Beyond Audition: Psychosocial Benefits of Music Training for Children With Hearing Loss

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Objectives: Children with hearing loss tend to have poorer psychosocial and quality of life outcomes than their typical-hearing (TH) peers particularly in the areas of peer relationships and school functioning. A small number of studies for TH children have suggested that groupbased music activities are beneficial for prosocial outcomes and help develop a sense of belonging. While one might question whether perceptual limitations would impede satisfactory participation in musical activities, findings from a few studies have suggested that group music activities may have similar benefits for children with hearing loss as well. It is important to note that the effect of music on psychosocial outcomes has primarily been investigated at an anecdotal level. The objective of this study was to explore the effect of a music training program on psychosocial and quality of life outcomes for children with hearing loss. It was hypothesized that music training would provide benefits for domains centered upon peer relationships and prosocial measures.

Design: Fourteen children aged 6 to 9 years with prelingual sensorineural hearing loss (SNHL) participated in a 12-week music training program that consisted of group-based face-to-face music therapy supplemented by online music apps. The design was a pseudorandomized, longitudinal study (9 participants were waitlisted, initially serving as a passive control group). Psychosocial wellbeing and quality of life were assessed using a questionnaire battery comprised of the Strengths and Difficulty Questionnaire (SDQ), the Pediatric Quality of Life Inventory, the Hearing Environments and Reflection on Quality of Life (HEAR-QL), and the Glasgow Children's Benefit Inventory. For comparative purposes, responses were measured from 16 TH children that ranged in age from 6 to 9 years.

Results: At baseline, children with SNHL had poorer outcomes for internalizing problems, and all measures of the HEAR-QL compared with the TH children. There were no differences for general psychosocial and physical health. After music training, SDQ internalizing problems such as peer relationships and emotional regulation were significantly reduced for the children with SNHL. There were no changes for any outcomes for the passive control group. Additional benefits were noted for emotional and learning factors on the Glasgow Children's Benefit Inventory. However, there were no significant changes for any psychosocial and quality of life outcomes as measured by the Pediatric Quality of Life Inventory or HEAR-QL instruments.

Conclusions: The present study provides initial evidence that music training has a positive effect on at least some psychosocial and quality of life outcomes for children with hearing loss. As they are at a greater risk of poorer psychosocial and quality of life outcomes, these findings are cause for cautious optimism. Children with hearing loss should be encouraged to participate in group-based musical activities.

Key words: Children, Cochlear implants, Deafness, Hearing aids, Music, Psychosocial, Quality of life.

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INTRODUCTION

The main goal of early intervention for children with hearing loss is the provision of audibility for the primary purpose of maximizing speech and language development (The Joint Committee on Infant Hearing 2019). Indeed, the majority of research investigating outcomes for children with hearing loss has been focused toward improving and understanding language and speech perception (Blamey et al. 2001; Schorr et al. 2008; Ching et al. 2017, 2018), 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 cognitive, educational, and behavioral problems (Stevenson et al. 2010; Hoffman et al. 2015). However, it is also important to consider that children's needs extend far beyond language.

Quality of Life and Psychosocial Outcomes

The World Health Organization Quality of Life Group (1998, p. 551) defines quality of life 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. This definition reflects the view that quality of life refers to a subjective evaluation that is embedded in a cultural, social, and environmental context." Within pediatric audiology, quality of life is often hypothesized as a resulting cascade of consequences that is directly influenced by the presence of prelingual deafness, subsequent intervention, and auditory and linguistic outcomes (Summerfield & Marshall 1999; Lin & Niparko 2006; Stacey et al. 2006).

There is high variability in quality of life outcomes for children with hearing loss. A meta-analysis (albeit of only 4 studies using the Pediatric Quality of Life Inventory [PedsQL] inventory for children aged between 6 and 18) by Roland et al. (2016) found statistically and clinically poorer outcomes on the domains of school and social functioning, but not physical and emotional domains, compared with typical-hearing (TH) peers.

Multiple studies have reported that children with hearing loss have a greater risk for poorer psychosocial outcomes than their TH peers (Moeller et al. 2007; Fellinger et al. 2008; Kant & Adhyaru 2009). A range of psychosocial problems are associated with hearing loss; overt behaviors such as aggression and problems around conduct are categorized as externalizing problems, while behaviors such as depression and anxiety are categorized as internalizing problems (American Psychiatric Association 2013; Theunissen et al. 2014). A systematic review by Stevenson et al. (2015) examined 33 studies utilizing quantitative questionnaires (stratified as either SDQ, or non-SDQ) that investigated emotional and behavioral difficulties. As a review article, the participants were wide-ranging, involving

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children and adolescents with mild to profound hearing loss aged between 6 and 21 years of age, with the majority appearing to be aided and/or implanted. The primary finding identified the area at most risk and concern was peer relationships.

Language and communication are well-established factors associated with psychosocial development in school-aged children, 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 (Polat 2003; Hoffman et al. 2015). This is likely due to the general behavior that from about 4 years of age, children tend to move away from dyadic interactions to larger groups, which generates higher levels of speechin-noise (SIN) (Benenson et al. 1997). Thus, children with better speech intelligibility are likely to communicate more effectively with their peers in group-based interactions (Punch & Hyde 2011).

According to a literature review by Xie et al. (2014) investigating peer interactions for children with hearing loss in inclusive settings, only one study has explored the efficacy of a social skills training program. This study by 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 twenty 1-hr sessions twice a week that dealt with cognitive and interpersonal problem solving, followed by six 1-hr social skills programs that was taught in conjunction with TH peers. However, the total duration of the study was not clearly reported. Psychosocial factors such as emotional and social adjustment, as well as self-image were improved at the end of the program. More than half of the children showed improvements of assertive behavior, inhibition, and thinking. These findings were supported by the children's teachers, along with self-reports.

The Rationale for Music as (Re)habilitation

In recent years, a small body of studies has suggested musicbased activities, in addition to auditory-verbal and social skills training, as a component of early intervention programs for children with hearing loss (for review, see Torppa & Huotilainen 2019). Given cochlear implants (CIs) are known to convey a degraded representation of pitch and timbre (Looi et al. 2012), what is the basis for considering music-based interventions as part of psychosocial (re)habilitation?

Music is ubiquitous. As a universal behavior or phenomena, music is a multisensory activity that typically involves the broad activation of auditory, visual, cognitive, and motor domains (Zimmerman & Lahav 2012). It is unsurprising then, that the nature and function of music is sufficiently broad and diverse. Nonetheless, a large-scale survey (n = 834) by Schäfer et al. (2013) categorized the psychological functions of music listening into three distinct, underlying dimensions: to regulate arousal and mood, to achieve self-awareness, and to express social relatedness.

From the perspective of group music making, survey data from 78 university-level music students reported 3 major themes of group music making as: a musical act, as a social act, and as personal skill development (Kokotsaki & Hallam 2007). As Hargreaves et al. (2003, p. 160) 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." Rhythm has also been implicated as a key link between music and social bonding, on the basis that synchronizing with another person may remove barriers as they work toward a shared musical experience (Overy 2012). Fundamentally, music is also fun, pleasurable, and rewarding (Blood & Zatorre 2001).

Taken together, music has many functions related to psychosocial wellbeing. Music provides a comprehensive, multisensory experience that leverages synchronicity to positive effect, and reward pathways for sustained learning, which is particularly relevant for children and adherence to training. Thus, music training provides a unique mechanism to explore psychosocial wellbeing and quality of life outcomes as part of an intervention program.

Evidence from longitudinal studies of TH children has vielded positive outcomes. Rickard et al. (2013) conducted a 2.5-year study across 9 schools with a total of 359 children from grades 1 to 3. Five schools received an age-specific music intervention program (children in grade 1 at baseline received 3×30 minutes Kodály classes a week, while children in grade 3 at baseline received a 1-hr, group string-instrumental class a week). The Kodály curriculum is based on principles of childhood development, with a large focus on singing and the voice (Kodály 1974). The control group consisted of the remaining four schools that continued with their standard school curriculum and music program. Hence, the five schools receiving the music intervention were essentially receiving greater exposure to music training. While schools which received the music intervention program showed no significant benefit to social skills (potentially because they were above average at baseline), significant benefits were noted for self-esteem.

Another longitudinal study with TH children by Williams et al. (2015) explored the contributions of early book reading and music activities between parent and child in the home environment. Data were collected from 3031 children when they were 2 to 3 years, and again at 4 to 5 years. A large number of outcomes were investigated such as vocabulary, numeracy, school readiness, attentional and emotional regulation, and prosocial skills. It is interesting that shared music activities (and not shared book reading) were associated with better prosocial outcomes. Williams et al. 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 a multimodal activity may make it more accessible and interactive than shared reading.

The psychosocial benefits of music training are also apparent in older TH children. A study by Schellenberg et al. (2015) investigated a music program with weekly 40-min classes over 10 mo for 8- to 9-year-old children (n = 38) in comparison to a control group that did not receive music training (n = 46). The music program focused on the use of the ukulele and students were encouraged to share their knowledge and skills, actively encouraging cooperative behavior. There was some evidence of benefit, but prosocial skills only improved from baseline for children that were already poor performers. For a review of music interventions and TH child development across a range of domains, see Hallam (2010) and Dumont et al. (2017).

A modest body of studies have recommended the use of music training for children with hearing loss as a complementary intervention (Chen et al. 2010; Torppa & Huotilainen 2019; Lo et al. 2020). The relatively limited research is likely due in part to the challenging logistical considerations of designing and facilitating a longitudinal training study for a hearing loss population (Gfeller 2016). Recent studies for children with hearing loss indicate preliminary evidence of positive associations between music, speech, and language outcomes such as singing, prosodic stress and reading (Torppa et al. 2020), melodic contour identification and Mandarin lexical tone perception (Cheng et al. 2018), piano instruction and prosody perception (Good et al. 2017). However, it should be noted that a recent systematic review on the benefits of music training for individuals with hearing loss found the evidence to support speech perception benefits is not yet sufficiently robust, or demonstrated convincingly (McKay 2021).

In contrast, the psychosocial and quality of life benefits from music-based interventions are mostly unexplored for children with hearing loss. It is often assumed that because children with hearing loss face difficulties with music perception (Roy et al. 2014; Lo et al. 2020), music is not a suitable activity. However, perceptual skills generally have little bearing on music appreciation and enjoyment (Looi et al. 2012). Furthermore, family attitudes and support appear to have a greater impact on music engagement than perceptual factors (Gfeller et al. 2019; Looi et al. 2019).

A 2-year pilot study by Yucel et al. (2009) explored a music training program for 18 (9 active, 9 control) pediatric unilateral CI recipients and bimodal users that focused on the use of a take-home electronic keyboard. Unfortunately, the ages were not clearly reported. Activities were centered on parents playing prescribed intervals and songs, and parents were encouraged to dance and play finger games. After training, statistically significant benefits were shown for a wide range of musical skills, 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 min of musical activities centered around play for 11 children with hearing loss aged between 9 and 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 socialization with peers, and a sense of belonging.

Summary and Study Goals

In summary, children with hearing loss face a range of psychosocial and quality of life challenges, with interpersonal relationships with peers implicated as the most salient issue. Findings from studies exploring music for TH children are mixed but generally positive, suggesting benefits for psychosocial skills such as prosocial behaviors and cooperation. Despite perceptual difficulties, children with hearing loss readily engage with music activities, but the skills, benefits, and outcomes remain relatively nascent and would benefit from further exploration.

The purpose of the present study was to explore the effect of a 12-week music training program on psychosocial and quality of life outcomes for children with hearing loss. To the authors' best knowledge, no study has explored psychosocial and quality of life outcomes with standardized generic and specific questionnaires. The present study forms the second part of a larger project that investigated the benefits of music training for children with hearing loss. The first study focused on perceptual outcomes, and significant benefits were noted relating to SIN, spectral resolution, timbre perception, as well as music appreciation (Lo et al. 2020). Based on these findings that suggested a general improvement for skills relevant for aural communication, it was hypothesized that music training would result in better outcomes for domains in which peer relationships and prosocial measures were 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 Hearing Environments and Reflection on Quality of Life (HEAR-QL-26) that were directly related to hearing-specific problems.

MATERIALS AND METHODS

Participants

Two groups of participants were tested in the study, stratified by hearing status (children with sensorineural hearing loss [SNHL] and TH). The SNHL 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 to 9.2 years (M = 7.5, SD = 1.1) when measured at the commencement of music training. Inclusion criteria for children with SNHL included prelingual onset of bilateral SNHL with moderate-to-profound thresholds, aided or implanted before the age of 3.5 years. Most children with SNHL (9/14) were enrolled in mainstream school settings, while the others attended schools for the deaf and hard-of-hearing with specialist support.

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. Relevant demographic data for children with SNHL can be found in Table 1.

The second group were 16 TH children (7 female, 9 male) that ranged in age from 6.3 to 8.7 years (M = 7.6, SD = 0.8) for comparative purposes (this group did not participate in music training and completed all the questionnaires to provide normative data for TH children). There was no significant difference between children with SNHL and TH for chronological age, t(25) = 0.86, p = 0.400, and formal music training, t(25) = -0.58, p = 0.569. At the start of the testing session, the TH children underwent pure-tone audiometric testing to confirm hearing thresholds (0.25 to $8 \text{ kHz} \le 20 \text{ dB HL}$). Relevant demographic data for TH children can be found in Table 2. All participants were native Australian English speakers. Exclusion criteria for all participants included any diagnosed psychological or developmental disorder.

Participant recruitment was multifaceted to encourage a broad sample of participants and reduce sampling bias. For children with hearing loss, direct invitations were sent to families via clinics within New South Wales that fit the inclusion criteria, and flyers were distributed to clinics and hearing/deafness advocacy groups for distribution in newsletters and on social media. To recruit TH children, flyers were distributed throughout Macquarie University campus. Written parental consent and participant assent were obtained before commencement of

| TABLE | 1. D | emographi | TABLE 1. Demographic information for children with SNHL | for ch | ildren with SN | IHL | | | | | | | |
|----------|-----------|---------------------------------|---|----------|----------------------|---------------------------|-------------------------|---------------------|-----------|----------|-------------------------------|---------------------|-------------|
| | | Age/ | Age at | | Formal | | | | | | | | |
| 0 0 | Group | Hearing Age* | First Fitting/ Implantation | Sex | Music Experience† | Degree of Hearing Loss | Device Configuration | Device | Processor | Strategy | Active Electrodes Etiology | Etiology | Schooling |
| HL1 | | 6.3/6.0 | 0.3 | L | 0 | L: profound | ō | L: CI422 (SRA) | L: CP910 | L: ACE | L: 22 | Unknown | Specialized |
| | | | | | | R: profound | | R: CI522 (SRA) | R: CP910 | R: ACE | R: 22 | | |
| HL3 | - | 8.3/7 | 1.3 | Σ | 3.7 | L: profound | Ū | L: CI24RE (ST) | L: CP810 | L: ACE | L: 7 | Pneumococcal | Mainstream |
| | | | | | | R: profound | | R: CI522 (ST) | R: CP810 | R: ACE | R: 22 | meningitis | |
| HL5 | - | 6.1/3.1 | 3.0 | ш | 0.7 | L: profound | Bimodal | L: CI24RE (ST) | L: CP910 | L: ACE | L: 22 | Enlarged vestibular | Specialized |
| | | | | | | R: moderate | | R: Siemens Motion M | | | | aqueduct | |
| HL6 | - | 7.8/7.5 | 0.3 | Σ | 1.3 | L: moderately severe | Bimodal | L: Phonak BTE | R: CP910 | R: ACE | R: 22 | Unknown | Mainstream |
| | | | | | | R: severe | | R: CI512 (CA) | | | | | |
| HL8 | - | 8.5/7.7 | 0.8 | ш | 4.2 | L: moderately severe | HA | L: Siemens Motion P | | | | Usher syndrome | Mainstream |
| | | | | | | R: moderately severe | | R: Siemens Motion P | | | | | |
| HL11 | N | 6.7/6.2 | 0.5 | ш | 0 | L: moderately severe | Bimodal | L: Siemens BTE | R: CP910 | R: ACE | R: 22 | Hypoplasia of the | Mainstream |
| | | | | | | R: profound | | R: CI24RE (CA) | | | | auditory nerve | |
| HL12 | 2 | 7.8/5.8 | 2.0 | Σ | 4.3 | L: profound | Ū | L: CI24RE (CA) | L: CP920 | L: ACE | L: 22 | Unknown | Mainstream |
| | | | | | | R: profound | | R: CI24RE (CA) | R: CP920 | R: ACE | R: 22 | | |
| HL14 | N | 6.7/4.9 | 1.8 | ш | 0.2 | L: profound | Ū | L: CI24RE (CA) | L: CP920 | L: ACE | L: 22 | Waardenburg | Specialized |
| | | | | | | R: profound | | R: CI24RE (CA) | R: CP920 | R: ACE | R: 21 | syndrome type 2 | |
| HL15 | N | 6.3/6.0 | 0.3 | Σ | 1.3 | L: profound | Ū | L: CI512 (unknown) | L: CP920 | L: ACE | L: 22 | Unknown | Mainstream |
| | | | | | | R: profound | | R: CI422 (unknown) | R: CP920 | R: ACE | R: 22 | | |
| HL16 | 0 | 8.6/8.5 | 0.1 | Σ | 4.8 | L: moderately severe | HA | L: Phonak BTE | | | | Genetic | Mainstream |
| | | | | | | R: moderately severe | | R: Phonak BTE | | | | | |
| HL17 | N | 6.8/6.7 | 0.1 | ш | 4.5 | L: profound | Bimodal | L: Concerto FLEX28 | L: Sonnet | L: FS4 | L: 12 | Connexin 26 | Mainstream |
| | | | | | | R: severe | | R: Siemens BTE | | | | | |
| HL18 | 2 | 9.2/7.2 | 2.0 | ш | 0 | L: profound | ō | L: CI24RE (ST) | L: CP910 | L: ACE | L: 22 | Unknown | Specialized |
| | | | | | | R: profound | | R: CI24RE (ST) | R: CP910 | R: ACE | R: 19 | | |
| HL19 | 2 | 6.8/6.3 | 0.5 | Σ | 0 | L: profound | ō | L: CI24RE (CA) | L: CP910 | L: ACE | L: 22 | Genetic | Specialized |
| | | | | | | R: profound | | R: CI24RE (CA) | R: CP910 | R: ACE | R: 22 | | |
| HL20 | N | 8.8/8.4 | 0.4 | Σ | 3.6 | L: profound | ō | L: CI512 (CA) | L: CP910 | L: ACE | L: 22 | Connexin 26 | Mainstream |
| | | | | | | R: profound | | R: CI512 (CA) | R: CP910 | R: ACE | R: 20 | | |
| | | | | | | | | | | | | | |
| *Measure | ∋d at th∈ | *Measured at the pretime point. | | | | | | | | | | | |

*FFormal 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 as follows: music lessons, singing groups, instrumental groups, dance classes, and group-based classes. As an example, 1 yr of weekly piano lessons = 0.7.*CI, cochlear implant; HA, hearing loss; SNHL, sensorineural hearing loss. Measured at the pretime point.

TABLE 2. Demographic information for TH children

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

TH, typical-hearing.

testing, and approval for this study was granted by the Macquarie University Human Research Ethics Committee (Medical Sciences); reference: 5201600081.

Experimental Design

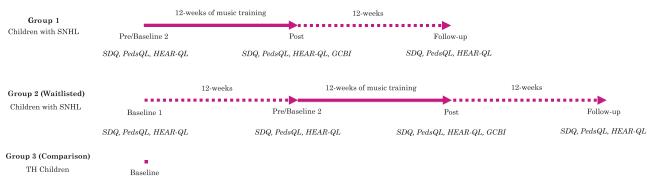
The present study forms the second part of a larger research program that investigated the benefits of music training for children with hearing loss. The first part was focused on the outcomes for perceptual skills and music appreciation (Lo et al. 2020), while the present study focused on psychosocial and quality of life outcomes. Using a longitudinal double-baseline waitlist design, data collection occurred over a period of 9 mo. Children with SNHL were pseudorandomly assigned to commence music training immediately (group 1, n = 5) or placed in the waitlisted group (group 2, n = 9) that commenced music training 12 weeks later. Because the study required a considerable time-commitment, participants were pseudorandomly assigned, to offer flexibility for participating families (i.e., if start dates were not suitable for participation they could opt for the other group).

Double-baseline testing occurred for group 2 (separated by 12 weeks) to provide a baseline measure of natural development and maturation over a 12-week period, thus the waitlisted group

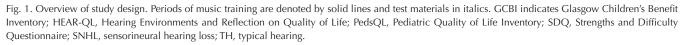
acted as a control group during this period. This technique maximized the statistical power of the small sample size, by not having to split the cohort into a training and control group. There is also an ethical rationale to this approach that allowed all participants to engage in a potentially beneficial program. On the other hand, the disadvantage of not having an active control group is that we cannot completely attribute specific benefits to the music program (i.e., would a nonmusic training program provide similar benefits?). Participants then completed the questionnaires after the full 12 weeks of music training (post); and finally, 12 weeks after training was completed to measure retention (follow-up). An additional cohort of age-matched TH children was included as a comparison group and completed the questionnaires once. An overview of this design can be viewed in Figure 1.

Materials and Methods

As the role of psychosocial wellbeing and quality of life is mostly unexplored for music-based interventions for children with hearing loss; our strategy was to utilize two widely used and validated questionnaires as a general measure of psychosocial wellbeing (Strengths and Difficulties Questionnaire) and quality of life (Pediatric Quality of Life Scale), alongside a measure designed specifically for a population with hearing loss (HEAR-QL). A combination of both generic and specific measures is recommended to fully comprehend the full extent of quality of life (Solans et al. 2008; Warner-Czyz et al. 2011). 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 (Looi et al. 2018, 2019). 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 before commencement of the present study. Musical activities were stratified into six categories (music lessons, singing groups, instrumental groups, special children's programs, dance classes, and group-based music classes). Each activity provided a score that was calculated by duration of participation (in years), multiplied by its frequency (1 = less oftenthan 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). As there were six activities, each







activity had a 1/6 weighting toward the total score. As an example, 1 year of weekly piano lessons = $1 \ge 4 \div 6 = 0.7$.

Strengths and Difficulties Questionnaire • The SDQ was developed by Goodman (1997) as a brief behavioral screening questionnaire that provides an overview of children's psychosocial wellbeing through behavior, emotion, and relationships, and has been used effectively for children with hearing loss such as in Wong et al. (2017). It consists of 25 items equally subdivided into 5 hypothesized subscales: Hyperactivity, Emotional symptoms, Conduct problems, Peer problems, and Prosocial. Example items for each of these 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. As computerized scoring is recommended, scores were calculated using the SDQ SPSS Syntax (Youthinmind Ltd. 2016).

Based on the age of present study's cohort, parent-reported versions (recommended by Goodman et al. (2010) 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 et al. (2010). The additional advantage of this approach was a reduction of measurement error (Goodman et al. 2010).

Pediatric Quality of Life Inventory • The PedsQL Inventory was developed by Varni et al. (1999) as a generic measure of health-related quality of life that consists of a Generic Core Scale (GCS) and various condition-specific modules. Healthrelated quality of life broadly encapsulates physical health, psychosocial health, and social interaction, and has become a dominant quality of life measure (Wallander & Koot 2016). The PedsQL has been used successfully in previous studies for children with hearing loss such as in Looi et al. (2016). The 23-item PedsQL GCS consists of 4 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 the "parent-report" and "child-report" formats were used. The self-reports for children aged 8 to 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 to 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 PedsOL scales, a higher score was indicative of a better health-related quality of life. To create the Psychosocial Health Summary Score, the mean was calculated as the sum of the items over the number of items answered in the Emotional, Social, and School Functioning Scales. The Physical Health Summary Score was the same as the Physical Functioning Scale Score. To create the Total Scale Score, the mean was computed as the sum of all the items over the number of items answered on all the Scales. Scoring rules are available at the PedsQL Scoring Algorithm site (Varni 2020).

Hearing Environments and Reflection on Quality of Life-26 • The HEAR-QL is a quality of life assessment tool designed specifically for children with hearing loss (Umansky et al. 2011). The 26-item HEAR-QL-26 is designed for self-report in children aged between 7 and 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 health-related quality of life. Mean scores for each subscale (Environments, Activities, and Feelings) and for the overall HEAR-QL are computed as the sum of scores for items on each subscale (or total measure) divided by the number of items completed.

The Glasgow Children's Benefit Inventory • Unlike the majority of questionnaires that make an assessment at a single point in time, the Glasgow Children's Benefit Inventory (GCBI) was designed as a postintervention health-related benefit measure (Kubba et al. 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. This questionnaire was only completed by the children with SNHL at the completion of the 12-week music training program.

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. After completing the perceptual tasks as described in Lo et al. (2020), questionnaires were presented in a fixed order at the following test sessions for parents: SDQ and PedsQL GCS (baseline, post, and follow-up), and GCBI (post); and for children: PedsQL GCS, HEAR-QL-26 (baseline, post, follow-up). Questionnaires were paper-based, and the experimenter read aloud each questionnaire item directly to the children, in order for them to be able to seek clarification at any time. Children responded either verbally or by pointing to their selection. Honest responses were emphasized at each session, and children were not allowed to consult or discuss their responses with their parents, who completed the questionnaires independently.

Participants were asked to respond to all questionnaires with a 1-mo recall period. Testing was administered by one of 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. Each test session generally took around 1 hr to complete. 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 for the children with SNHL, with a focus on maximizing access to a broad range of musical skills and activities. The curriculum consisted of weekly, 40-min, 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. This resulted in a total of 8 hr of face-to-face music training over a 12-week period. Music therapy is "a research-based practice and profession in which music is used to actively support people as they strive to improve their health, functioning and wellbeing" (Australian Music Therapy Association 2012). Examples of music therapy activities include drumming, singing, dancing, and improvisation, all of which involved group interaction. The full curriculum is available (see Appendix in Supplemental Digital Content, http://links.lww.com/EANDH/A838).

The activities were based on the Nordoff-Robbins (or Creative Music Therapy) approach that places an emphasis on active music activities between the music therapist and their clients (Nordoff et al. 2007). The Nordoff-Robbins approach placed a significant emphasis on interactive, social, groupbased activities which often required cooperation and turn-taking. Generally, instructions involved a combination of verbal and musical commands which became increasingly musical (and less verbal) as the participants progressed through the curriculum. Specific rehabilitative characteristics of the music training include (a) music consists of spectrally complex and diverse sounds that require attentive listening which may push the auditory system beyond its typical requirements (Limb & Roy 2014); (b) the multisensory nature of music such as playing instruments or using movement and dancing provides helpful visual and vibrotactile cues that can supplement auditory perception (Zimmerman & Lahav 2012; Vongpaisal et al. 2016); (c) the use of group-based musical activities often involved more than one person playing or singing at a time, thus, encouraging turn-taking, cooperation, and prosocial behaviors (Kirschner & Tomasello 2010; Schellenberg et al. 2015).

The first two authors consulted with the music therapist to provide an understanding of hearing loss and hearing technology to guide development of the curriculum. Accommodations included being aware of the importance of facial and body cues due to difficulties with auditory perception, and techniques to incorporate vision and motor skills to support listening. It should be noted that many of these strategies were already inherent to the Nordoff-Robbins approach and music therapy in general.

Participants were also expected to complete a series of online music activities 3 times a week (approximately 15 to 30 min depending on ability) with MusicFirst Junior (Music Sales Group 2020)—an online-based suite of music apps designed for children aged between 6 and 12 years. These apps are compatible for PC/Mac/smart devices and 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 set 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 the music apps (which were single-user experiences) include

"drawing" melodic contours and creating compositions, and identification of high, low, fast, or slow sounds typically through multiple-choice or matching games.

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 (e.g., the possibility of adjusting difficulty based on individual performance and ability, and replicability), which also have the additional benefit of data-logging the activities. Thus, this hybrid approach of face-to-face group-based activities, supplemented by onlinebased apps encouraged maximum participation in a broad range of musical activities during a limited timeframe.

RESULTS

Statistical Analyses

IBM SPSS Statistics (version 22) was used to perform main hypothesis testing using generalized linear mixed (GZLM) models. A significant advantage with the GZLM model was to account for differences in the distribution of responses. For example, the majority of SDQ responses was not normally-distributed and were better suited to an analysis with a gammadistribution. Asymmetric responses in the SDQ have also been noted in the longitudinal Millennium Cohort Study (n = 11,972 observations) (Tzavidis et al. 2016). Another advantage with GZLM models is that it can accommodate 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 GCS 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 = 0.05.

For the music training analyses of the children with SNHL (n = 11), the following fixed effects were entered: time (baseline 1, pre/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-parametrization (due to the small sample size) by consolidating chronological age and age at fitting/implantation as one variable. While formal music training (as measured by the RMFQ) and compliance with the face-toface and online music training were fixed effects of interest, the model failed to converge, hence they were not included in the analyses. The inclusion of device and hearing age was selected given the assumption that both fixed effects would have a significant impact on perception and development. Given the children were relatively young (aged between 6 and 11 years), there was also less time and opportunity for formal music experience to be widely divergent when compared with older cohorts.

For all analyses of the children with SNHL, participants were entered as random effects with random intercepts (random slopes were of interest but failed to converge). Visual inspection of Q-Q plots indicated that SDQ measures for Internalising, Externalising, and Total scores were gamma-distributed (and thus analyzed 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 pre/baseline 2 as a reference point, except for the GCBI which was only measured at the post-training time point. The use of pre/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-training), and the retention of any benefit (follow-up).

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 pretraining measurement. Initial comparisons to TH children were made using independent-samples t tests in respect to raw pre/baseline 2 measures (as the models to calculate each group's estimated marginal means were not equivalent).

Attendance and Compliance

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 1 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%). In addition, 1 participant also left the study in week 8 due to a surgical procedure.

Strengths and Difficulty Questionnaire

A summary of results for the SDQ scales can be found in Table 3 and Figure 2. The total range of scores for Internalising and Externalising problems is from 0 to 20, the Prosocial scale is from 0 to 10, and the Total difficulties is from 0 to 40.

At baseline, children with SNHL had SDQ Internalising problems that were significantly higher by 3.0 points, 95% CI (1.1–4.9) than their TH peers, t(26) = 3.21, p = 0.003. In

addition, compared with TH peers, Externalising problems were 1.2 points higher, 95% CI (-2.3 to 4.7), Prosocial scores were 0.6 points lower, 95% CI (-2.1 to 0.9), and Total difficulties were 4.2 points higher, 95% CI (-0.6 to 9.0), but these were not significantly different (t(26) = 0.70, p = 0.488; t(26) = -0.85, p = 0.402; t(26) = 1.79, p = 0.085, respectively).

The change scores for the children with SNHL are as follows: 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 = 0.001, and this was retained at the follow-up time point with a decrease of 2.5 points, F(3,16) = 5.4, p = 0.036. 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 = 0.012; however, this improvement was not maintained at follow-up, F(3,12) = 2.4, p = 0.148. A posthoc analysis comparing children with SNHL at the post-training time point and TH children were not significantly different for SDQ Internalising problems, t(24) = -0.19, p = 0.854, and Total difficulties, t(24) = -0.09, p = 0.927 indicating parity after the music training program. There was no change across time for Externalising problems or the Prosocial scale. Device and hearing age were not significant factors in this model.

Pediatric Quality of Life Inventory GCS

A summary of results for the PedsQL GCS across time points for children with SNHL can be seen in Table 4. The total range of scores is from 0 to 100. At baseline, children with SNHL had slightly lower scores than their TH peers, although there was no statistically significant difference between them for any PedsQL measure (Table 5).

In terms of the effect of training for the children with SNHL, 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. Interrater reliability was examined

TABLE 3. Results from the GZLM for SDQ scales across time points for children with SNHL

| | Parent Report | | | 95% CI | | |
|------------------------|---------------|-------|--------|--------|-------|--|
| SDQ Scales | (M, SE) | t | p | Lower | Upper | |
| Internalizing Problems | | | | | | |
| Baseline 1 | 5.1 (1.3) | -0.62 | 0.559 | 2.7 | 9.5 | |
| Pre/baseline 2 | 6.0 (1.0) | | | 4.2 | 8.7 | |
| Post | 2.5 (0.4) | -4.20 | 0.001* | 1.7 | 3.7 | |
| Follow-up | 3.5 (0.7) | -2.32 | 0.036* | 2.2 | 5.5 | |
| Externalizing Problems | | | | | | |
| Baseline 1 | 6.5 (1.3) | 1.07 | 0.306 | 4.0 | 10.4 | |
| Pre/baseline 2 | 5.2 (1.2) | | | 3.1 | 8.6 | |
| Post | 4.4 (0.9) | -0.73 | 0.476 | 2.7 | 7.3 | |
| Follow-up | 5.0 (0.9) | -0.24 | 0.815 | 3.2 | 7.7 | |
| Prosocial | | | | | | |
| Baseline 1 | 7.8 (0.9) | -0.71 | 0.531 | 6.0 | 10.0 | |
| Pre/baseline 2 | 8.1 (0.9) | | | 6.3 | 10.4 | |
| Post | 9.1 (1.1) | 1.65 | 0.126 | 6.9 | 12.2 | |
| Follow-up | 8.0 (1.0) | -0.19 | 0.853 | 5.9 | 10.8 | |
| Total Difficulties | () | | | | | |
| Baseline 1 | 11.3 (1.9) | -0.16 | 0.874 | 7.7 | 16.6 | |
| Pre/baseline 2 | 11.7 (2.1) | | | 8.0 | 17.2 | |
| Post | 6.9 (0.9) | -2.87 | 0.012* | 5.0 | 9.5 | |
| Follow-up | 8.8 (1.1) | -1.54 | 0.148 | 6.5 | 11.9 | |

* $p \leq 0.05$, relative to measurement at pre/baseline 2.

GZLM, generalized linear mixed; SDQ, Strengths and Difficulty Questionnaire; SNHL, sensorineural hearing loss.

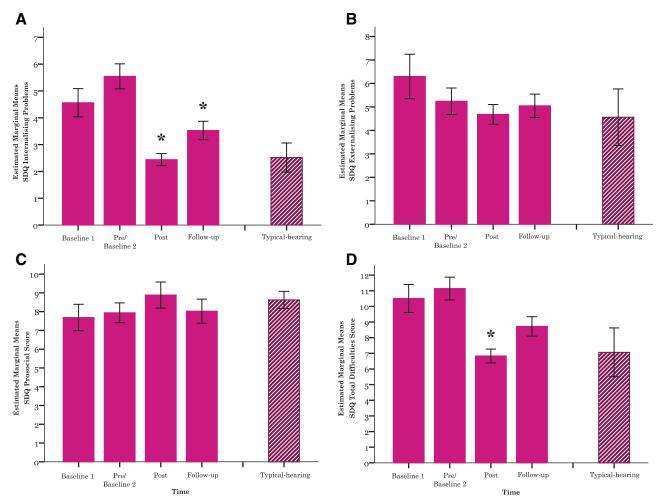


Fig. 2. Bar graphs of estimated marginal means for SDQ subscales across time with a comparison of TH children's performance: (A) internalizing problems, (B) externalizing problems, (C) prosocial, (D) total difficulties. $*p \le 0.05$ compared with pre/baseline 2. Error bars: ± 1 SE. SDQ indicates Strengths and Difficulty Questionnaire; TH, typical hearing.

TABLE 4. Results from the GZLM for PedsQL GCS across time points for children with SNHL

| | Parent Report | | | 95% | 6 CI | Child Report | Child Report | | 95% CI | |
|---------------------|---------------|-------|-------|-------|-------|--------------|--------------|-------|--------|-------|
| PedsQL Scale | (M, SE) | t | р | Lower | Upper | (M, SE) | t | р | Lower | Upper |
| Psychosocial Health | | | | | | | | | | |
| Baseline 1 | 61.1 (4.1) | -1.26 | 0.230 | 50.5 | 71.6 | 65.1 (5.9) | -0.27 | 0.979 | 50.4 | 79.7 |
| Pre/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 | 0.203 | 69.0 | 80.7 | 60.3 (5.1) | -1.07 | 0.333 | 49.1 | 71.6 |
| Follow-up | 69.2 (3.2) | 0.16 | 0.872 | 62.0 | 76.5 | 63.1 (6.0) | -0.38 | 0.714 | 47.8 | 78.5 |
| Physical Health | | | | | | | | | | |
| Baseline 1 | 75.5 (5.3) | -0.66 | 0.525 | 63.1 | 87.9 | 66.0 (6.0) | 0.20 | 0.853 | 52.1 | 79.9 |
| Pre/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 | 0.960 | 69.9 | 89.0 | 62.5 (7.7) | -0.47 | 0.651 | 45.4 | 79.6 |
| Follow-up | 74.0 (8.7) | -0.59 | 0.569 | 54.4 | 93.6 | 66.4 (10.3) | 0.12 | 0.909 | 43.3 | 89.5 |
| Total Score | | | | | | | | | | |
| Baseline 1 | 65.8 (4.0) | -1.14 | 0.293 | 45.6 | 86.1 | 65.3 (5.9) | -0.74 | 0.944 | 51.3 | 79.3 |
| Pre/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 | 0.372 | 70.3 | 81.9 | 61.3 (5.2) | -1.00 | 0.345 | 49.7 | 72.9 |
| Follow-up | 70.4 (3.8) | -0.30 | 0.769 | 61.7 | 79.0 | 64.6 (6.1) | -0.21 | 0.839 | 50.3 | 78.8 |

GCS, Generic Core Scale; GZLM, generalized linear mixed; PedsQL, Pediatric Quality of Life Inventory; SNHL, sensorineural hearing loss.

between parent and child responses across all time points. For the psychosocial health measure, ICC = 0.37, with a 95% CI (-0.27 to 0.69); for the physical health measure, ICC = 0.01, with a 95% CI (-0.82 to 0.48); and for the total score, ICC = 0.31, with a 95% CI (-0.34 to 0.65)—all of which suggest poor reliability on average (Cicchetti 1994; Koo & Li 2016).

Hearing Environments and Reflection on Quality of Life-26

A summary of results for all HEAR-QL-26 domains across all time points for children with SNHL can be seen in Table 6. The total range of scores is from 0 to 100. At baseline, compared with 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 to -9.3), Activities were 14.5 points lower, 95% CI (-27.3 to -1.8), Feelings were 25.7 points lower, 95% CI (-42.7 to -8.7), and Totals were 20.4 points lower, 95% CI (-30.1 to -10.8), all of which were significantly different [t(24) = -3.68, p < 0.001; t(10) = -2.54, p = 0.0.30; t(12) = -3.29, p = 0.007; and t(24) = -4.36, p < 0.001 respectively]. 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.

Glasgow Children's Benefit Inventory

The GCBI was administered to the children with SNHL at the post-training time point and was evaluated with a Wilcoxon signed-rank test with a hypothesized median of interest set to 0 = no change. Table 7 indicates that after music training, a statistically significant improvement was observed for overall life, p = 0.015; the capacity to do things, p = 0.014; better behavior, p = 0.020; progress and development, p = 0.009; learning, p = 0.046; and confidence, p = 0.025, 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.

DISCUSSION

The aim of the present study was to evaluate psychosocial and health-related quality of life 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 internalizing and externalizing problems, psychosocial and physical health, as well as hearing-specific questions targeting environments, activities, and feelings. The primary finding was that internalizing problems were significantly reduced at the posttraining time point, which were also retained at follow-up. The SDQ Total Score was also significantly improved at the posttraining time point, though not retained at follow-up. Somewhat surprisingly, there were no benefits for prosocial outcomes. In addition, responses from the GCBI suggest a generally positive effect of training, with benefits primarily around emotional and learning factors. It should be noted that significant benefits were only found from the parent reports of the SDQ and GCBI, with no significant improvements from any of the child-reported questionnaires.

At baseline, children with hearing loss had poorer outcomes for internalizing problems, and all measures of the hearingspecific questionnaire when compared with the TH children; 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, rather than the effects of natural maturation and development.

Parent reports of children with hearing loss generally indicate greater internalizing and peer problems than parent reports of TH children (van Eldik et al. 2004; Barker et al. 2009; Hoffman et al. 2015). The results from the present study are encouraging, as internalizing problems (measured by the SDQ as the sum of peer and emotional problems) were improved after training and maintained 3 mo after training had ceased. Posthoc analyses also suggest their post-training results were at parity with their TH peers. Anecdotal evidence suggests that music training supports prosocial behaviors for children with hearing loss (Innes-Brown et al. 2013). However, the prosocial scale in the present study showed no change across time. In TH children, the evidence is mixed, with a review by Dumont et al. (2017) indicating positive findings in three studies (one of which was designated as high quality), and another study reporting no benefits. In the present study, total scores on the SDQ were also significantly improved at the posttraining time point, but not maintained at follow-up. This suggests that sustaining longer-term benefits may require ongoing participation in musical activities.

Measuring quality of life in children presents a number of challenges. While best practice is to use parent- and selfreported measures in tandem to provide a comprehensive understanding of a child's quality of life, many measures (and studies) are designed to rely exclusively on parent reports; particularly if children may not have the capacity (e.g., due to age, illness, disability) to reliably self-report (Upton et al. 2008; Umansky et al. 2011). The SDQ is one such case, which is

| TABLE 5. Independent | t t-tests of PedsQ | L between childrer | with SNHL | and TH |
|----------------------|--------------------|--------------------|-----------|--------|
| | | | | |

| | | | | 95 | % Cl |
|--|------------------|-------|-------|-------|-------|
| SDQ Scales | M, SE Difference | Т | р | Lower | Upper |
| PedsQL Psychosocial Health (parent reported) | -7.8 (5.3) | -1.46 | 0.158 | -18.8 | 3.2 |
| PedsQL Psychosocial Health (child reported) | -7.9 (5.3) | -1.49 | 0.150 | -18.9 | 3.1 |
| PedsQL Physical Health (parent reported) | -1.0 (8.2) | -0.12 | 0.907 | -17.7 | 15.8 |
| PedsQL Physical Health (child reported) | -5.6 (6.1) | -0.92 | 0.365 | -18.1 | 6.9 |
| PedsQL Total (parent reported) | -5.4 (5.9) | -0.92 | 0.368 | -17.5 | 6.7 |
| PedsQL Total (child reported) | -7.1 (5.0) | -1.43 | 0.166 | -17.4 | 3.2 |

PedsQL, Pediatric Quality of Life Inventory; SDQ, Strengths and Difficulty Questionnaire; SNHL, sensorineural hearing loss; TH, typical hearing.

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| | Parent Report | | | 95 | % Cl |
|-----------------|---------------|-------|-------|-------|-------|
| HEAR-QL Domains | (M, SE) | t | Р | Lower | Upper |
| Environments | | | | | |
| Baseline 1 | 63.0 (7.7) | 0.17 | 0.871 | 44.3 | 81.7 |
| Pre/baseline 2 | 61.6 (4.6) | | | 50.8 | 72.4 |
| Post | 57.0 (3.6) | -1.01 | 0.332 | 48.1 | 65.8 |
| Follow-up | 62.5 (6.5) | 0.13 | 0.900 | 47.6 | 77.4 |
| Activities | | | | | |
| Baseline 1 | 79.5 (7.7) | 0.30 | 0.770 | 61.6 | 97.5 |
| Pre/baseline 2 | 77.3 (5.2) | | | 65.9 | 88.8 |
| Post | 74.4 (4.8) | -0.76 | 0.473 | 63.1 | 85.7 |
| Follow-up | 82.8 (7.1) | 0.86 | 0.414 | 66.4 | 99.2 |
| Feelings | | | | | |
| Baseline 1 | 67.3 (7.2) | 0.27 | 0.790 | 46.0 | 88.5 |
| Pre/baseline 2 | 64.5 (7.8) | | | 46.8 | 82.2 |
| Post | 58.9 (8.5) | -0.50 | 0.630 | 38.8 | 79.1 |
| Follow-up | 46.7 (7.9) | -1.67 | 0.124 | 26.3 | 67.0 |
| Total | | | | | |
| Baseline 1 | 70.6 (7.3) | 0.32 | 0.757 | 52.7 | 88.5 |
| Pre/baseline 2 | 68.0 (5.0) | | | 56.4 | 79.7 |
| Post | 63.5 (3.5) | -0.95 | 0.365 | 54.9 | 72.0 |
| Follow-up | 65.7 (5.0) | -0.39 | 0.706 | 54.6 | 76.9 |

TABLE 6. Results from the GZLM for HEAR-QL-26 across time points for children with SNHL

GZLM, generalized linear mixed; HEAR-QL-26, Hearing Environments and Reflection on Quality of Life; SNHL, sensorineural hearing loss.

parent-reported for the age range of the present study's cohort. Internalizing problems have been noted as being easy to miss by parents, as these behaviors are less visible and obvious than externalized ones (Clarke-Stewart et al. 2003). Nonetheless, irrespective of this, internalized behaviors were notably improved for up to a 6-mo period in this study (as measured from pre/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 significantly different difficulties for psychosocial or physical health when compared with their TH peers. Consequently, there may have been little room for measurable improvement. Given the SDQ indicated improvement around internalizing behaviors, it might be expected that the PedsQL GCS may also have shown

TABLE 7. GCBI results after music training for children with SNHL

| Has Your Child's Participation in the Music Program | р | Observed Median |
|---|--------|---------------------|
| 1. Made their overall life better or worse? | 0.015* | 1 = a little better |
| 2. Affected the things they do? | 0.014* | 1 = a little better |
| 3. Made their behavior better or worse? | 0.020* | 1 = a little better |
| 4. Affected their progress and development? | 0.009* | 1 = a little better |
| 5. Affected how lively they are during the day? | 0.059 | 0 = no change |
| 6. Affected how well they sleep at night? | 0.317 | 0 = no change |
| 7. Affected their enjoyment of food? | 1.000 | 0 = no change |
| 8. Affected how self-conscious they are with people? | 0.317 | 0 = no change |
| 9. Affected how well they get on with the rest of the family? | 0.157 | 0 = no change |
| 10. Affected their ability to spend time and have fun with friends? | 0.102 | 0 = no change |
| 11. Affected how embarrassed they are with other people? | 0.317 | 0 = no change |
| 12. Affected how easily distracted they have been? | 0.059 | 0 = no change |
| 13. Affected their learning? | 0.005* | 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? | 0.020* | 1 = a little better |
| 16. Affected how irritable they are? | 0.180 | 0 = no change |
| 17. Affected how they feel about themselves? | 0.059 | 0 = no change |
| 18. Affected how happy and content they are? | 0.046* | 0 = no change |
| 19. Affected their confidence? | 0.025* | 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? | 0.157 | 0 = no change |
| 21. Affected their ability to enjoy leisure activities such as swimming and sports, and general play? | 0.083 | 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? | 0.317 | 0 = no change |
| 24. Affected how much medication they need to take? | 1.000 | 0 = no change |

* $p \le 0.05$, relative to a hypothesized median = 0 (no change).

GCBI, Glasgow Children's Benefit Inventory; SNHL, sensorineural hearing loss.

some positive change, particularly on the Psychosocial Health Summary Score. However, a study of TH children and adolescents by Stevanovic (2013) found poor correlations between the two questionnaires, suggesting less overlap than expected. The PedsQL GCS also utilized data from both child and parent, which may be a significant factor, noting that interrater reliability between parents and children for the PedsQL GCS was poor. This was unsurprising, as existing research has shown that quality of life concordance between parent and child ratings is highly inconsistent (Upton et al. 2008), and these differences reflect separate perspectives, all of which are relevant and valuable (Jozefiak et al. 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 was similarly poor. However, on the basis of previous findings and given the small sample size, this is not an unexpected finding. In addition, Looi et al. (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, or that hearing age could be a more suitable measure when interpreting age-related guidelines. It is interesting that 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-training 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. Children tend to evaluate their quality of life 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.

To assess hearing-related quality of life, the HEAR-QL-26 was specifically used, as it is a validated self-reported 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 were no significant changes in HEAR-QL scores across time points. Part one of this study focused on perceptual outcomes, and there were significant benefits resulting from the music training such as improved SIN, spectral resolution, and timbre perception (Lo et al. 2020). Thus, it was surprising this did not transfer to better hearing-related quality of life. 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 period of time before these improvements are reflected in a self-reported questionnaire. At face-value, 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. A study by Klatte et al. (2010) investigated the effect of noise and reverberation in classroomlike settings with first and third grade TH children. While noise and reverberation had a clear negative effect on their speech perception performance, the children's subjective appraisal of disturbance ratings were low, suggesting they were unable to estimate the effect of disruption.

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 with "0" indicating no change. However, there are no reporting guidelines as to what constitutes a clinically significant score for the GCBI. As a comparison, Roland et al. (2016) performed a meta-analysis of 5 studies using GCBI outcomes after children received bone-anchored hearing aids resulted in a total average score of +43, 95% CI (25-56), while Sparreboom et al. (2012) found a benefit of +16.6 (no confidence interval reported) for 30 children with prelingual deafness that received sequential bilateral cochlear implantation (first implant, M = 1.8years; second implant, M = 5.3 years). Overall, the findings from the present study suggest 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 internalizing behaviors, although evidence that music may improve attention and executive functioning, factors 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 internalizing behaviors (as measured by the SDQ), as well as learning and emotion factors (as measured by the GCBI). First, participation in the face-to-face group-based music therapy sessions required engagement in activities geared toward turn-taking, coordinated re/action, peer interaction, imitation, and emotional expression. This shared intentionality through joint music-making activities was geared toward encouraging shared and cooperative behaviors, which Kirschner and Tomasello (2010) hypothesize improves peer interactions and creates a sense of belonging. The children were also provided the opportunity to develop their skills using a variety of musical tasks such as singing, dancing, playing instruments, as well as leading various activities, which may have facilitated feelings of competence, and a sense of achievement (Hallam et al. 2016).

Second, there may have been a social benefit primarily for the children who were mainstream schooled (n = 8). Anecdotally, the majority of participants who received mainstream-education were also the only child with hearing loss at their school. Many studies have reported that children with hearing loss in mainstream schools often report experiences of loneliness and social isolation (Schorr 2006; Most et al. 2012). The "hearing aid effect" is noted as a generalized stigma associated with wearing HAs (Cameron et al. 2008), that may result in anxieties around acceptance within peer groups (Punch & Hyde 2011). Similarly, a study by Dammeyer et al. (2018) investigated the experiences and attitudes of 65 adolescent CI recipients aged 11 to 15 years and found that 55.4% reported feeling different from others their age, and 18.5% reported attempts to hide their CIs "all the time" or "often." However, as the music therapy sessions were conducted exclusively with other children with hearing loss, this may have alleviated social anxieties and resulted in a greater sense of belonging.

Finally, the improvements may have been a result of better communication skills. A number of perceptual enhancements were noted, as reported in part one of this study (Lo et al. 2020), including SIN perception and question/statement prosody. These results are compatible with the hypothesis that Summerfield and Marshall (1999) put forward, suggesting that outcomes such as speech perception (which the authors classified as a short-term outcome) have a cascading effect on quality of life (classified as long-term by the authors), although it should be noted that the time scale that constitutes short-term and long-term effects are not well defined. In addition, while SIN and prosodic perception are an important perceptual aspect of communication, the present study did not directly assess broader communication ability which consists of a myriad of dynamic and interlinked 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 of the present study. Without an active control group, it is plausible that improvements were not music-specific and could have been attained from any groupbased social activity. The small sample size is not unusual for longitudinal studies of children with hearing loss incorporating training programs or interventions; however, it does reduce statistical power and generalizability. 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, whereas the children were likely naive to the overarching aims of the study.

An open question that remains is the time course that one would expect quality of life 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 mo to establish significant change from pre/baseline 2 to the follow-up time point; that some improvement was noted warrants cautious optimism. Another point to note is that the majority of children in this study were using CIs which are generally associated with poorer music perception than children with less hearing loss (Looi et al. 2012; Innes-Brown et al. 2013). Would music training benefit children with poorer hearing loss, as they have more room for improvement? Or does better auditory acuity facilitate better musical learning outcomes? Lengthier longitudinal studies confirming the reliability of these findings, with larger samples, investigating a variety of group-based interventions, and across a range of ages are likely to provide additional insights and perspectives.

There is a paucity of evidence linking music training to psychosocial and quality of life 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 utilized a range of generic and disease-specific, validated questionnaires, with the preliminary finding that music training may improve psychosocial and health-related quality of life outcomes for children with hearing loss. The primary areas of improvement were related to internalizing behaviors, along with factors associated with emotions and learning. These findings provide initial evidence that the general psychosocial and health-related quality of life benefits noted in music studies for TH children may be applicable to pediatric populations with hearing loss. While the mechanisms are likely different and at present not clear, 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 quality of life outcomes (Theunissen et al. 2014; Stevenson et al. 2015). Overall, this study has shown a positive

effect of group-based musical activity supplemented with music apps for children with hearing loss.

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