a study of music training. The purpose of the study is to determine the benefits that music may provide for children with hearing loss. We anticipate benefits for music perception, music appreciation, speech perception, and social wellbeing.

The study is being conducted by Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au) to meet the requirements of a PhD thesis, under the supervision of A/Prof Catherine M. McMahon (ph: 9850 8775; cath.mcmahon@mq.edu.au) from the Department of Linguistics, Prof Bill Thompson (ph: 9850 4083; bill.thompson@mq.edu.au) from the Department of Psychology, and Dr Valerie Looi (valerie.looi@scic.org.au), from the SCIC Cochlear Implant Program, an RIDBC Service.

If you decide to participate, your child will be asked to play a series of MusicFirst apps from home, and attend face-to-face group-based music classes at the Australian Hearing Hub, Macquarie University, where free parking is available. Your child will engage with musical tasks such as: matching melodies, learning about instruments, and creating music. The duration of the music training is 3 months. Training with MusicFirst apps will take approximately 30 minutes a day, 3 times a week. There will also weekly face-to-face group-based music therapy classes run by Nordoff-Robbins Music Therapy Australian that will be recorded (audio and video). There are 5 x 1 hour test sessions that will measure the performance of your child's musical, speech perception, and social abilities. For your convenience, some of these test sessions can coincide with the music therapy classes. Finally, you and your child will be asked to answer a set of questionnaires that provide information about your child's quality of life, and the quality of hearing and communication. After the training has been completed, there will be one follow-up test session 3 months later. There are no perceived or actual risks to participating in this study.

Any information or personal details gathered in the course of the study are confidential, except as required by law. Recordings will only be accessible to the music therapist to monitor progress, and may be presented by the researchers to the scientific community at conferences. All other data will be de-identified and only made accessible to the researchers named above. All data is securely stored and retained for a period of 5 years. A summary of the results of the data can be made available to you on request by contacting Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au)

Participation in this study is entirely voluntary: you are not obliged to participate and if you decide to participate, you are free to withdraw at any time without having to give a reason and without consequence. If you choose not to participate or withdraw at any time, this will in no way affect your care at the SCIC, RIDBC, Australian Hearing, The Shepherd Centre, Macquarie University, or any other place of treatment.

Consent of participants under 18 years is to be obtained through a parent or legal guardian.

I (parent/guardian's name), _____ the parent/legal guardian of (child's name), _____ give permission for my child to participate in this study. I understand the information above and any questions I have asked have been answered to my satisfaction. I agree to participate in this research, knowing that I can withdraw from further participation in the research at any time without consequence. I have been given a copy of this form to keep.

☐ I have read and understand the information above and any questions I have asked have been answered to my satisfaction.

☐ I agree for my child/ren and myself to participate in this research, knowing that I can withdraw from further participation in the research at any time without consequence.

☐ I agree for my child/ren and myself to be recorded during face-to-face music classes.

☐ I agree for my audio and video recordings to be used for conference presentations.

☐ I agree to be contacted about future research.

☐ I have been given a copy of this form to keep.

Participant's Name: _____
(Block letters)

Participant's Signature: _____ Date: _____

Investigator's Name: _____
(Block letters)

Investigator's Signature: _____ Date: _____

The ethical aspects of this study have been approved by the Macquarie University Human Research Ethics Committee. If you have any complaints or reservations about any ethical aspect of your participation in this research, you may contact the Committee through the Director, Research Ethics & Integrity (telephone (02) 9850 7854; email ethics@mq.edu.au). Any complaint you make will be treated in confidence and investigated, and you will be informed of the outcome.

(INVESTIGATOR'S [OR PARTICIPANT'S] COPY)

Phone: +61 (0)2 9850 8775
Fax: +61 (0)2 9850 9199
Email: cath.mcmahon@mq.edu.au

Supervisor: A/Prof Catherine M. McMahon

Participant Information and Consent Form

Name of Project: Benefits of music training for children

You and your child are invited to participate in a study of music and language. The purpose of the study is to understand the connections between musical skills and language abilities in children.

The study is being conducted by Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au) to meet the requirements of a PhD thesis, under the supervision of A/Prof Catherine M. McMahon (ph: 9850 8775; cath.mcmahon@mq.edu.au) from the Department of Linguistics, Prof Bill Thompson (ph: 9850 4083; bill.thompson@mq.edu.au) from the Department of Psychology, and Dr Valerie Looi (valerie.looi@scic.org.au), from the SCIC Cochlear Implant Program, an RIDBC Service.

If you decide to participate, your child will attend 2 x 1 hour test sessions that will measure the performance of your child's musical, speech perception, and social abilities over the course of 12 weeks. Additionally, your child will undergo a basic hearing screening test. Finally, you and your child will be asked to answer a set of questionnaires that provide information about your child's quality of life, and the quality of hearing and communication. There are no perceived or actual risks to participating in this study.

Any information or personal details gathered in the course of the study are confidential, except as required by law. All data will be de-identified and only made accessible to the researchers named above. All data is securely stored and retained for a period of 5 years. A summary of the results of the data can be made available to you on request by contacting Chi Yhun Lo (ph: 9850 8106; chi.lo@mq.edu.au).

Participation in this study is entirely voluntary: you are not obliged to participate and if you decide to participate, you are free to withdraw at any time without having to give a reason and without consequence. If you choose not to participate or withdraw at any time, this will in no way affect your care at Macquarie University, or any other place of treatment.

Consent of participants under 18 years is to be obtained through a parent or legal guardian.

I (parent/guardian's name), _____ the parent/legal guardian of (child's name), _____ give permission for my child to participate in this study. I understand the information above and any questions I have asked has been answered to my satisfaction. I agree to participate in this research, knowing that I can withdraw from further participation in the research at any time without consequence. I have been given a copy of this form to keep.

☐ I have read and understand the information above and any questions I have asked has been answered to my satisfaction.

☐ I agree for my child/ren and myself to participate in this research, knowing that I can withdraw from further participation in the research at any time without consequence.

☐ I agree to be contacted about future research.

☐ I have been given a copy of this form to keep.

Participant's Name: _____
(Block letters)

Participant's Signature: _____ Date: _____

Investigator's Name: _____
(Block letters)

Investigator's Signature: _____ Date: _____

The ethical aspects of this study have been approved by the Macquarie University Human Research Ethics Committee. If you have any complaints or reservations about any ethical aspect of your participation in this research, you may contact the Committee through the Director, Research Ethics & Integrity (telephone (02) 9850 7854; email ethics@mq.edu.au). Any complaint you make will be treated in confidence and investigated, and you will be informed of the outcome.

(INVESTIGATOR'S [OR PARTICIPANT'S] COPY)



Free music lessons for children with hearing loss

CHILDREN AGED 6 TO 11 YEARS WITH
COCHLEAR IMPLANTS AND/OR HEARING AIDS

What your child will do:

Play musical apps at home
(30 minutes a day, 3 times a week)

Face-to-face group music therapy
(12 sessions, once a week)

Test sessions
(5 sessions, 1 hour each)

Other:

3 month duration
(Starting August 2017, enrol by 8th May)

Weekly sessions at Macquarie University
Saturday (9:30 to 10:00, and 10:15 to 10:45).
Weekday afternoon (subject to interest)

Receive \$15 at commencement and conclusion

Chi Yhun Lo
T: +61 (2) 9850 8106
E: chi.lo@mq.edu.au



COCHLEAR
IMPLANT PROGRAM
An RIDBC service



MACQUARIE
University



Help us understand the connection between music and language

CHILDREN AGED 6 TO 9 YEARS WITH COCHLEAR
IMPLANTS AND/OR HEARING AIDS

The study involves two test sessions (1 hour each, separated by 3 months) in which
your child will play various listening activities.

You will receive \$15 at the first and second sessions.

Testing is on-site at Macquarie University.

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Help us understand the connection between music and language

CHILDREN AGED 6 TO 9 YEARS

The study involves two test sessions (1 hour each, separated by 3 months) in which
your child will play various listening activities.

They will receive a free hearing test, and \$15 at the first and second sessions.

Testing is on-site at Macquarie University.

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MACQUARIE
University

Appendix B — Curriculum

Week 1 — Music therapy

Activities	Equipment	Goals
Hello	Guitar	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
Drum <ul style="list-style-type: none"> • raindrop to thunder 		<ul style="list-style-type: none"> • Explore dynamics • Creative expressions (modes of playing) • Working as a team
<ul style="list-style-type: none"> • Patterns (rhythm + modes) 		<ul style="list-style-type: none"> • Auditory memory
“Shake” <ul style="list-style-type: none"> • Choices of instruments • Position • Body parts 		<ul style="list-style-type: none"> • Choices of contrasting sounds • Concepts (auditory): start/stop; position; body parts • Listen for single step instructions with music • Creative movement (dance)
“I have a sound”		<ul style="list-style-type: none"> • Confidence • Explore vocal sounds • Expand range of vocal sounds • Leader-follower • Relationship
Aeroplane	Paper plane	<ul style="list-style-type: none"> • Creative vocal expressions • Confidence in leading in a group
Parachute		<ul style="list-style-type: none"> • Auditory discrimination (fast/slow; loud/soft) • teamwork
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 2 — Music therapy

Activities	Equipment	Goals
Hello	Guitar	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
Drum <ul style="list-style-type: none"> • Recap raindrop to thunder • Pass round the circle (2 directions) • Pass with eyes closed 		<ul style="list-style-type: none"> • Explore dynamics • Creative expressions (modes of playing) • Working as a team • Directional sounds
“Shake” <ul style="list-style-type: none"> • Choices of instruments • Position • Body parts • (eyes closed) • Listen for single step instructions with music (from MT and peers) 		<ul style="list-style-type: none"> • Choices of contrasting sounds • Concepts (auditory): start/stop; • Position (up/down; R/L; front/back); • Body parts • Creative movement (dance)
“I have a sound” (with mic)		<ul style="list-style-type: none"> • Confidence • Explore vocal sounds • Expand range of vocal sounds • Leader-follower • Relationship
Aeroplane	Paper plane	<ul style="list-style-type: none"> • Creative vocal expressions • Confidence in leading in a group
Parachute		<ul style="list-style-type: none"> • Auditory discrimination (fast/slow; loud/soft)
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Call-response • Relating as a group—social skills

Week 3 — Music therapy

Activities	Equipment	Goals
Hello	Guitar	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm) • volume increase
Drum <ul style="list-style-type: none"> • ‘I LIKE ...’ • Recall others’ likings • Pass round the circle (2 directions) • Pass with eyes closed 		<ul style="list-style-type: none"> • Explore speech rhythms • Creative expressions (modes of playing) • Working as a team • directional sounds
“Shake” <ul style="list-style-type: none"> • Guess what instrument • Choices of instruments • Position • Body parts • loud/soft highlight 	3 pairs: Shaker Cabasa Castanet Clapper Jingle stick Bells	<ul style="list-style-type: none"> • Sound discrimination • Concepts (auditory): start/stop; • Position (up/down; R/L; front/back; side to side); body parts • Listen for single step instructions with music • (from MT and peers)
“I have a sound” (with mic)	Long/short sounds High/low sounds taught	<ul style="list-style-type: none"> • Confidence • Explore vocal sounds • Expand range of vocal sounds • Leader-follower • Relationship
Parachute <ul style="list-style-type: none"> • Rotate seats when music stops 	Range of pitches	<ul style="list-style-type: none"> • Auditory discrimination (fast/slow; loud/soft) • Teamwork • Add high/low movement correspond with pitch
“It’s time to go now”		<ul style="list-style-type: none"> • Pitch and rhythm discrimination • Call-response • Volume increase

Week 4 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Guitar Pitch chart Horn	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
“Shake” <ul style="list-style-type: none"> • Choices of instruments • Position • Body parts 	Guess the sound bag Discuss qualities	<ul style="list-style-type: none"> • Choices of contrasting sounds • Concepts (auditory): start/stop; position; body parts • Listen for single step instructions with music
<ul style="list-style-type: none"> • Drum • Speech pattern “I like...” • Pattern up to 3–4 sounds (rhythms, modes) 	Group 2 with noise	<ul style="list-style-type: none"> • Auditory memory • Rhythm • Social
Movement with pitch (up and down)	Strings	<ul style="list-style-type: none"> • Pitch perception
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 5 — Music therapy

Activities	Equipment	Goals
Hello (re-position) 3 horns each	Guitar Pitch chart Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
Drum <ul style="list-style-type: none"> • Speech pattern about what they did in holidays • Pattern up to 3–4 sounds (rhythms, modes) 	L/S F/S Sounds of diff emotions	<ul style="list-style-type: none"> • Auditory memory • Rhythm • Social
“Shake” <ul style="list-style-type: none"> • Choices of instruments • Position • Body parts No visual cue 	Discuss about the sound Blindfold	<ul style="list-style-type: none"> • Choices of contrasting sounds • Concepts (auditory): start/stop; position; body parts • Listen for single step instructions with music
Movement with pitch (up and down)	Strings Keyboard <ul style="list-style-type: none"> • Octaves • 6ths 	<ul style="list-style-type: none"> • Pitch perception
Emotions sing “If you are...”	Puppet or instruments	
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 6 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Guitar Pitch chart Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
Drum <ul style="list-style-type: none"> • Speech pattern “I like ...” over drum beat 	Bongos	<ul style="list-style-type: none"> • Auditory • Sharing/team • Rhythm
“Shake” <ul style="list-style-type: none"> • Choices of instruments • Position • Body parts • No visual cue 	Discuss about the sound Blindfold	<ul style="list-style-type: none"> • Contrasting sounds • Concepts (auditory): start/stop; position; body parts • Listen for single step instructions with music
“I can Sing” (with mic)		<ul style="list-style-type: none"> • Long/short sounds • High/low sounds—increase awareness and execution of pitch range • Leader-follower relationship
Movement with pitch (up and down) 5ths and 3rds	Strings Keyboard / Vocal—continuous sound	<ul style="list-style-type: none"> • Pitch perception
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 7 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Guitar Pitch chart Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • Singing (pitch; rhythm)
Movement with pitch (up and down) 5ths, 3rds and 2nds	Strings Keyboard / Vocal – continuous sound	<ul style="list-style-type: none"> • Pitch perception
Drum <ul style="list-style-type: none"> • Speech pattern “activities during the week” over drum beat • “Scared” sounds 	Bongos	<ul style="list-style-type: none"> • Auditory • Sharing/team (pairs) • Rhythm
Percussions location	Discuss about the sound Blindfold	<ul style="list-style-type: none"> • Distinguish sounds • Listen for directs
Feelings song: “If you are happy/sad/surprised” <ul style="list-style-type: none"> • Pretend sounds 	Puppets	<ul style="list-style-type: none"> • Sing • Sing with different tone of voice/speed
Keyboard improvisation with emotions—happy		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with percussion
“It’s time to go now”	With gesture cues	<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 8 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Pitch chart No guitar Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • “Same” pitch
Movement with pitch (up and down) 5ths, 3rds, same, (2nds) <ul style="list-style-type: none"> • Each SINGS for others 	Strings; Keyboard / Vocal – continuous sound	<ul style="list-style-type: none"> • Pitch perception • Lead the singing
Drum <ul style="list-style-type: none"> • Speech pattern “What makes you...” over drum beat 	Bongos	<ul style="list-style-type: none"> • Auditory • Sharing/team • Rhythm
Percussions Location <ul style="list-style-type: none"> • 2 instruments 	Blindfold	<ul style="list-style-type: none"> • Distinguish sounds • Listen for directs
Feelings song: “There are times...” (new song) <ul style="list-style-type: none"> • Say the sentence “This is a stick” 	Puppets	<ul style="list-style-type: none"> • Sing • Sing with different tone of voice/speed • Say the sentence “This is a stick” in various emotions
Keyboard improvisation with emotions (sad)		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with vocals/percussion
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 9 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Pitch chart No guitar Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • “Same” pitch
Movement with pitch (up and down) 5ths, 3rds, same, (2nds) <ul style="list-style-type: none"> • Each SINGS for others 	Strings Vocal	<ul style="list-style-type: none"> • Pitch perception • Lead the singing
Drum <ul style="list-style-type: none"> • With “feelings” pic 	Bongos	<ul style="list-style-type: none"> • Auditory speech over sound • Sharing/team
Percussions Location <ul style="list-style-type: none"> • 2 instruments 	Blindfold	<ul style="list-style-type: none"> • Distinguish sounds • Listen for directs
Feelings song: “There are times...” <ul style="list-style-type: none"> • Say the sentence “This is a stick” in various emotional context 	Puppets	<ul style="list-style-type: none"> • Sing • Sing with different tone of voice/speed
Keyboard improvisation with emotions		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with percussion
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 10 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Pitch chart No guitar Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • “Same” pitch
Aeroplane point up/down Sing “HELLO” Descending or ascending	Vocal	<ul style="list-style-type: none"> • Pitch perception • Sing the perceived ascending/descending interval
Drum <ul style="list-style-type: none"> • I feel... when I... 	Bongos Feelings cards	<ul style="list-style-type: none"> • Auditory –speech over sound • Sharing/ team • Feelings
Percussions Location <ul style="list-style-type: none"> • Cymbal/ratchet/tambourine/2-tone block 	Blindfold	<ul style="list-style-type: none"> • Distinguish sounds • Listen for directs
Feelings song: “There are times...” <ul style="list-style-type: none"> • Say the sentence “This is a stick” with various emotions 	Puppets	<ul style="list-style-type: none"> • Sing • Sing with different tone of voice/speed/
Keyboard improvisation with emotions (scared)		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with vocals/ percussion
Do Re Mi	Bells	<ul style="list-style-type: none"> • Team-work • Sing
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 11 — Music therapy

Activities	Equipment	Goals
Hello (re-position)	Pitch chart No guitar Horns	<ul style="list-style-type: none"> • Social skill (acknowledge the members) • “Same” pitch
Aeroplane point up/down Sing “HELLO” Descending or ascending	Vocal	<ul style="list-style-type: none"> • Pitch perception • Sing the perceived ascending/descending interval
Drum <ul style="list-style-type: none"> • Express and guess 	Feelings cards	<ul style="list-style-type: none"> • Sharing/team • Feelings
Feelings song: “There are times...” <ul style="list-style-type: none"> • Say the sentence “This is a stick” with various emotions 	Puppets	<ul style="list-style-type: none"> • Sing • Sing with different tone of voice/speed/
Keyboard improvisation with emotions (choice)		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with percussion
Do Re Mi	Bells	<ul style="list-style-type: none"> • Team-work • Pitch
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 12 — Music therapy

Activities	Equipment	Goals
Hello	Pitch chart No guitar Horns (random allocation)	<ul style="list-style-type: none"> • Team work • “Same” pitch
Aeroplane point up/down Sing “HELLO” Descending or ascending	Vocal	<ul style="list-style-type: none"> • Pitch perception • Sing the perceived ascending/descending interval
Choice of instruments improvisation with emotions (choice)		<ul style="list-style-type: none"> • Creative expressions • Others match sounds with percussion
Do Re Mi	Bells	<ul style="list-style-type: none"> • Team-work • Pitch
“It’s time to go now”		<ul style="list-style-type: none"> • Singing • Pitch and rhythm discrimination • Relating as a group—social skills

Week 1 — Music apps

Families were introduced the music apps, provided login details, software installation, and training.

Week 2 — Music apps

Week 2 was used as a technical test. With all the login details distributed, and software installed, only one app was enabled (Morton Subotnick's Music Academy—Pitch Draw). Families were expected to report any technical difficulties, so they could be rectified prior to Week 3.

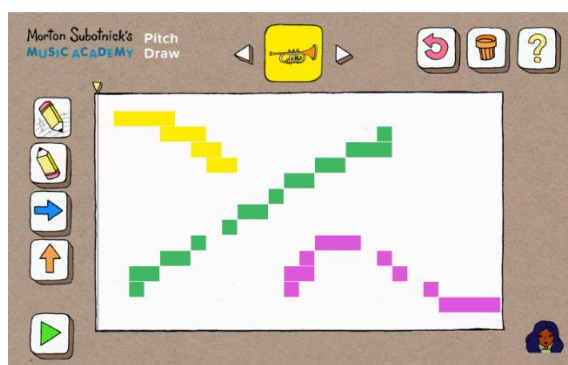


Figure 10. Morton Subotnick's Music Academy—Pitch Draw.

Week 3 — Music apps

Monday/Tuesday

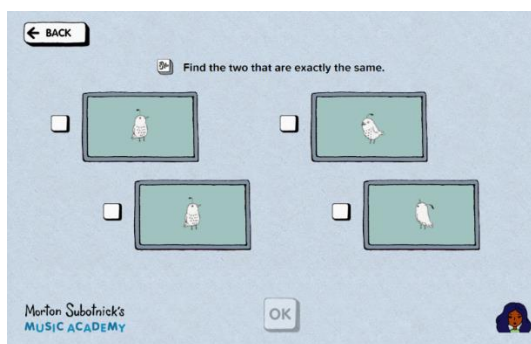
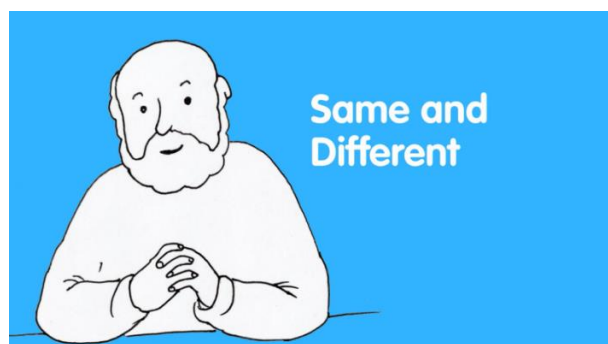


Figure 11. Morton Subotnick's Music Academy—Same and Different Video (L) and Practice Two the Same (R).

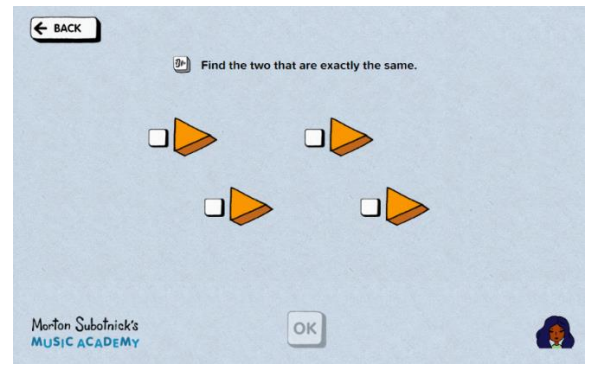
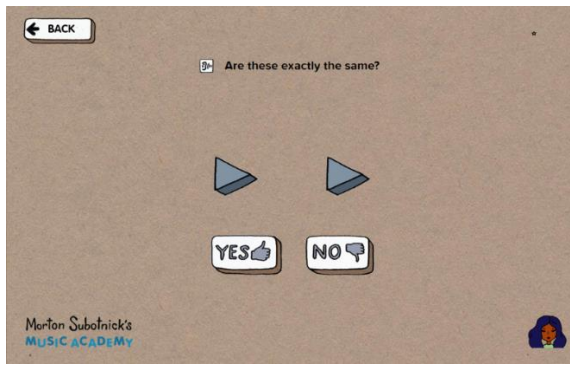


Figure 12. Morton Subotnick's Music Academy—Same and Different (L) and Two the Same (R).

Wednesday/Thursday

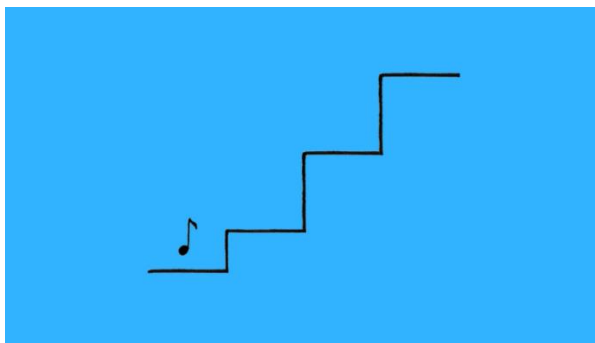


Figure 13. Morton Subotnick's Music Academy—Up and Down Video (L) and Going Up and Down (R).

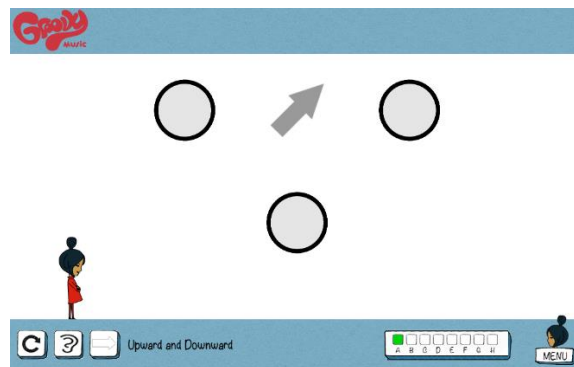


Figure 14. Groovy Music—Upward and Downward.
[click the shapes to find the melody that goes upward, drag the arrow to the melody that goes upward]

Week 4 — Music apps

Monday/Tuesday

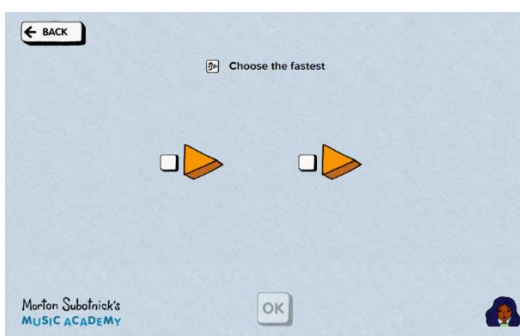


Figure 15. Morton Subotnick's Music Academy—Timing Fast and Slow Video and Practice Fastest or Slowest.

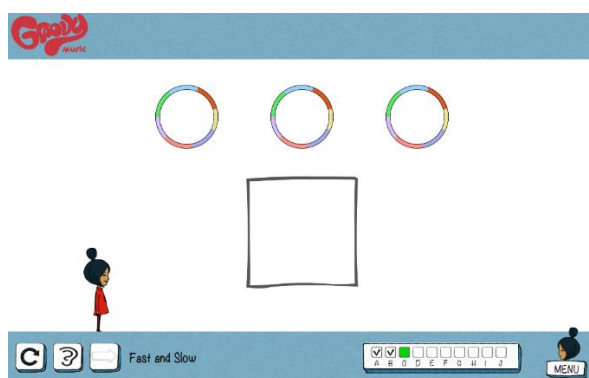


Figure 16. Groovy Music—Fast and Slow.
[Find the fast/slow music and drag it to the box]

Wednesday/Thursday

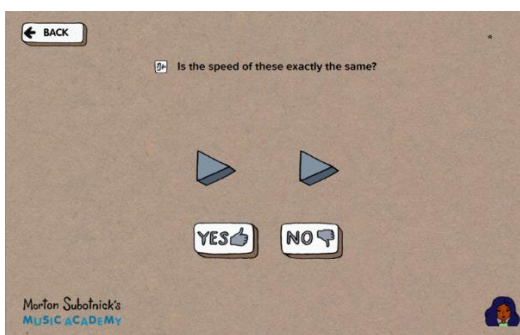
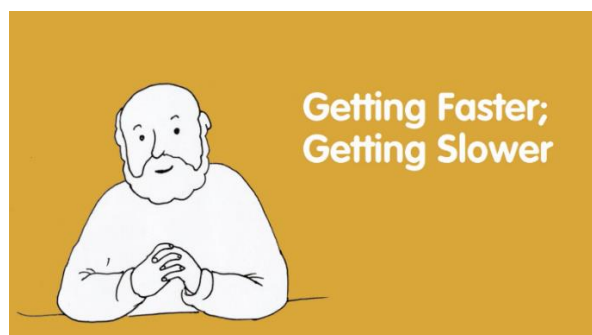


Figure 17. Morton Subotnick's Music Academy—Getting Faster; Getting Slower Video (L) and Same or Different Speed (R).

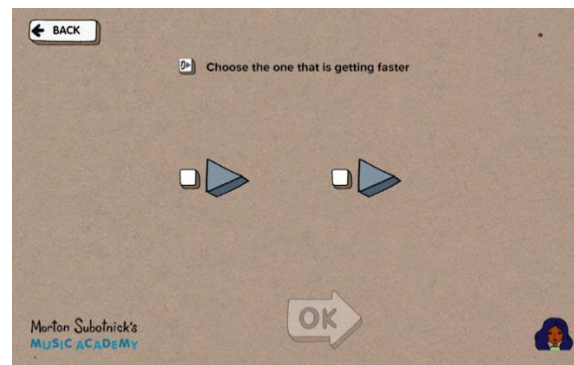
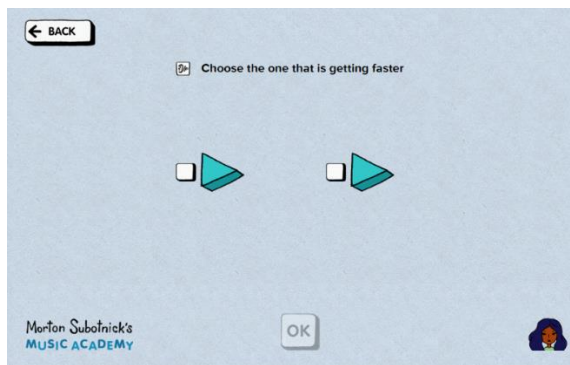


Figure 18. Morton Subotnick's Music Academy—Practice Getting Faster and Slower (L) and Getting Faster or Slower.

Week 5 — Music apps

Monday/Tuesday

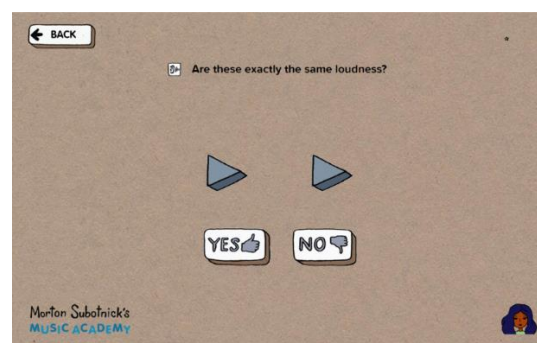
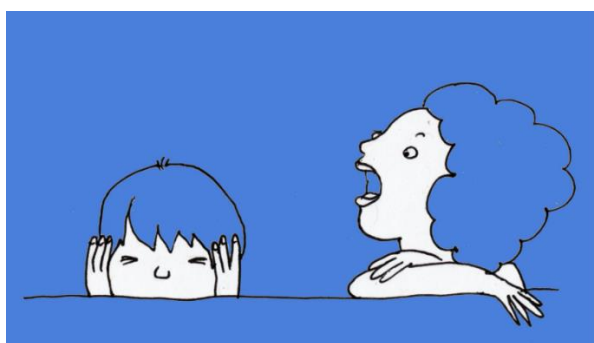


Figure 19. Morton Subotnick's Music Academy—About Loud and Soft Video (L) and Same or Different Loudness (R).

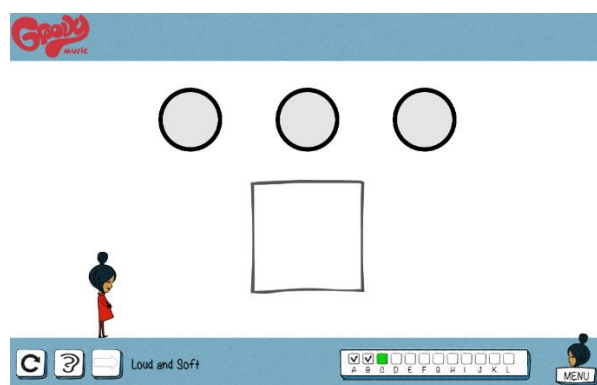


Figure 20. Groovy Music—Loud and Soft.
[Find the loud/soft music and drag it to the box]

Wednesday/Thursday



Figure 21. Morton Subotnick's Music Academy—Individual Instruments Video (L) and Same or Different Musical Instruments (R).

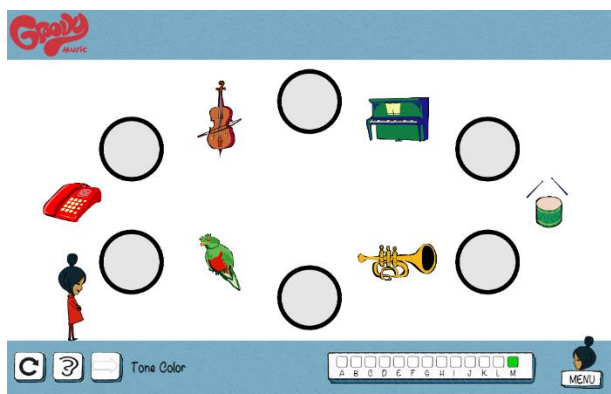


Figure 22. Groovy Music—Tone Color.
[Click on each shape to listen to their sound. Drag each shape to the matching picture]

Week 6 — Music apps

Monday/Tuesday

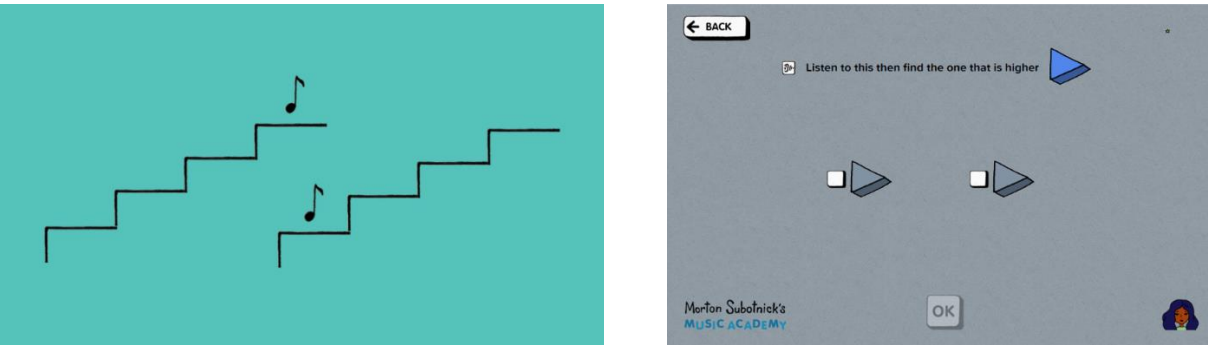


Figure 23. Morton Subotnick's Music Academy—Higher and Lower Video (L) and Higher or Lower (R).

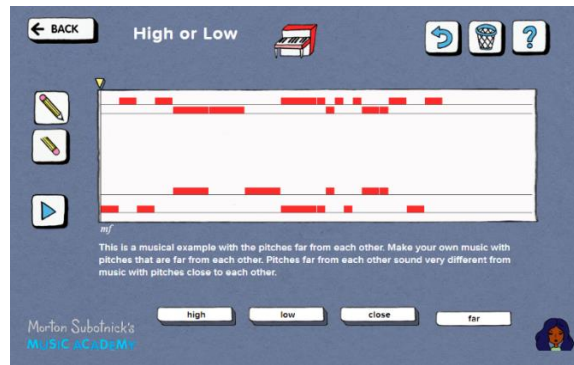


Figure 24. Morton Subotnick's Music Academy—High or Low.

Wednesday/Thursday

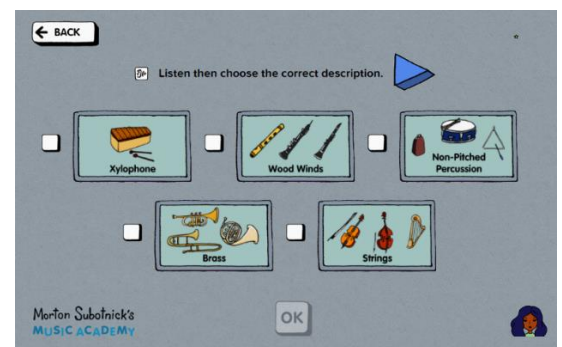


Figure 25. Morton Subotnick's Music Academy—Instrument Families Video (L) and Instrument Families (R).

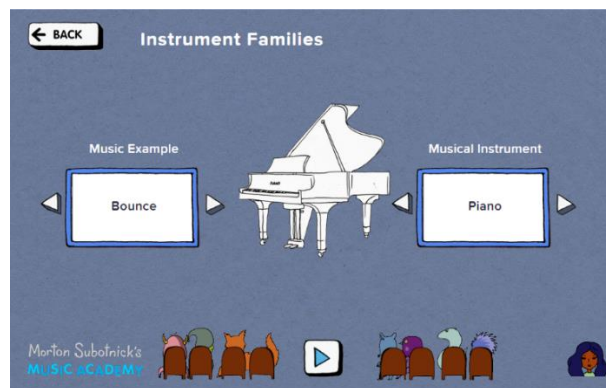


Figure 26. Morton Subotnick's Music Academy—Instrument Families (Creative Task).

Week 7 — Music apps

Monday/Tuesday



Figure 27. Morton Subotnick's Music Academy—The Families in the Orchestra Video (L) and Find the Section of the Orchestra (R).

Wednesday/Thursday

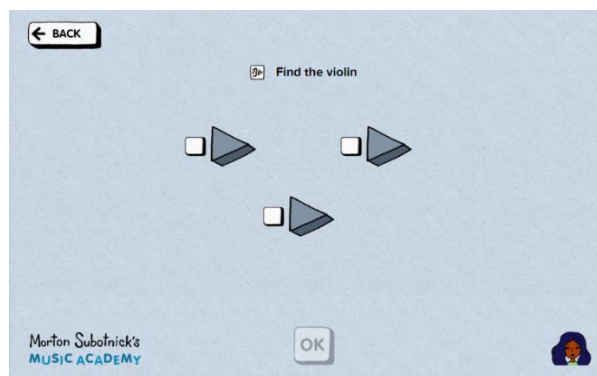


Figure 28. Morton Subotnick's Music Academy—Practice Find the Instrument (L) and Find the Instrument (R).

Week 8 — Music apps

Monday/Tuesday

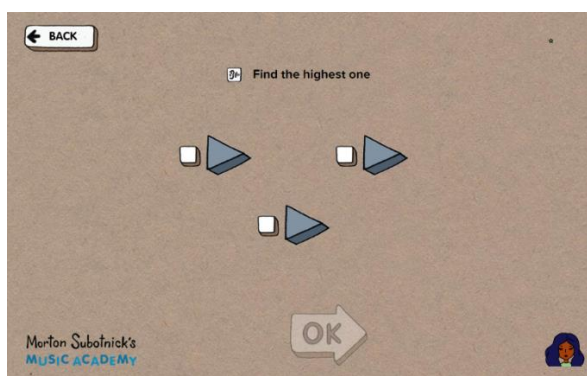
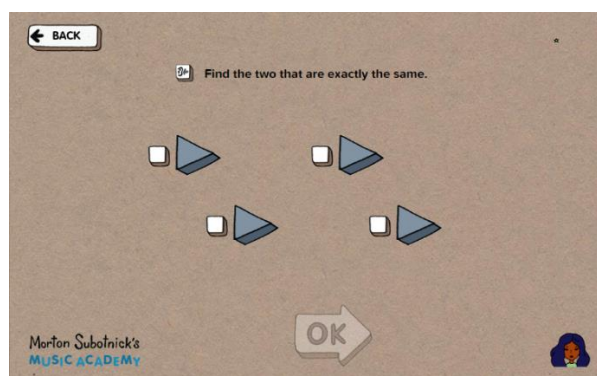


Figure 29. Morton Subotnick's Music Academy—Two the Same (L) and Find the Highest/Lowest (R).

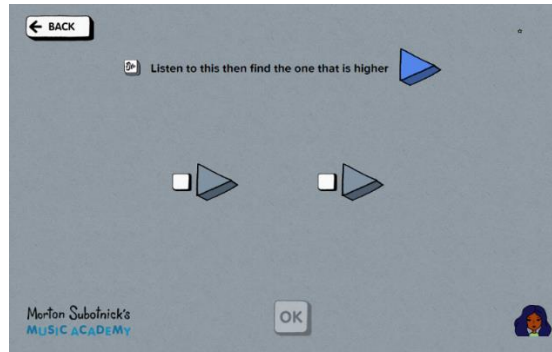


Figure 30. Morton Subotnick's Music Academy—Listen and Find the Higher/Lower.

Wednesday/Thursday

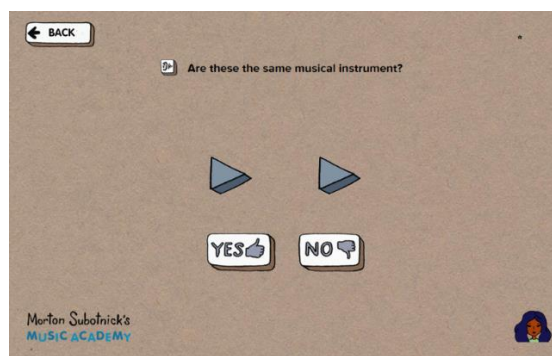


Figure 31. Morton Subotnick's Music Academy—Same or Different (Instruments).

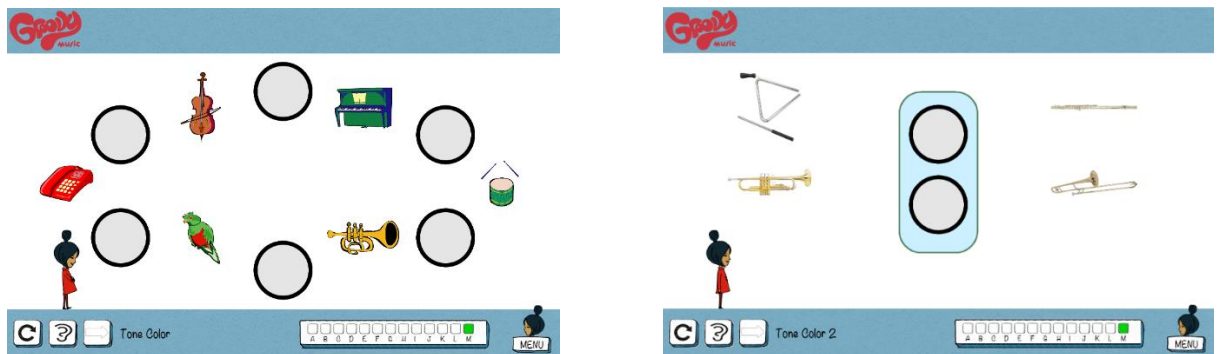


Figure 32. Groovy Music—Tone Color 1 (L) and 2 (R).
[Click on each shape to listen to their sound. Drag each shape to the matching picture]

Week 9, 10, 11, and 12

For weeks 9–12, all of the apps from weeks 3–8 were made available for revision.

Creative activities

From week 3–8, the following “creative” apps were available on Friday, Saturday and Sunday. From weeks 9–12 (the revision period), the following apps were made available each day. An audiobook of “Peter and the Wolf”—a symphonic fairy tale for children was also available at these weeks.

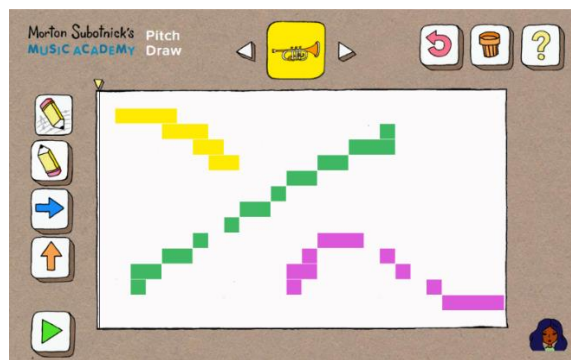


Figure 33. Morton Subotnick's Music Academy—Pitch Draw.

[Notes are drawn with pitch frequency on the y-axis, and time on the x-axis. Multiple instruments are available, denoted by different colours.]

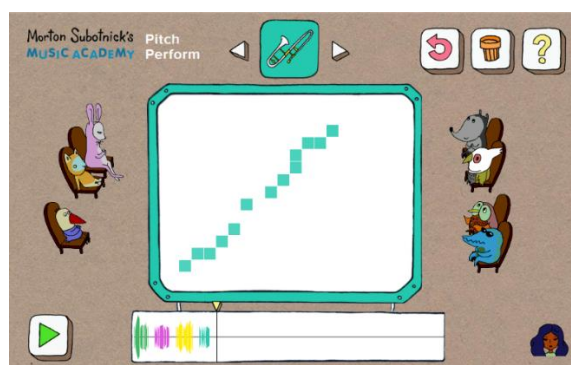


Figure 34. Morton Subotnick's Music Academy—Pitch Perform.

[Similar to Pitch Draw, but multiple segments are drawn and connected to form a larger composition]

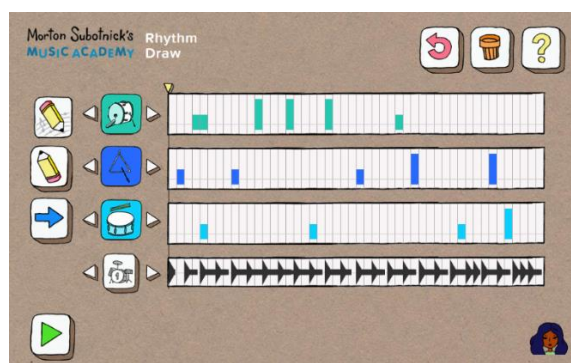


Figure 35. Morton Subotnick's Music Academy—Rhythm Draw.

[A basic beat is selected (bottom of the figure), and multiple percussive instruments' rhythms can be drawn at varying intensity]

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Table 11. Independent samples *t*-test of outcome measures and age between children with SNHL and TH.

		Levene's Test for Equality of Variances		<i>t</i> -test for Equality of Means						
		F	<i>p</i>	<i>t</i>	df	<i>p</i>	Mean Difference	SE Difference	95% CI of the Difference	
									Lower	Upper
SIN	Equal variances assumed	18.86	.000	-5.23	25	.000	3.8	.7	-5.3	-2.3
	Equal variances not assumed			-4.55	12	.001*	3.8	.8	-5.6	-2.0
Pitch	Equal variances assumed	0.04	.836	-1.80	25	.083	2.1	1.1	-4.4	0.3
	Equal variances not assumed			-1.78	21	.090	2.1	1.2	-4.5	0.4
Timbre	Equal variances assumed	8.30	.008	4.95	25	.000	-30.7	6.2	17.9	43.4
	Equal variances not assumed			5.56	23	.000*	-30.7	5.5	19.3	42.1
Spectral Resolution	Equal variances assumed	3.69	.066	6.66	25	.000*	-4.5	.7	3.1	5.8
	Equal variances not assumed			7.28	25	.000	-4.5	.6	3.2	5.7
Emotional Prosody	Equal variances assumed	18.81	.000	3.37	25	.002	-13.1	3.9	5.1	21.1
	Equal variances not assumed			2.95	13	.012*	-13.1	4.4	3.5	22.7
Question/ Statement Prosody	Equal variances assumed	2.26	.145	1.32	25	.197	-10.2	7.7	-5.7	26.1
	Equal variances not assumed			1.42	25	.168	-10.2	7.2	-4.6	25.0
Age	Equal variances assumed	1.23	.278	0.86	25	.400	-0.3	.3	-0.4	1.0
	Equal variances not assumed			0.83	20	.414	-0.3	.4	-0.4	1.0

**p* ≤ .05

Table 12. Estimates of fixed effects for speech-in-noise at double baselines for children with SNHL.

Parameter	Estimate	SE	df	t	Sig.
Intercept	6.03	3.96	8	1.52	.165
Time (Baseline 1)	-0.26	0.49	12	-0.53	.605
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	0.90	1.30	9	0.70	.502
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	0.79	0.77	12	1.02	.330
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	1.63	1.96	9	0.83	.427
Device (Bimodal)	1.04	2.13	9	0.49	.636
Device (HA)	0 ^b	0.00			
Hearing age	-0.66	0.47	8	-1.40	.200
Formal music experience	0.03	0.35	8	0.09	.934

b. This parameter is set to zero because it is redundant.

Table 13. Estimates of fixed effects for spectral resolution at double-baselines for children with SNHL.

Parameter	Estimate	SE	df	t	p
Intercept	6.81	2.10	10	3.24	.008*
Time (Baseline 1)	-0.42	0.25	11	-1.67	.123
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	0.91	0.60	14	1.53	.149
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	0.02	0.38	11	0.05	.964
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	-2.42	0.90	8	-2.68	.029*
Device (Bimodal)	-2.49	0.93	7	-2.69	.031*
Device (HA)	0 ^b	0.00			
Hearing age	-0.38	0.26	11	-1.45	.174
Formal music experience	0.20	0.15	7	1.28	.241

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 14. Estimates of fixed effects for emotional prosody at double-baselines for children with SNHL.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	28.82	14.45	8	1.99	.081
Time (Baseline 1)	-6.68	2.43	13	-2.75	.017*
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	-1.85	4.27	8	-0.43	.676
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	3.56	3.68	12	0.97	.352
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	-5.75	6.89	8	-0.83	.428
Device (Bimodal)	7.62	7.54	8	1.01	.342
Device (HA)	0 ^b	0.00			
Hearing age	8.42	1.74	8	4.83	.001*
Formal music experience	0.11	1.30	8	0.08	.935

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 15. Estimates of fixed effects for question/statement prosody at double-baselines for children with SNHL.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	14.67	25.51	8	.58	.580
Time (Baseline 1)	-9.73	7.04	12	-1.38	.192
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	10.46	8.80	8	1.19	.267
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	9.82	10.72	12	.92	.377
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	6.40	12.07	8	.53	.610
Device (Bimodal)	-4.18	13.20	8	-.32	.760
Device (HA)	0 ^b	0.00			
Hearing age	7.12	3.06	8	2.33	.048*
Formal music experience	3.16	2.27	8	1.39	.202

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 16. Estimates of fixed effects for pitch at double-baselines for children with SNHL.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	15.50	3.00	9	5.17	.001
Time (Baseline 1)	0.65	0.63	12	1.03	.323
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	1.15	1.25	10	0.92	.382
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	0.14	0.96	12	0.14	.889
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	2.18	1.40	8	1.56	.158
Device (Bimodal)	-0.51	1.53	8	-0.33	.747
Device (HA)	0 ^b	0.00			
Hearing age	-1.69	0.36	8	-4.75	.001*
Formal music experience	-0.59	0.26	8	-2.22	.057

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 17. Estimates of fixed effects for timbre at double-baselines for children with SNHL.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	-42.02	16.94	9	-2.48	.036
Time (Baseline 1)	4.60	3.81	12	1.21	.249
Time (Baseline 2)	0 ^b	0.00			
Group 1 (1-week retest)	11.33	6.80	7	1.67	.138
Group 2 (12-weeks retest)	0 ^b	0.00			
Baseline 1 * 1-week retest	-2.78	5.78	12	-0.48	.639
Baseline 1 * 12-weeks retest	0 ^b	0.00			
Baseline 2 * 1-week retest	0 ^b	0.00			
Baseline 2 * 12-weeks retest	0 ^b	0.00			
Device (CI)	1.51	7.93	8	0.19	.854
Device (Bimodal)	11.29	8.66	8	1.30	.229
Device (HA)	0 ^b	0.00			
Hearing age	8.90	2.02	8	4.41	.002*
Formal music experience	-0.69	1.50	8	-0.46	.656

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

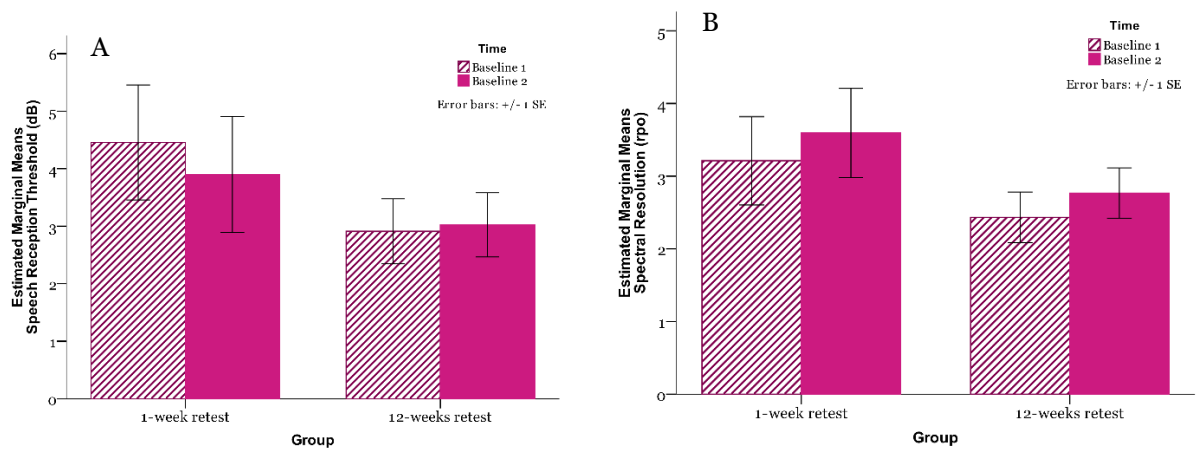


Figure 36. Bar graph of estimated marginal means for speech reception threshold (L) and spectral resolution (R) at double baselines for children with SNHL in each group.

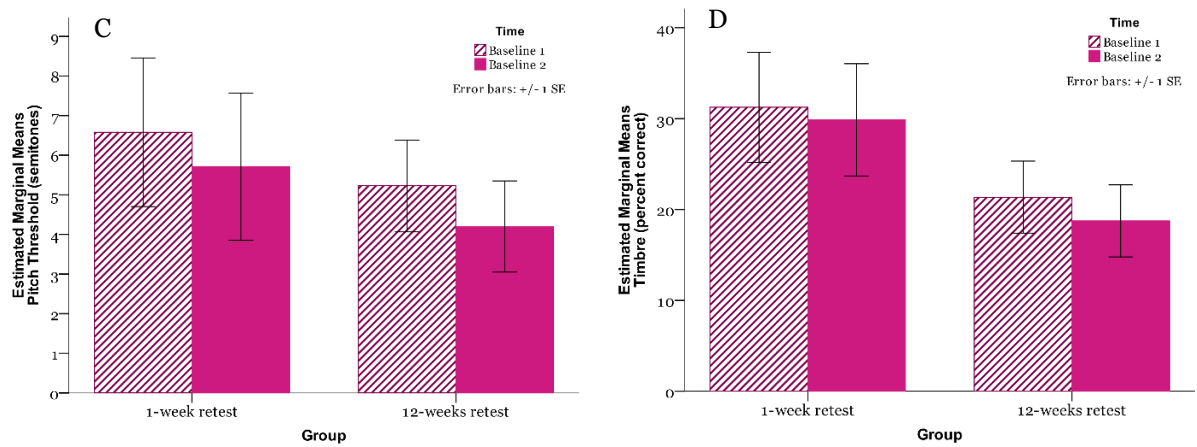


Figure 37. Bar graph of estimated marginal means for pitch threshold (L) and timbre (R) at double baselines for children with SNHL in each group.

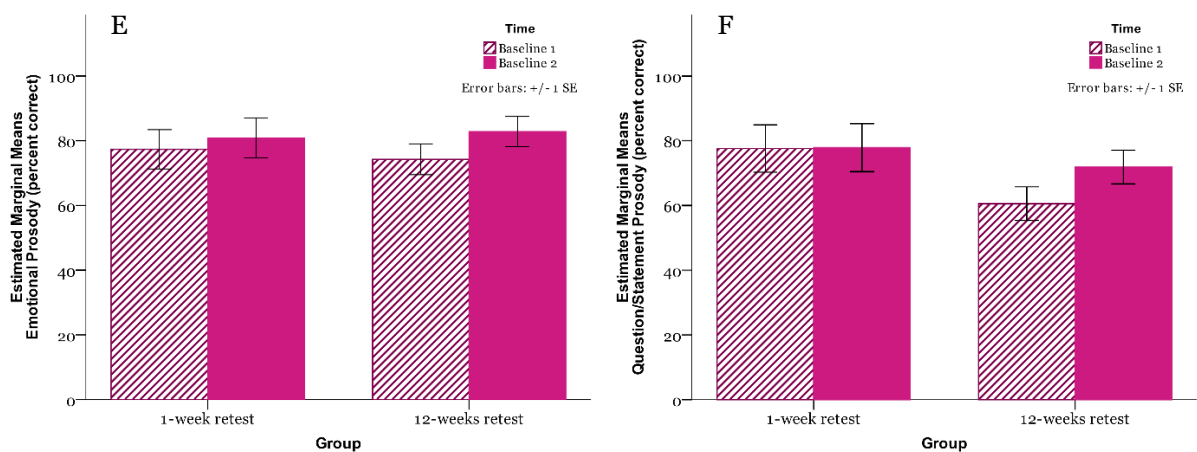


Figure 38. Bar graph of estimated marginal means for emotional prosody (L) and question/statement prosody at double baselines for children with SNHL in each group.

Table 18. Estimates of fixed effects for speech-in-noise at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	3.56	1.80	27	1.98	.058
Time (Baseline 1)	-0.06	0.38	28	-0.16	.875
Time (Baseline 2)	0 ^b	0.00			
Age	-0.49	0.24	27	-2.03	.052
Formal music experience	0.02	0.07	27	0.28	.785

b. This parameter is set to zero because it is redundant.

Table 19. Estimates of fixed effects for pitch at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	14.66	4.04	28	3.63	.001*
Time (Baseline 1)	-0.60	0.85	28	-0.70	.489
Time (Baseline 2)	0 ^b	0.00			
Age	-1.50	0.54	28	-2.78	.010*
Formal music experience	-0.28	0.15	28	-1.83	.078

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 20. Estimates of fixed effects for timbre at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	14.16	30.49	25	.46	.646
Time (Baseline 1)	-5.34	6.75	26	-.79	.436
Time (Baseline 2)	0 ^b	0.00			
Age	3.80	4.08	25	.93	.360
Formal music experience	3.95	1.14	25	3.46	.002*

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 21. Estimates of fixed effects for spectral resolution at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	-.40	2.81	27	-.14	.889
Time (Baseline 1)	-.19	0.61	27	-.32	.752
Time (Baseline 2)	0 ^b	0.00			
Age	1.09	0.37	26	2.93	.007*
Formal music experience	-.01	0.10	26	-.12	.902

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 22. Estimates of fixed effects for emotional prosody at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	64.74	12.38	19	5.231	.000*
Time (Baseline 1)	-4.22	3.37	21	-1.251	.225
Time (Baseline 2)	0 ^b	0.00			
Age	3.66	1.66	19	2.210	.040*
Formal music experience	0.65	0.46	19	1.398	.178

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

Table 23. Estimates of fixed effects for question/statement prosody at double baselines for TH children.

Parameter	Estimate	SE	df	<i>t</i>	<i>p</i>
Intercept	18.23	28.96	27	.63	.534
Time (Baseline 1)	-0.19	6.21	27	-.03	.976
Time (Baseline 2)	0 ^b	0.00			
Age	7.44	3.85	27	1.93	.064
Formal music experience	2.55	1.08	27	2.36	.026*

* $p \leq .05$

b. This parameter is set to zero because it is redundant.

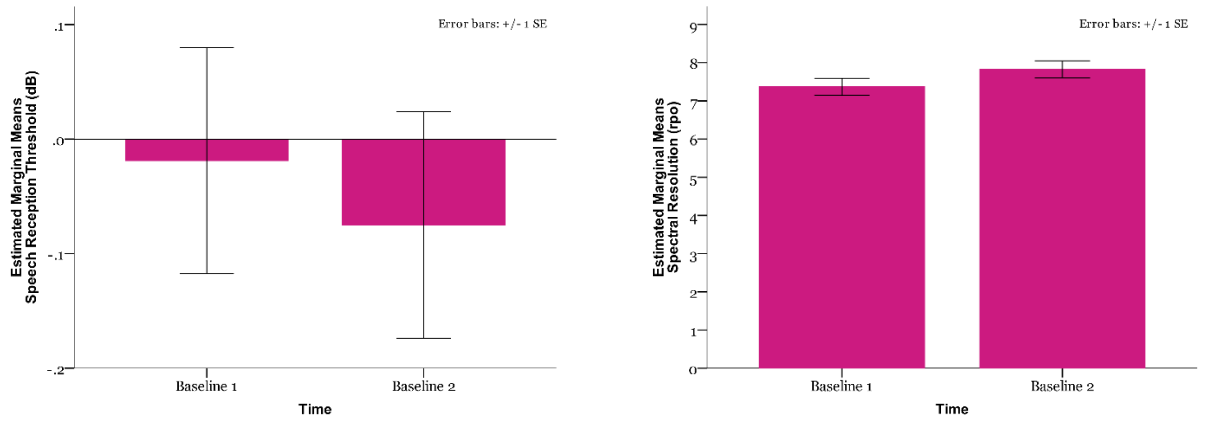


Figure 39. Bar graph of estimated marginal means for speech reception threshold (L) and spectral resolution (R) at double baselines for TH children.

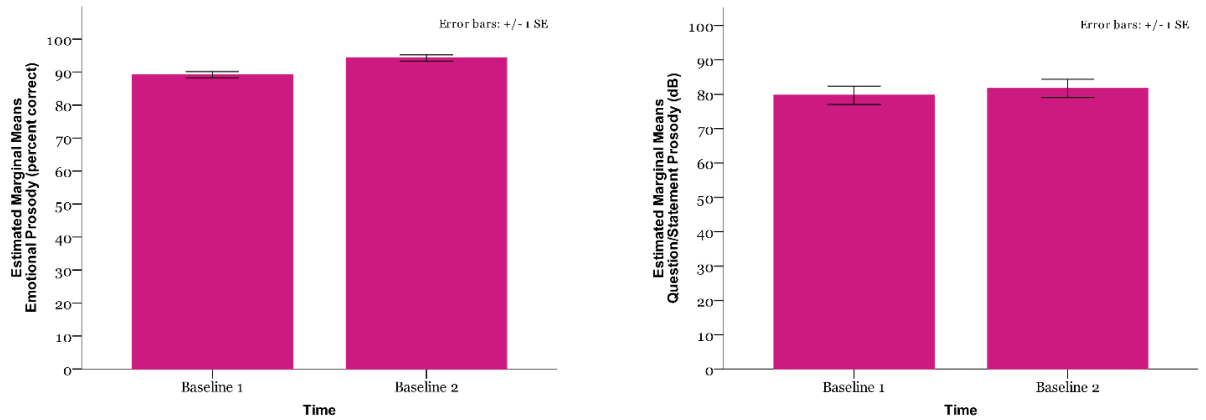


Figure 40. Bar graph of estimated marginal means for emotional prosody (L) and question/statement prosody (R) at double baselines for TH children.

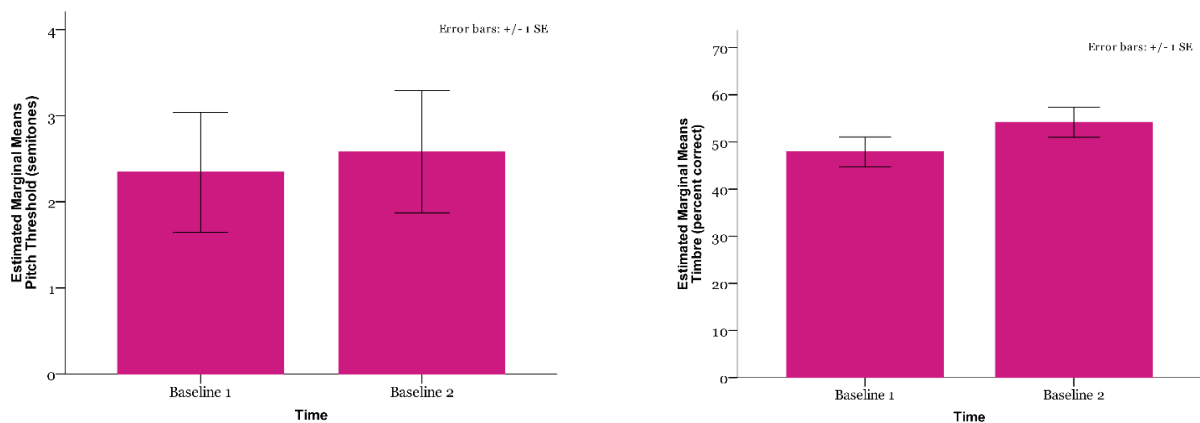


Figure 41. Bar graph of estimated marginal means for pitch threshold (L) and timbre (R) at double baselines for TH children.

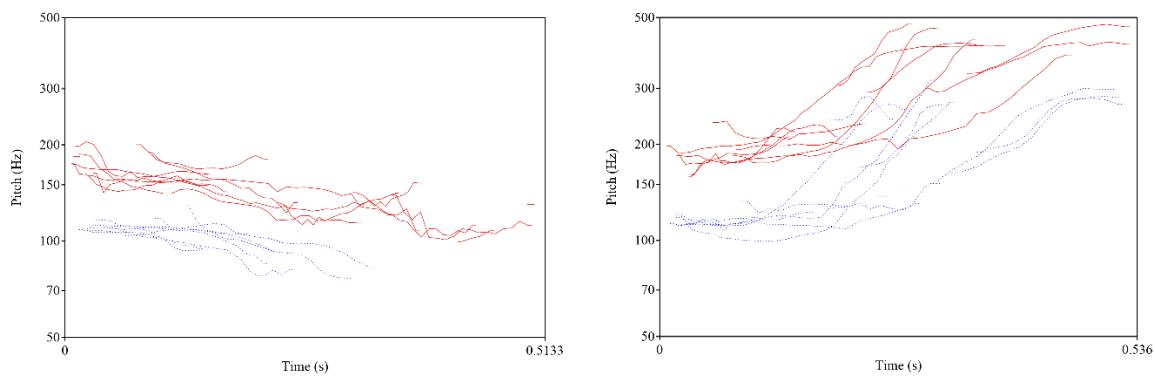


Figure 42. Intonation curves for the Question/Statement prosody [statement utterances (L) and question utterances (R), male (blue) and female (red)]

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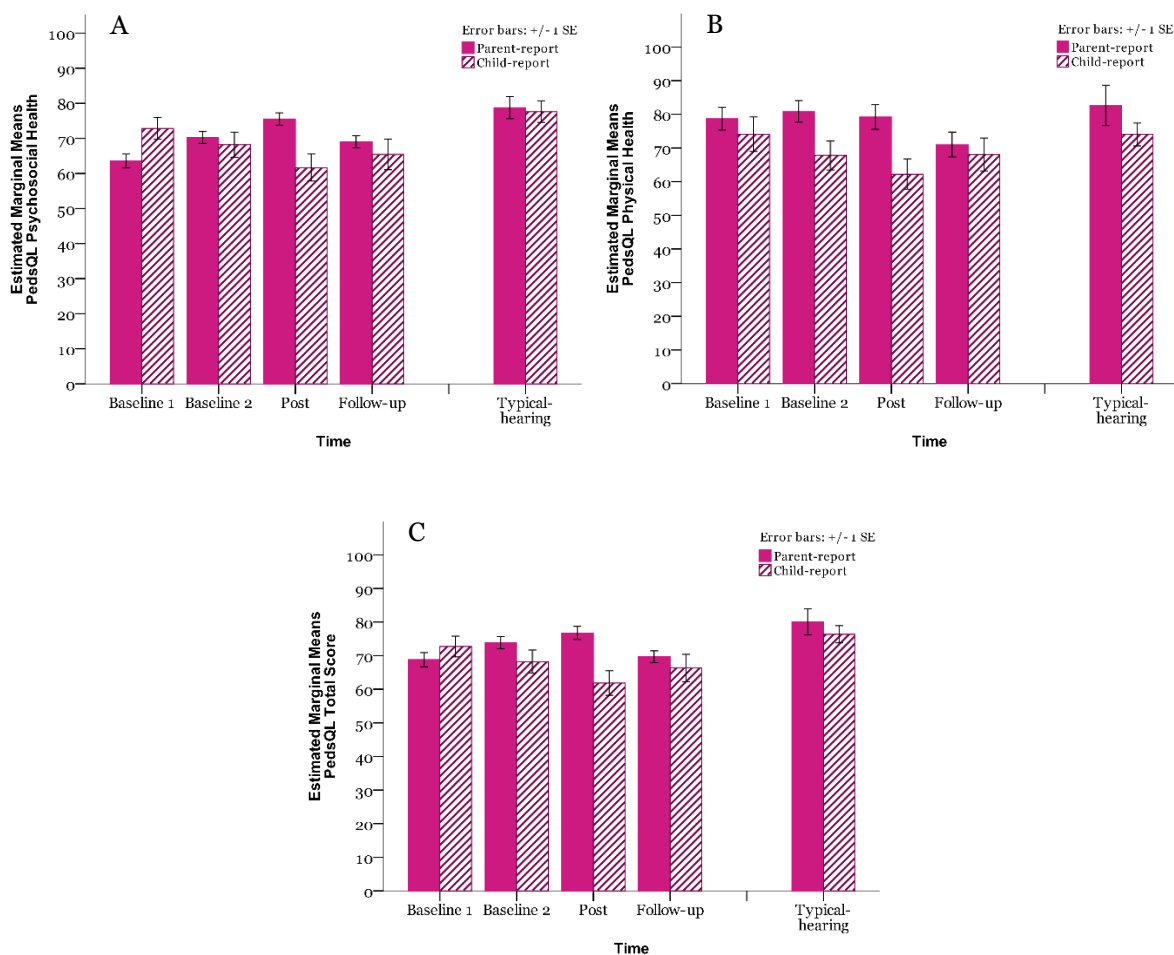


Figure 43. Bar graphs of estimated marginal means for PedsQL GCS across time with a comparison of TH children's performance: (A) Psychosocial Health, (B) Physical Health, (C) Total score.

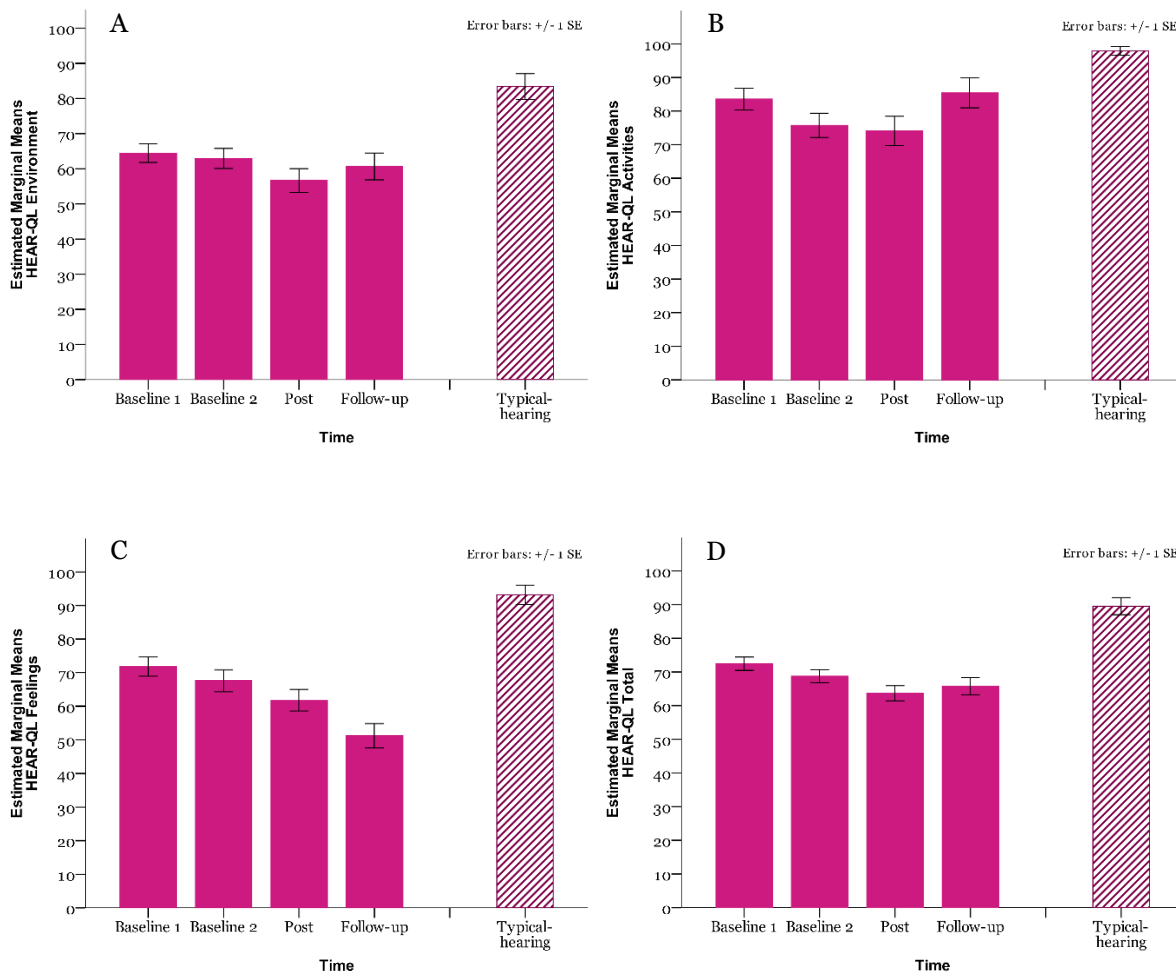


Figure 44. Bar graphs of estimated marginal means for HEAR-QL-26 domains across time with a comparison of TH children's performance: (A) Environment, (B) Activities, (C) Feelings, (D) Total.

Table 24. Independent samples *t*-test of reported measures between children with SNHL and their parents.

		Levene's Test for Equality of Variances		t-test for Equality of Means						
		F	p	t	df	p	Mean Difference	SE Difference	95% CI of the Difference	
									Lower	Upper
SDQ Internalising Problems (Parent report)	Equal variances assumed	.30	.592	3.21	26	.003*	3.0	.9	1.1	4.9
	Equal variances not assumed			3.10	20	.006	3.0	1.0	1.0	5.0
SDQ Externalising Problems (Parent report)	Equal variances assumed	.09	.769	0.70	26	.488	1.2	1.7	-2.3	4.7
	Equal variances not assumed			0.73	26	.475	1.2	1.6	-2.2	4.6
SDQ Prosocial (Parent report)	Equal variances assumed	.06	.815	-0.85	26	.402	-0.6	.7	-2.1	0.9
	Equal variances not assumed			-0.84	22	.411	-0.6	.7	-2.2	0.9
SDQ Total (Parent report)	Equal variances assumed	.27	.605	1.79	26	.085	4.2	2.3	-0.6	9.0
	Equal variances not assumed			1.81	24	.083	4.2	2.3	-0.6	9.0
PedsQL Psychosocial Health (Parent report)	Equal variances assumed	.39	.535	-1.46	26	.158	-7.8	5.3	-18.8	3.2
	Equal variances not assumed			-1.41	21	.174	-7.8	5.5	-19.3	3.7
PedsQL Psychosocial Health (Child report)	Equal variances assumed	.42	.521	-1.49	24	.150	-7.9	5.3	-18.9	3.1
	Equal variances not assumed			-1.42	16	.174	-7.9	5.6	-19.7	3.9
PedsQL Physical Health (Parent report)	Equal variances assumed	1.24	.275	-0.12	26	.907	-1.0	8.2	-17.7	15.8
	Equal variances not assumed			-0.12	26	.902	-1.0	7.7	-16.9	15.0
PedsQL Physical Health (Child report)	Equal variances assumed	1.59	.219	-0.92	24	.365	-5.6	6.1	-18.1	6.9
	Equal variances not assumed			-0.88	16	.392	-5.6	6.4	-19.1	7.9
PedsQL Total (Parent report)	Equal variances assumed	.01	.923	-0.92	26	.368	-5.4	5.9	-17.5	6.7
	Equal variances not assumed			-0.92	24	.365	-5.4	5.9	-17.5	6.7
PedsQL Total (Child report)	Equal variances assumed	2.81	.107	-1.43	24	.166	-7.1	5.0	-17.4	3.2
	Equal variances not assumed			-1.31	14	.212	-7.1	5.4	-18.7	4.5
HEAR-QL Environment (Child report)	Equal variances assumed	.10	.753	-3.68	24	.001*	-21.2	5.8	-33.1	-9.3
	Equal variances not assumed			-3.75	21	.001	-21.2	5.6	-32.9	-9.4
HEAR-QL Activities (Child report)	Equal variances assumed	28.04	.000	-3.13	24	.005	-14.5	4.7	-24.1	-4.9
	Equal variances not assumed			-2.54	10	.030*	-14.5	5.7	-27.3	-1.8
HEAR-QL Feelings (Child report)	Equal variances assumed	6.83	.015	-3.81	24	.001	-25.7	6.7	-39.6	-11.8
	Equal variances not assumed			-3.29	12	.007*	-25.7	7.8	-42.7	-8.7
HEAR-QL Total (Child report)	Equal variances assumed	1.31	.263	-4.36	24	.000*	-20.4	4.7	-30.1	-10.8
	Equal variances not assumed			-4.07	15	.001	-20.4	5.0	-31.1	-9.7

* $p \leq .05$