CEO Clinical and Experimental Otorhinolaryngology Vol. 14, No. 1: 15-28, February 2021
https://doi.org/10.21053/ceo.2020.00101
pISSN 1976-8710 eISSN 2005-0720

Review

Efficacy of Music Training in Hearing Aid and Cochlear Implant Users: A Systematic Review and Meta-Analysis

Nor Farawaheeda Ab Shukor[1,2] · Jihyeon Lee[1,3,4] · Young Joon Seo[3,4] · Woojae Han[1,2]

[1]Laboratory of Hearing and Technology, Research Institute of Audiology and Speech Pathology, Hallym University, Chuncheon; [2]Division of Speech Pathology and Audiology, College of Natural Sciences, Hallym University, Chuncheon; [3]Department of Otorhinolaryngology and [4]Research Institute of Hearing Enhancement, Yonsei University Wonju College of Medicine, Wonju, Korea

This study aimed to evaluate the efficacy of music training on the improvement of musical perception among hearing-impaired listeners using a systematic review and meta-analysis. Article search was conducted from five databases, Scopus, ScienceDirect, Web of Knowledge, CINAHL, and PubMed. A total of 186 participants from 10 studies investigating the music training effects on individuals fitted with hearing assistive devices and outcome measurements were included. The meta-analysis showed standardized mean difference as a measure of the effect size, in musical improvement between the pre- and post-training. Although the funnel plot yielded an asymmetrical graph, the Egger's regression showed no significant publication bias. Interestingly, subgroup analysis showed that the training effect was greater in children than in adults. With a necessity of longer training period to significantly improve their musical perception, cochlear implant only users had better effect compared to bi-modal users with both cochlear implant and hearing aids. However, the difference in the training effect between the users with and without previous musical experience was nonsignificant. The present study concludes that auditory music training brings hearing-impaired listeners into better musical perception while informing that training effects differ depending on age, duration of the training, and the type of hearing device used.

Keywords. *Hearing Loss; Rehabilitation; Auditory Training; Music Perception*

INTRODUCTION

Disabling hearing loss is a common problem faced by over 466 million people throughout the world, accounting for 6.1% of the world's population [1]. The effect of hearing loss on an individual is highly dependent on the severity of the loss, the individual's lifestyle and communication needs, and other factors [2]. Most people suffering from hearing loss can benefit from advanced hearing assistive devices, such as hearing aids and cochlear implants, which record sounds from the surrounding environment, adapt the signal to compensate for the characteristics of the user's hearing loss, and replay the adapted sounds into the user's ear [3]. The primary therapeutic needs of individuals with hearing devices include a better understanding of speech [4].

Aural (re)habilitation is one of the key factors in improving communication skills and promoting the normal development of the speech-language of hearing device users [5]. In the last two decades, studies have investigated music as an auditory training approach. Interestingly, the part of the brain that plays a major role in perceiving speech also plays an important role in the processing of music and other meaningful auditory signals [6,7]. Confirming that speech and music share neural networks, Gfeller [6] suggested that listening to or performing music might have a positive effect on the development of more efficient and robust auditory processes. In addition, Anderson and Kraus [7] found that the perceptual requirements associated with music listening also had implications for auditory training. In light of these results, significant interest has emerged in adapting music training for aural (re)habilitation [8]. Although the significant effects of

• Received January 16, 2020
Revised March 9, 2020
Accepted April 6, 2020

• Corresponding author: **Woojae Han**
Division of Speech Pathology and Audiology, College of Natural Sciences, Hallym University, 1 Hallimdaehak-gil, Chuncheon 24252, Korea
Tel: +82-33-248-2216, Fax: +82-33-256-3420
E-mail: woojaehan@hallym.ac.kr

Table 1. Full electronic search strategies for all databases used in the study

Database	Keyword				
PubMed, Scopus, Web of Knowledge, CINAHL	"Music" OR "Music Training" OR "Music therapy" OR "Acoustic Stimulation" OR "Auditory Training" OR "Auditory Rehabilitation" OR "Auditory stimulation" OR "Musical Stimulation"	AND	"Speech perception" OR "Speech improvement" OR "Auditory perception" OR "Music perception" OR "Speech intelligibility" OR "Loudness perception" OR "Pitch perception" OR "Pitch discrimination"	AND	"Training time" OR "Training duration" OR "Training content" OR "Musical exposure" OR "Music Content"
ScienceDirect	"Music Training" OR "Music therapy" OR Auditory Training"	AND	"Auditory perception" OR "Music perception" OR "Speech intelligibility" OR "Loudness perception"	AND	"Training duration" OR "Training content"
	"Acoustic Stimulation" OR "Music therapy" OR "Musical Stimulation"	AND	"Speech improvement" OR "Music perception" OR "Loudness perception" OR "Pitch perception"	AND	"Training content" OR "Musical exposure" OR "Music Content"

music training have led to further clinical research on whether it can improve speech and/or musical perception as an aural (re)habilitation approach [9], many early studies only explored the role of sub-cortical responses in the detection of musical elements, such as pitch, frequency, timing, and timbre [10]. Further, no previous studies reported specific and concrete findings regarding the clinical implications of music training and its effects in hearing-impaired listeners who wear hearing devices.

Herein, we aimed to determine the therapeutic efficacy of music training in patients with hearing loss who have been fitted with hearing aids, cochlear implants, or both devices, through a systematic review and meta-analysis. Furthermore, another purpose of the present study was to assess potential positive clinical effects of music training based on users' age, the type of hearing device, musical experience before training, and training period.

MATERIALS AND METHODS

Search strategy

The systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [11]. Articles search of the five databases, e.g., Scopus, ScienceDirect, Web of Knowledge, CINAHL, and PubMed, was done systematically in October 2019. The articles that been included in this study have been published from year 1980 to 2019. Table 1 provides full electronic search strategies with a string of keywords.

HIGHLIGHTS

- In patients with hearing loss, music training should be considered as auditory training as it can improve their musical perception.
- Compared to adults with hearing loss, the pediatric patients showed greater benefits from the music training.
- Cochlear implant users have a greater effect on music training compared to the bimodal users.
- Unexpectedly, the previous musical experience was not a significant factor in their music training.
- Long duration of the music training (at least longer than 12 months) provides better performance in terms of musical perception.

Eligibility criteria and study selection

The inclusion criteria for studies in the review were specified in terms of participants, interventions, comparisons, outcomes, and study design (PICOS) as follows [11]. Studies were included in the systematic review if they (1) involved individuals (children and adults) with hearing loss fitted with hearing aids or cochlear implant unilaterally or bilaterally, (2) included participants who underwent music training as rehabilitation, (3) compared pre- and post-rehabilitation effect or repeated measures (experiments with additional purposes), (4) incorporated outcome measure(s) related to speech perception/intelligibility, auditory perception, musical perception, or communication improvement, and (5) integrated study design of randomized controlled trials, non-randomized controlled trials, cohort studies, and repeated measures (experiments with additional purposes) to report the results of pre- and post-training.

Titles and abstracts of the articles were screened according to the selection criteria and identified for preliminary articles as inclusion. Additional information was identified manually by two independent authors (NFAS and WH) who also checked any relevant articles that may not have been returned by the initial database search.

Data extraction

Two authors independently extracted data from each study following PICOS criteria [11]. Both children and adult participants who had hearing loss and have been fitted with either hearing aids or cochlear implants were included as the participants. In the intervention, all studies included the music training sorted by the stimuli used, frequency, duration of the training, and study settings of music training. All outcome measures related to musical perception after conducting the music training were collected. For comparison, we used pre- and post-training results to study the effectiveness of the music training. No restrictions were specified in terms of the duration of rehabilitation and follow-up. Finally, primary outcomes included one or more of the following: (1) detection of music perception including pitch, rhythm, frequency discrimination, and melody; (2) improvement in music perception including pitch, rhythm, frequency discrimination, and melody; (3) improvement in melodic contour identification that indicates tone recognition and speech perception; (4) improvement in the perception of recognition of musical instruments. Secondary outcomes comprised the improvement in musical performance, i.e., singing and music appreciation.

Risk of bias and quality of study

The methodological quality and risk of bias for the included studies were assessed using the standardized Physiotherapy Evidence Database (PEDro) scale. This scale's items assessed the (1) specific eligibility criteria, (2) randomization of the allocation of the subjects, (3) concealed allocation of subjects, (4) pretherapeutic interventions baseline, (5) blinding of all subjects, (6) blinding of therapists, (7) blinding of assessors who measured key outcomes of the study, (8) measure of at least one key outcome form 85% of the recruited subjects, (9) intention to treat analysis (mentions that all subjects received treatment or control conditions as allocated, (10) statistical comparison between groups for at least one key outcome, and (11) point measure for size of treatment effect and variability measure for at least one key outcome.

One point was added to the studies only if a criterion was clearly stated on literal reading. The point would not be added if the criteria were missing or not clearly stated. For criteria 4 and 7 through 11, key outcomes provided the primary measure of the effectiveness or lack of the effectiveness of the therapy. Based on the suggestion of Moseley et al. [12], studies scoring 9–10 on the PEDro scale were considered methodologically to be of "excellent" quality. Scores ranging from 6 to 8 were considered "good" quality, while studies scoring 4 or 5 were of "fair" quality, and studies scoring below 4 were considered "poor" quality.

Statistical analysis and publication bias

Data analysis was run using Comprehensive Meta-Analysis ver. 3 (Biostat Inc., Englewood, NJ, USA). Meta-analysis was performed using all studies that contained musical perception data having pre- and post-music training, appropriate outcome measures, and intervention. Means and standard deviation of pre- and post-training and also their correlation were used to calculate effect size of the music training.

Since most studies employed different measures outcome for musical perception, the effect sizes were calculated as standardized mean difference (SMD) in which it is necessary to standardize the result of all the studies to uniform scale before they can be combined; thus SMD expresses the size of the intervention effect in each study relative to the variability observed in that study [13]. All effect sizes were pooled using a random-effects model with 95% confidence intervals (CIs) [14]. A mixed-effects Q test for between-subgroup analysis of variance was used to compare the effects of four subgroups [14]. The funnel plot and Egger's regression asymmetry test were used to assess publication bias [15].

Heterogeneity analysis

Cochrane's Q and I^2 values were calculated to test for homogeneity of variance across the studies. The Q value represented the total amount of variance among the set of studies. I^2 was calculated using the formula, $I^2=100\%\cdot(Q–df)/Q$. According to Higgins and Altman [13], it provided a precise and easily interpreted measure of heterogeneity. I^2 values of 25%, 50%, and 75% represented low, medium, and high heterogeneity, respectively. A significant Q value indicated that the data were heterogeneous.

RESULTS

The search returned 9,021 articles, the titles of which were screened for relevance to the topic. After eliminating duplicate articles, a total of 4,092 articles remained. Subsequently, the following criteria were used to screen for eligibility. After first screening the titles and abstracts, 4,044 articles were excluded from the full-text assessment. A preliminary review of those titles narrowed down the potentially relevant articles to 48 journal articles, the full text of which could be accessed. The full texts of the remaining articles were then reviewed for inclusion based on the PICOS criteria and relevance. After applying our inclusion and exclusion criteria, 15 articles were left. Fig. 1 presents the selection process of the articles. Five studies were then excluded due to unsuitable outcome measures; thus, 10 full articles were included in this meta-analysis.

Characteristics of studies in the meta-analysis

Ten studies involving 186 participants met the PICOS inclusion criteria. The participants consisted of both adult and pediatric patients with hearing loss who had been fitted with hearing aids and cochlear implants. The age of the 101 adult participants who had consistently used hearing aids before post-lingual cochlear implantation [16-19] ranged from 18 to 88 years old, and the age of the 85 children ranged from 1 to 15 years old [20-25]. Among the participants, 89 were female and 76 were male; however, the sex of 21 participants in two studies by Fuller et al. [16] and Yucel et al. [23] was not specified.

The music training programs used in these studies consisted of various kinds of stimuli and musical programs. Fu et al. [22] and Yucel et al. [23] used musical tones as stimuli, while the training stimuli in two other studies consisted of musical instrument tones at low, mid, and high frequencies [17,24]. Three

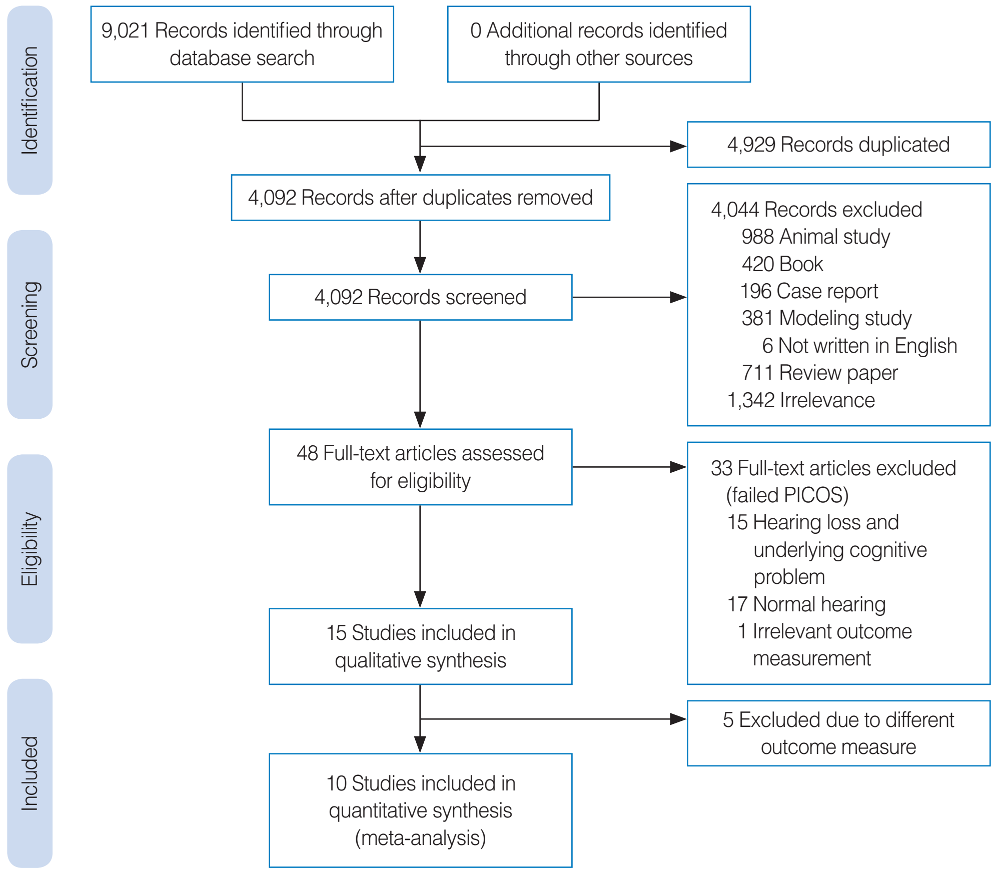


Fig. 1. Flow diagram of studies selected for the systematic review based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) criteria. PICOS, participants, interventions, comparisons, outcomes, and study design.

studies directly measured cochlear implant users' melodic perception using a melodic contour identification task [16,18,25], and three other studies conducted self-developed music training to enhance perceptual detection and to evaluate its improvement [19-21].

Most studies had a training period of 5 weeks to 3 months [16-18,22,25], while two studies [20,21] utilized training that lasted 3 months to 11 months, which were classified as an intermediate duration, and other two studies conducted training with a long duration (more than 12 months) [23,24]. Five studies were conducted in home settings [17,18,21-23] and the other studies implemented the training in rehabilitation centers [10,16,19,24,25]. The characteristics of the 10 studies are summarized in Table 2.

Overall effectiveness of music training

The outcomes of each study are also summarized in Table 2. In the pooled analysis (Fig. 2A), participants' musical perception was significantly higher after music training (SMD=2.092, 95% CI, 1.333–2.850, $P<0.001$). Although a funnel plot showed that the data were asymmetrical (Fig. 2B), the Egger regression test detected no publication bias in the studies (intercept=2.313; standard error, 1.376; $P=0.1312$). Due to high heterogeneity ($I^2=86.57$), we conducted a subgroup analysis.

Subgroup analysis

Four subgroup analyses were conducted to investigate the effects of age, the hearing device used, the participants' musical experience before the training, and music training duration on improvements in musical perception. Table 3 presents the effect sizes for subgroups, 95% CIs, and heterogeneity.

Age

To investigate the effects of age, the participants were divided into adults (age ≥18 years) and children (age <18 years). Fig. 3A presents a subgroup analysis of the effect of age on the outcome of musical perception after music training. A statistically significant subgroup effect was found ($P=0.013$), indicating that the effect of music training was different according to participants' age. The rehabilitation effect was greater for children than for adults. The pooled effect size estimated for adults (SMD= 1.118; 95% CI, 0.014–2.21) was notably lower than that of children (SMD=2.658; 95% CI, 1.640–3.676), implying that the treatment effect was stronger in children than in adults.

Table 2. Comparison of participants' characteristics and training outcomes in the 10 included studies

Study	Design	Participant			Intervention: music training		Study setting	Outcome	Finding
		Hearing loss and hearing device	No. of participants, age and sex	Control group	Stimuli	Frequency and duration			
Yucel et al. (2009) [23]	Randomized controlled trial	Profoundly hearing-impaired children Unilaterally implanted with cochlear implants; HiRes stimulation mode	n=18 (music group: 9) Age: not specified Age at implantation: 39–96 months Sex: NA	Normal hearing children: n=9 Age: not specified	An electronic keyboard (YAMAHA PSR-295) Listening to two pairs of notes Based on note discrimination	10 min/day every day for 2 years Evaluation after 1, 3, 6, 12, and 24 months (24 months)	Home training	Improvement of musical perception (pitch, rhythm, and melody)	Music group: Ling's six-sound detection ($P<0.05$) Word identification ($P<0.05$) Daily sentences ($P>0.05$) (not significant) Music stage questionnaire ($P<0.05$)
Di Nardo et al. (2015) [21]	Experimental study (repeated measure)	Bilateral hearing loss Mono-aural nucleus CI users	n=10 (6 boys, 4 girls) Age: 5–12 years Hearing age with CI, ±26 months	None	Auditory music training program (home-learning program) Frequency bands: 262–523 Hz, 523–1,046 Hz, and 1,046–1,976 Hz (used for most of the song, 36 notes)	6-Month training period At least 2 hours weekly (6 months)	Home training	Improvement in musical perception (frequency discrimination, pitch recognition, and appraisal)	Musical pitch discrimination test showed a significant improvement in musical perception after music training ($P=0.001$) Music test result (pre-and post-training: $P=0.015$) (melodic version: $P=0.007$)
Fu et al. (2015) [22]	Repeated measures	Children with congenital hearing loss Mandarin-speaking Had at least 2 years of experience with CI	n=14 (boys: 7 girls: 7) Age: 5.5–9.7 years (mean, 7.8 years) Mean CI usage: 4.9 years (2.3–7.7 years)	None	Five tones, three tones, five piano stimuli (different root notes used for training) 23 Root notes (250 ms with 50 ms of silence between each note) All stimuli presented at 70 dBA	Half-hour per day Every day for 10 weeks 2.5 months	Home training	MCI Tone recognition and speech perception	Mean performance improved for all outcome measures Five tones (mean improvement, 57.3 points; SE, 11.1) Three tones (mean improvement, 45.8 points; SE, 10.9) Five piano stimuli (mean improvement, 45.8 points; SE, 8.2)

(*Continued to the next page*)

Table 2. Continued

Study	Design	Participant			Intervention: music training		Study setting	Outcome	Finding
		Hearing loss and hearing device	No. of participants, age and sex	Control group	Stimuli	Frequency and duration			
Cheng et al. (2018) [25]	Non-randomized controlled trial	Mandarin-speaking CI children Prelingually deaf and diagnosed with severe-profound SNHL before 1 year old	CI group: n=16 (5 girls, 11 boys) Age: 1.7–6.1 years; mean, 6.3 years CI experience: 0.8–6.0 years; mean, 2.8 years	Normal group n=22 (11 girls, 11 boys) Age: 4.5–9.3 years; mean, 6.2 years	(1) MCI stimuli: nine melodic contours (rising, rising-flat, rising-falling, flat-rising, flat, flat-falling, falling-rising, falling-flat, falling) Five notes of equal duration (250 ms, 50 ms of silence between notes) (2) Lexical tone stimuli: 64 stimuli (4 tones× 4 monosyllables× 4 talkers)	15 min/session Three sessions per training day 5 day/wk for 8 weeks 2 months	Rehabilitation center	Music (MCI and tone recognition) and speech perception (sentence recognition)	Significant effect recorded for (1) MCI mean improvement: 22.0 (range, 5.7–47.2) (2) Tone recognition: 14.5 (range, 4.7–32.8) (3) Sentence recognition: 14.5 (range, 1.5–34.3)
Fuller et al. (2018) [16]	Randomized controlled trial	Dutch-speaking adults who were CI users Used CI more than 1 year Some participants used hearing aids in the contralateral ear.	(1) Pitch/timbre group (n=6); age: 56–73 years; CI experience: 5–11 years (2) Music group (n=6); age: 59–71 years; CI experience: 3–10 years	Control (non-music training), n=4 Age: 66–80 years CI experience: 4–6 years	(1) Pitch and timbre: MCI (five training sessions) MCI: one test Instrument identification/daily sound identification MCI (five training sessions) MCI (one test) (2) Music therapy Listening to music and emotional speech Listening to musical speech Singing Playing an instrument Improvising music Session questionnaire	2 hr/session (15-minute break) Weekly session for 6 weeks 1.5 months	Rehabilitation center	(1) Word identification (2) Sentence identification (both consisting of speech perception)	(1) Word identification: Timbre: $P<0.001$ Music: $P=0.708$ Overall: $P=0.005$ (significant) (2) Sentence identification: Timbre: $P=0.339$ Music: $P=0.328$ Overall: $P<0.05$ (significant)

(Continued to the next page)

Table 2. Continued

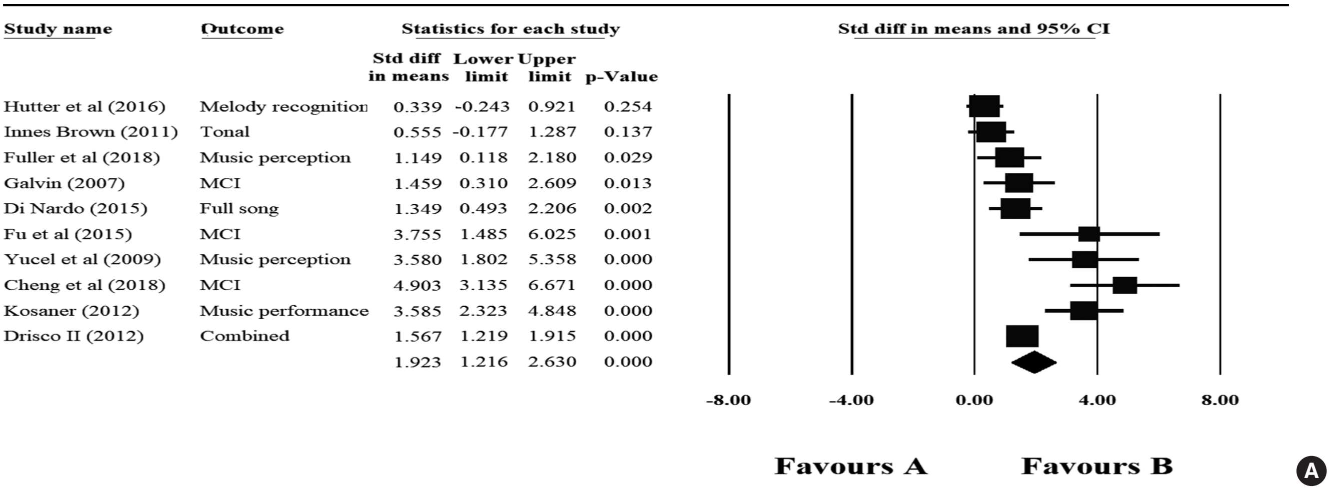
Study	Design	Participant			Intervention: music training		Study setting	Outcome	Finding
		Hearing loss and hearing device	No. of participants, age and sex	Control group	Stimuli	Frequency and duration			
Driscoll (2012) [17]	Randomized controlled trial	Post-lingual deaf adults aged >18 Read/understand written English Have access to a computer with internet and sound capabilities Used CI, HA, or both	n=71 (21 men, 50 women) Age: 26–88 years (mean, 62.59 years) Bilateral: 21, unilateral: 50 Divided into three groups	n=24, feedback on correct musical cues	Recording of solo performance of eight musical instruments: (1) Representing a range of low, middle, and high frequencies (2) Five melodies from each instrument	15 Sessions 10 min/session Three times/wk over 5 weeks 1.2 months	Home training	Improved recognition of musical instruments No significant improvement in timbre recognition	(1) Musical background questionnaire recognition test: Week 3: significant difference observed ($P<0.001$) (2) Significant improvement observed from week 3 to week 5 ($P=0.011$) (3) Individuals with bilateral CI scored significantly higher than those with unilateral CI ($P=0.02$)
Galvin et al. (2007) [18]	Non-randomized controlled trial	Adults with no musical experience CI users	n=6 (4 men, 2 women) Wearing CI Age: 26–75 years	n=9 Normal hearing	Melodic contour identification training (nine five-note melodic patterns)	4 subjects: 30 min/day; everyday (1 to 2 months) Two subjects: 3 hours per day, 1 week 2 months	Home training Rehabilitation center	Improvement in: MCI performance (vowel recognition) FMI test (music perception performance)	MCI performance (percent correct): 15.5%–45.4% improvement FMI performance: with rhythm cues: 9.1% improvement ($P=0.373$, not significant) Without rhythm cues: 20.8% improvement ($P=0.020$, significant)
Hutter et al. (2015) [19]	Experimental study (repeated measures)	Adults >18 years Post-lingual deafness, Unilaterally implanted CI users	n=12 (6 women, 6 men) Age: mean, 54 years	No control group	Five modules of music therapy: (1) Variability of voice and speech (2) Diverse components of music (3) Playfully used components of speech (4) Speech in diverse hearing surroundings (5) Complex hearing	10 Individualized sessions 50 min/session	Rehabilitation center	Hearing performance in musical parameters (1) Pitch discrimination (2) Melody recognition (3) Timbre identification	(1) Pitch discrimination: $P=0.027$ (no significant difference) (2) Melody recognition: $P<0.018$ (significant) (3) Timbre identification: $P=0.004$ (significant but only in unilateral CI users)

(*Continued to the next page*)

Table 2. Continued

Study	Design	Participant			Intervention: music training		Study setting	Outcome	Finding
		Hearing loss and hearing device	No. of participants, age and sex	Control group	Stimuli	Frequency and duration			
Kosaner et al. (2012) [24]	Experimental study (repeated measures)	Pediatric CI Unilaterally implanted CI users	n=25 Divided into three groups: Group A: n=12 (3 girls, 9 boys; mean age: 26 months) Group B: n=7 (5 girls, 2 boys; mean age: 43 months) Group C: n=6 (3 girls, 3 boys; mean age: 72 months)	No control group	Live or recorded music Each group: a set of six songs and six rhymes Tonal music with range pitch, timbre, intensity, and frequency Music associated with animals and actions and stories related to music was created.	Groups A and B participated with parents in one group session (45 minutes, one individual session of 20–30 min/wk for 18 months). Group C: one group and one individual session/wk for 3 months	Rehabilitation center	Improvement in performance of the musical component	Recognizing song, tunes, and timbre Responding to music and rhythm Singing Overall improvement Group A: $P<0.001$ Group B: $P<0.001$ Group C: $P=0.027$
Innes-Brown et al. (2013) [20]	Non-randomized controlled study	School-age children Using HA and CI	n=11 (4 girls, 7 boys) (CI: 6, HA: 5)	n=9 Normal hearing (5 girls, 4 boys)	The "music club": musical activities based on round play Session were divided into vocal play, physical music, and singing games. All games targeted rhythm, tempo, pitch, and timbre.	Conducted every week, 45 min/session	Rehabilitation center	Auditory perception (rhythmic, tonal, and timbre perception)	Rhythmic test: pre-post training: $P<0.01$ Tonal test: pre-post training: $P=0.04$ Timbre test: pre-post training: $P=0.01$

NA, not applicable; CI, cochlear implant; MCI, melodic contour identification; SE, standard error; SNHL, sensorineural hearing loss; HA, hearing aid; FMI, familiar melody identification.



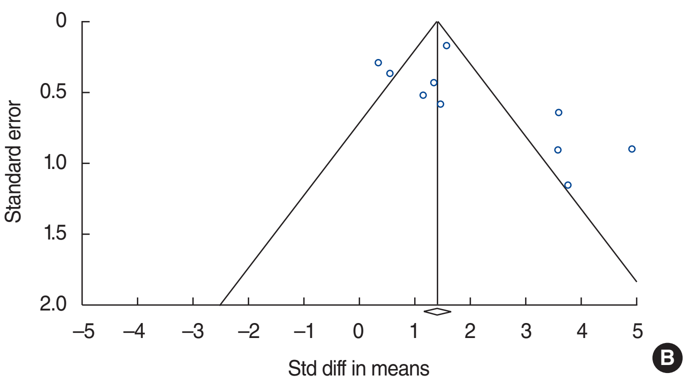


Fig. 2. Forest plot of a pooled analysis of all 10 included studies (A), and a funnel plot of standard errors by the standardized difference (std diff) in means yielded an asymmetrical graph, indicating potential publication bias (B). MCI, melodic contour identification; CI, confidence interval.

Table 3. Summary of the meta-analysis by effect size and heterogeneity, including four subgroup analyses

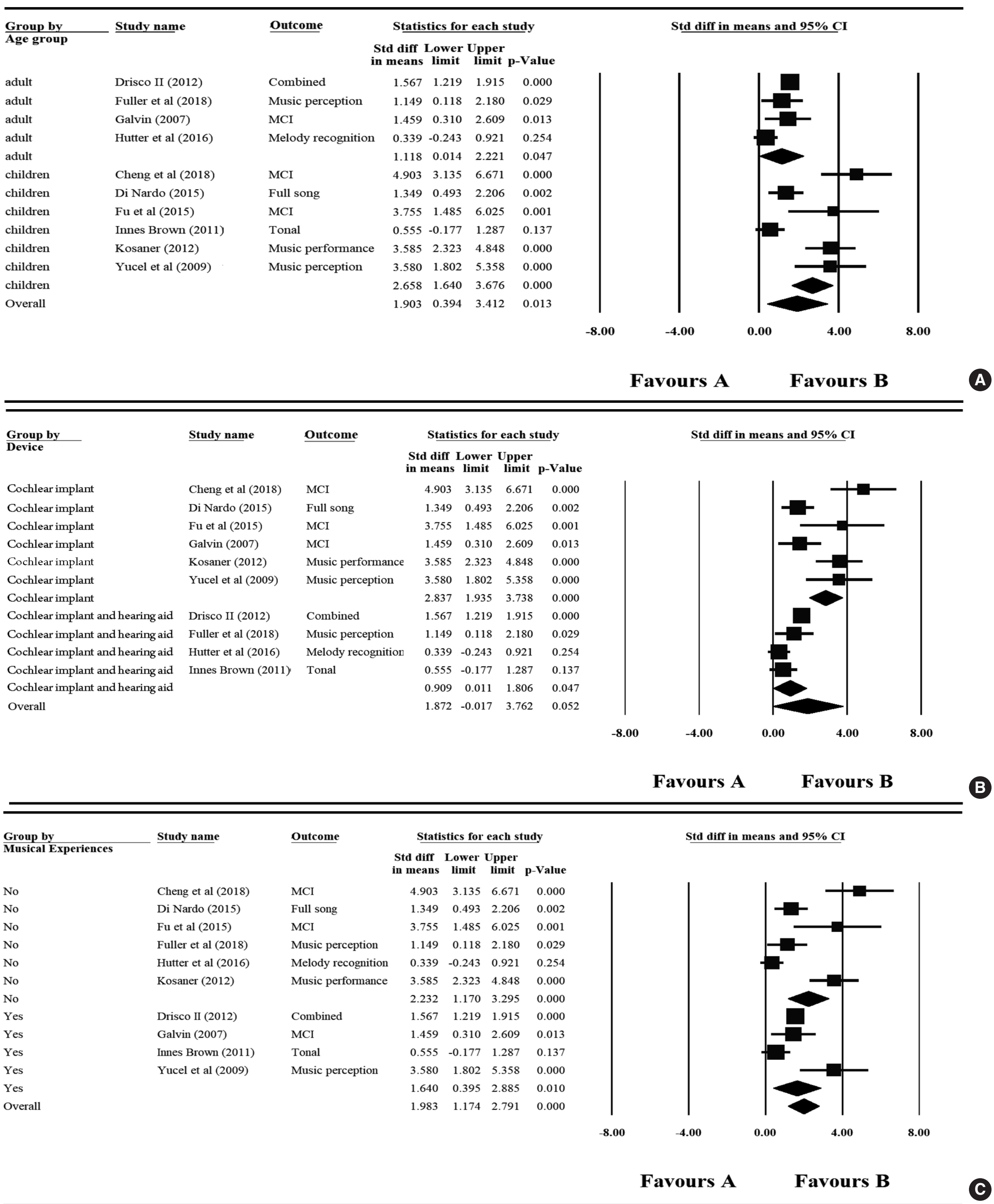
Characteristics	Study (n)	Effect size and 95% CI				Homogeneity test			
		SMD	Lower limit	Upper limit	*P*-value	Q	Df (Q)	*P*-value	I^2
Overall	10	2.092	1.333	2.850	<0.001	67.014	9	<0.001	86.570
Age		1.903	0.3412	3.412	0.013	25.523	5	0.090	39.420
Adult	4	1.118	0.014	2.221	0.047	12.769	3	0.005	16.506
Children	6	2.658	1.640	3.676	<0.001	38.370	5	<0.001	18.969
Device		1.862	0.550	3.174	0.005	45.610	7	<0.001	63.791
Cochlear implant only	6	2.452	1.450	3.453	<0.001	43.600	5	<0.001	58.532
Cochlear implant and hearing aid	4	1.101	–0.274	2.476	0.116	11.680	3	0.009	44.315
Musical experience		1.872	–0.017	3.762	0.052	51.523	9	0.003	83.791
Yes	4	0.909	0.011	1.806	0.047	15.510	3	0.001	77.774
No	6	2.837	1.935	3.738	<0.001	22.497	5	<0.001	80.658
Music training period		2.023	0.661	2.911	0.004	31.334	9	0.131	24.510
<3 mon	6	1.791	0.949	2.633	<0.001	33.074	5	<0.001	22.882
>3 to <12 mon	2	0.941	–0.392	2.275	0.167	1.906	1	0.467	21.536
≥12 mon	2	3.583	1.973	5.193	<0.001	0.000	1	0.996	0.000

CI, confidence interval; SMD, standardized mean difference.

Hearing devices

Hearing device users were divided into those with a cochlear implant only and those with a cochlear implant and hearing aid (i.e., bi-modal users). The pooled data for the hearing device subgroups are shown in Fig. 3B. Unfortunately, due to moderate to substantial heterogeneity between the trials within each of these subgroups and the relatively small number of participants, the results obtained might have failed to detect subgroup differences precisely. Regardless, the effect size for cochlear implant–only users was notably larger (SMD=2.232; 95% CI, 1.170–

Meta-Analysis



Group by Age group	Study name	Outcome	Std diff in means	Lower limit	Upper limit	p-Value
adult	Drisco II (2012)	Combined	1.567	1.219	1.915	0.000
adult	Fuller et al (2018)	Music perception	1.149	0.118	2.180	0.029
adult	Galvin (2007)	MCI	1.459	0.310	2.609	0.013
adult	Hutter et al (2016)	Melody recognition	0.339	-0.243	0.921	0.254
adult			1.118	0.014	2.221	0.047
children	Cheng et al (2018)	MCI	4.903	3.135	6.671	0.000
children	Di Nardo (2015)	Full song	1.349	0.493	2.206	0.002
children	Fu et al (2015)	MCI	3.755	1.485	6.025	0.001
children	Innes Brown (2011)	Tonal	0.555	-0.177	1.287	0.137
children	Kosaner (2012)	Music performance	3.585	2.323	4.848	0.000
children	Yucel et al (2009)	Music perception	3.580	1.802	5.358	0.000
children			2.658	1.640	3.676	0.000
Overall			1.903	0.394	3.412	0.013

Group by Device	Study name	Outcome	Std diff in means	Lower limit	Upper limit	p-Value
Cochlear implant	Cheng et al (2018)	MCI	4.903	3.135	6.671	0.000
Cochlear implant	Di Nardo (2015)	Full song	1.349	0.493	2.206	0.002
Cochlear implant	Fu et al (2015)	MCI	3.755	1.485	6.025	0.001
Cochlear implant	Galvin (2007)	MCI	1.459	0.310	2.609	0.013
Cochlear implant	Kosaner (2012)	Music performance	3.585	2.323	4.848	0.000
Cochlear implant	Yucel et al (2009)	Music perception	3.580	1.802	5.358	0.000
Cochlear implant			2.837	1.935	3.738	0.000
Cochlear implant and hearing aid	Drisco II (2012)	Combined	1.567	1.219	1.915	0.000
Cochlear implant and hearing aid	Fuller et al (2018)	Music perception	1.149	0.118	2.180	0.029
Cochlear implant and hearing aid	Hutter et al (2016)	Melody recognition	0.339	-0.243	0.921	0.254
Cochlear implant and hearing aid	Innes Brown (2011)	Tonal	0.555	-0.177	1.287	0.137
Cochlear implant and hearing aid			0.909	0.011	1.806	0.047
Overall			1.872	-0.017	3.762	0.052

Group by Musical Experiences	Study name	Outcome	Std diff in means	Lower limit	Upper limit	p-Value
No	Cheng et al (2018)	MCI	4.903	3.135	6.671	0.000
No	Di Nardo (2015)	Full song	1.349	0.493	2.206	0.002
No	Fu et al (2015)	MCI	3.755	1.485	6.025	0.001
No	Fuller et al (2018)	Music perception	1.149	0.118	2.180	0.029
No	Hutter et al (2016)	Melody recognition	0.339	-0.243	0.921	0.254
No	Kosaner (2012)	Music performance	3.585	2.323	4.848	0.000
No			2.232	1.170	3.295	0.000
Yes	Drisco II (2012)	Combined	1.567	1.219	1.915	0.000
Yes	Galvin (2007)	MCI	1.459	0.310	2.609	0.013
Yes	Innes Brown (2011)	Tonal	0.555	-0.177	1.287	0.137
Yes	Yucel et al (2009)	Music perception	3.580	1.802	5.358	0.000
Yes			1.640	0.395	2.885	0.010
Overall			1.983	1.174	2.791	0.000

Fig. 3. Subgroup analysis. Effect sizes according to age group (A), hearing device (B), previous musical experience (C). CI, confidence interval; std diff, standardized difference; MCI, melodic contour identification.

(*Continued to the next page*)

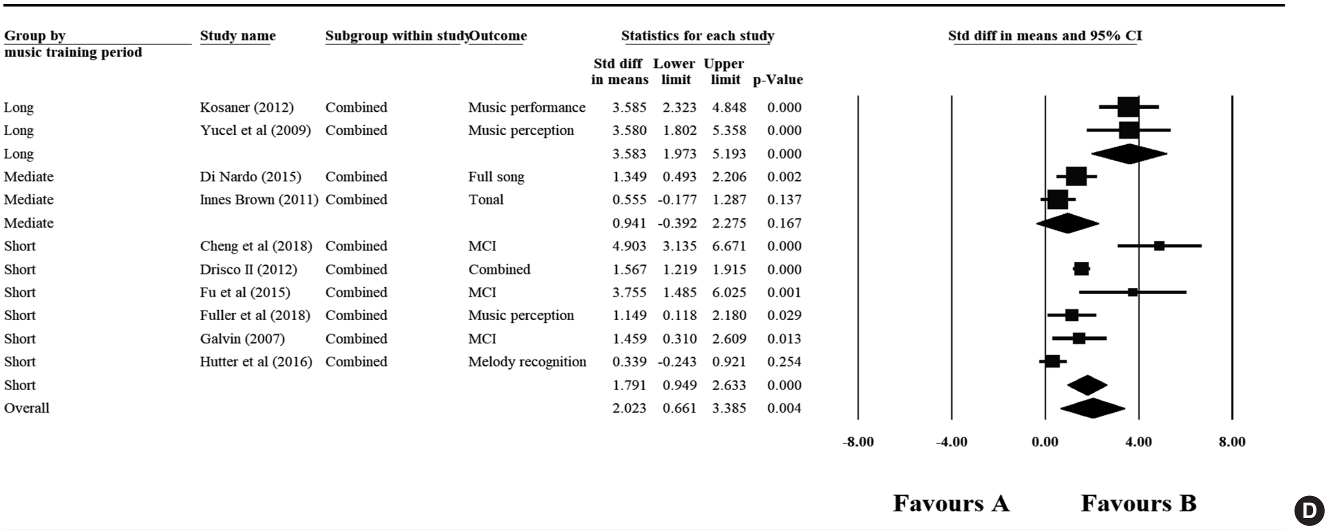


Fig. 3. (Continued) Subgroup analysis. Effect sizes according to the duration of music training (D). CI, confidence interval; std diff, standardized difference; MCI, melodic contour identification.

Table 4. Quality assessment based on the PEDro scale for the 10 included studies

Study	1	2	3	4	5	6	7	8	9	10	11	Total	Quality
Hutter et al. (2015) [19]	1	1	1	1	0	0	0	0	1	1	1	7/11	Good
Innes-Brown et al. (2013) [20]	0	0	1	0	1	0	0	1	1	1	1	6/11	Good
Kosaner et al. (2012) [24]	0	0	0	1	0	0	0	1	1	1	1	5/11	Fair
Galvin et al. (2007) [18]	0	1	1	0	0	0	0	1	0	1	1	5/11	Fair
Driscol (2012) [17]	1	1	1	0	0	0	0	1	0	1	1	6/11	Good
Fuller et al. (2018) [16]	1	1	0	1	1	0	0	1	1	1	1	8/11	Good
Cheng et al. (2018) [25]	1	0	1	1	0	0	0	1	1	1	1	7/11	Good
Fu et al. (2015) [22]	1	0	0	1	0	0	0	1	1	1	1	6/11	Good
Di Nardo et al. (2015) [21]	1	1	1	1	1	0	0	1	1	1	0	8/11	Good
Yucel et al. (2009) [23]	1	1	0	1	1	0	0	1	0	1	1	7/11	Good

Scale of item score: 0=absent; 1=present. The Physiotherapy Evidence Database (PEDro) scale criteria were (1) specific eligibility criteria; (2) random allocation; (3) concealed allocation; (4) similarity at baseline on key measures; (5) subject blinding; (6) therapist blinding; (7) assessor blinding; (8) >85% follow-up of at least 1 key outcome; (9) intention to treat analysis; (10) between-group statistical comparison for at least 1 key outcome; and (11) point estimates (size of treatment effect) and measures of variability provided for at least 1 key outcome. Studies scoring 9–10 on the PEDro scale were considered to be of "excellent" quality methodologically. Scores ranging from 6 to 8 were of "good" quality while studies scoring 4 or 5 were of "fair" quality, and studies scoring below 4 were considered "poor" quality.

3.295) than that for bi-modal users (SMD=1.640; 95% CI, 0.395–2.885), which indicates that a stronger treatment effect of music training was observed for cochlear implant–only users than for bi-modal users.

Previous musical experience

A subgroup analysis according to previous musical experience showed no statistically significant difference (P=0.052), indicating that participants' musical experience before music training did not affect the effectiveness of training (Fig. 3C). Nonetheless, the plausibility of this subgroup effect should be considered, as substantial heterogeneity was found among the trials within each of these subgroups (without musical experience: I^2=77.77%; with musical experience: I^2=80.66%).

Training period

The studies were divided according to three training periods (short, intermediate, and long). The training period showed a significant effect (P=0.004), with low heterogeneity (I^2=24.51%), indicating that training duration might significantly affect training effectiveness in terms of musical perception (Fig. 3D). A long duration of training showed notably stronger effects (SMD=3.583, 95% CI, 1.973–5.193) than a short duration of training (SMD=1.791, 95% CI, 0.949–2.633), which implies that long-duration music training is more effective as a treatment than short-duration training. Intermediate-duration training, lasting from 3 months to 11 months, had the smallest effect size (SMD=0.941, 95% CI, –0.392 to 2.275).

Risk of bias and quality of evidence assessment

Table 4 shows the quality scores for each question of the PEDro. Two authors independently analyzed the scores and then verified them on the official PEDro website. The overall mean PEDro score was 6.5. The quality of the articles ranged from fair to good, with the quality of two studies being assessed as fair [18,24] and the quality of the remaining studies classified as good [16,17,19-23,25] due to a lack of information about the random allocation of subjects, the blinding process, and whether the key outcome were analyzed by "intention-to-treat" or not. None of the studies provided information about the blinding of the therapists involved in therapy sessions (i.e., whether they were unable to distinguish between the treatments applied to different groups) or about the subject concealment.

DISCUSSION

Is music training effective as aural rehabilitation for hearing aid and cochlear implant users?

Music therapy can address several objectives of auditory training. Previous studies have suggested that music training can improve perception, localization, differentiation, and recognition of sound and attention towards the sound [5]. Furthermore, appropriate musical input is more effectively heard and assimilated than speech; thus, it is more likely to stimulate a natural motivation to use residual hearing [6]. The present study evaluated the efficacy of music training on musical perception among individuals with hearing loss who had been fitted with either hearing aids, cochlear implants, or both. The synthesis of data from these 10 studies suggested that significant improvements in musical perception were achieved in these individuals. Significant differences emerged according to age (between adults and children), the type of hearing device used, and the duration of training. However, nonsignificant differences in terms of improvements in musical perception were observed between participants with musical experience and those with no musical experience before starting music training.

Does music therapy affect adult and pediatric patients differently?

A significantly different effect of music training was found between children (below 18 years of age) and adults (18 years of age and older). The effect size for children was significantly larger than that for adults, suggesting that music training may provide greater benefits for children than for adults. From the perspective of neuroplasticity, it is logical that pediatric users of hearing aids and cochlear implants may benefit more from music training than adult users [6]. Chronological age has long been linked to neuroplasticity, with greater neuroplasticity associated with younger age and/or immaturity [26]. The capacity for synaptic plasticity, with consequences for learning and memory, is not constant throughout the lifespan and typically declines with age at variable rates [27]. It peaks relatively soon after birth, with some research indicating that infants' brain plasticity is about two times higher than that of adults. The effects of music training in children might therefore be stronger due to their greater brain plasticity. A clear differentiation of the effects of musical therapy across different age groups would help in the development and implementation of programs according to patients' needs. In addition to age, however, it would have been desirable to consider in this study whether individuals were affected by pre-lingual or post-lingual deafness. In previous studies, significantly different therapeutic effects have been found for these two groups [26]. However, due to limitations in the data that could be extracted, it was not possible to clearly subcategorize the participants into pre-lingual and post-lingual deafness groups. Future research comparing differences between individuals with pre-lingual and post-lingual deafness through high-quality randomized controlled trials is needed to confirm the therapeutic effectiveness of aural rehabilitation in these two groups.

Can the type of hearing assistive device influence the effects of music training?

The usage of different hearing assistive devices is known to be linked to differences in speech recognition performance by hearing-impaired individuals. Research by Gfeller et al. [28] found that bilateral cochlear implants provided a positive impact on the recognition of music with lyrics, whereas bi-modal users who were fitted with hearing aids and cochlear implants showed better perception and enjoyment of instrumental music. This finding provides natural support for the possibility that the perception of music is likely to be meaningfully improved by combining acoustic and electric stimulation.

Regardless, the current study found that the trainees who used only cochlear implants showed greater improvements in musical perception after music training than those who used both cochlear implants and hearing aids. One possible explanation for this seemingly contradictory finding is that most trainees who were only fitted with cochlear implants in this study were young children. As discussed above, the effects of music training differed between children and adults due to neuroplasticity. Therefore, in further research, music training should be applied to carefully differentiated subgroups among participants of the same age depending on the mode and type of devices.

Is previous musical experience a key factor for inducing positive results of music training?

A nonsignificant effect of previous musical experience on trainees' musical perception was found. However, the authors suggest that it may not be possible to draw a definitive conclusion for this subgroup because of the high level of heterogeneity that was present among the relevant studies. For example, the studies did not provide clear and specific information on how long and how often the trainee had musical experience and how intensive their experience was. Furthermore, the effects of music on the brain have been previously investigated by several researchers who compared the brain structure of musicians and non-musicians. For instance, in the study conducted by George and Coch [29], musicians demonstrated faster updating of auditory and visual working memory representations and more efficiently drew upon working memory resources to process deviant auditory stimuli than non-musicians with no musical experience.

The relationship between previous musical experience and the results of music training still need to be further explored through high-quality evidence-based studies, such as randomized controlled trials with a larger number of participants, as it remains possible that musical experience may have a significant effect on the outcomes of music training.

Is there a most effective duration of music training?

We analyzed the effectiveness of music training in terms of the training period. From the pooled results in this subgroup analysis, short and long training durations had a significant positive effect on improving participants' musical perception, whereas a non-significant effect was observed for the intermediate duration. From the perspective of audiological practice, we suggest a long training duration (e.g., 12 months or longer) to optimize the effectiveness of rehabilitation programs for hearing-impaired individuals. The effects of various training durations on the effectiveness of music training are expected to serve as a basis for the development of further music training programs to improve the auditory perception of hearing-impaired individuals with hearing devices.

Limitations of the study

The studies included in this review varied in terms of the severity of the disease, the type and duration of intervention, and the quality of the methodology; therefore, caution is needed when interpreting our findings. The outcome measure of musical perception was used to assess the efficacy of music training among individuals with hearing loss. Participants' listening abilities, duration of the training, and the content of training varied considerably across studies. This not only resulted in a limited ability to make direct comparisons but may also explain the inconsistent pattern of results that was observed across studies. Furthermore, because of the limited number of suitable articles that have been published, the number of participants was rather small and may not represent the entire population. Moreover, we only included studies that were published in English. In light of the small number of trials and participants and heterogeneity among studies, we still do not have enough evidence to confidently conclude whether there is a true subgroup effect for the type of device and musical experience. It is, therefore, useful to consider the plausibility of the demonstrated subgroup effects for these two groups. High heterogeneity is expected due to differences across studies, but it can nonetheless be useful to present the pooled data from comparable studies.

Future directions of auditory training with music

The findings of this systematic review regarding the effectiveness of music training in terms of improvements in musical perception can provide further directions for research in this field. We believe that the effectiveness of music training for the improvement of musical perception may be linked to the effectiveness of music training in aural rehabilitation and speech-language development because speech and music share many common properties. The auditory perception of speech and music involves the ability to distinguish between different sounds, their pitches, duration, intensities, and timbres, and their changes over time [6]. These properties enhance the listener's ability to interpret sounds and attach meaning to them. Furthermore, these commonalities between music and speech allow music and music therapy to provide an alternative and pleasurable tool to enhance traditional auditory training techniques. Specifically, based on the studies reviewed herein, patients' discrimination levels significantly improved through music training, and the practice that patients received in the setting of music generalized to environmental sounds.

Overall, music training can be implemented as an aural rehabilitation approach, as it effectively improves aural perception and musical perception in patients with hearing loss. In addition, it is important to consider that the effects of music training differ by the age of the trainee, the type of hearing device used, and the duration of music training. The finding of this study can serve as a reference for clinicians, patients, and health policymakers regarding the application of music training in clinical or rehabilitation programs. However, further high-quality randomized controlled trials are needed to confirm the effectiveness of aural rehabilitation in hearing-impaired patients.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

ACKNOWLEDGMENTS

This work was supported by a National Research Foundation of Korea (NRF) grant funded by the Korean government (MSIT) (2019R1F1A1053060).

ORCID

Nor Farawaheeda Ab Shukor
https://orcid.org/0000-0001-6964-2704

Jihyeon Lee https://orcid.org/0000-0003-0896-9062
Young Joon Seo https://orcid.org/0000-0002-2839-4676
Woojae Han https://orcid.org/0000-0003-1623-9676

AUTHOR CONTRIBUTIONS

Conceptualization: all authors. Data curation and formal analysis: NFAS, WH. Funding acquisition: WH. Methodology: NFAS, JL. Project administration: YJS, WH. Visualization: NFAS, JL. Writing–original draft: NFAS. Writing–review & editing: all authors.

REFERENCES

1. World Health Organization. WHO global estimates on prevalence of hearing loss. Prevention of Deafness [Internet]. Geneva: World Health Organization; 2018 [cited 2020 April 30]. Available from: https://www.who.int/pbd/deafness/estimates/en/.
2. Committee on Accessible and Affordable Hearing Health Care for Adults; Board on Health Sciences Policy; Health and Medicine Division; National Academies of Sciences, Engineering, and Medicine; Blazer DG, Domnitz S, Liverman CT. Hearing health care for adults: priorities for improving access and affordability. 2. Hearing loss: extent, impact, and research needs [Internet]. Washington (DC): National Academies Press; 2016 [cited 2020 April 30]. Available from: https://www.ncbi.nlm.nih.gov/books/NBK385309/.
3. Mielke M, Grunewald A, Bruck R. An assistive technology for hearing-impaired persons: analysis, requirements and architecture. Conf Proc IEEE Eng Med Biol Soc. 2013;2013:4702-5.
4. Gfeller K, Driscoll V, Kenworthy M, van Voorst T. Music therapy for preschool cochlear implant recipients. Music Ther Perspect. 2011 Jun;29(1):39-49.
5. Hull RH. Fourteen principles for providing effective aural rehabilitation. Hear J. 2005 Feb;58(2):28-30.
6. Gfeller K. Music-based training for pediatric CI recipients: a systematic analysis of published studies. Eur Ann Otorhinolaryngol Head Neck Dis. 2016 Jun;133 Suppl 1(Suppl 1):S50-6.
7. Anderson S, Kraus N. Auditory training: evidence for neural plasticity in older adults. Perspect Hear Hear Disord Res Res Diagn. 2013 May;17:37-57.
8. Asaridou SS, McQueen JM. Speech and music shape the listening brain: evidence for shared domain-general mechanisms. Front Psychol. 2013 Jun;4:321.
9. Kraus N, Skoe E, Parbery-Clark A, Ashley R. Experience-induced malleability in neural encoding of pitch, timbre, and timing. Ann N Y Acad Sci. 2009 Jul;1169:543-57.
10. Kraus N, Chandrasekaran B. Music training for the development of auditory skills. Nat Rev Neurosci. 2010 Aug;11(8):599-605.
11. Moher D, Liberati A, Tetzlaff J, Altman DG; PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. PLoS Med. 2009 Jul;6(7):e1000097.
12. Moseley AM, Herbert RD, Sherrington C, Maher CG. Evidence for physiotherapy practice: a survey of the Physiotherapy Evidence Database (PEDro). Aust J Physiother. 2002;48(1):43-9.
13. Higgins JP, Altman DG. Assessing risk of bias in included studies. In: Higgins JP, Green S, editors. Cochrane handbook for systematic reviews of interventions. Hoboken (NJ): Wiley; 2008. p. 187-241.
14. Borenstein M, Hedges LV, Higgins JP, Rothstein HR. Introduction to meta-analysis. 1st ed. Chichester: John Wiley & Sons; 2009.
15. Egger M, Davey Smith G, Schneider M, Minder C. Bias in meta-analysis detected by a simple, graphical test. BMJ. 1997 Sep;315(7109):629-34.
16. Fuller CD, Galvin JJ 3rd, Maat B, Baskent D, Free RH. Comparison of two music training approaches on music and speech perception in cochlear implant users. Trends Hear. 2018 Jan-Dec;22:2331216518765379.
17. Driscoll VD. The effects of training on recognition of musical instruments by adults with cochlear implants. Semin Hear. 2012 Nov;33(4):410-8.
18. Galvin JJ 3rd, Fu QJ, Nogaki G. Melodic contour identification by cochlear implant listeners. Ear Hear. 2007 Jun;28(3):302-19.
19. Hutter E, Argstatter H, Grapp M, Plinkert PK. Music therapy as specific and complementary training for adults after cochlear implantation: a pilot study. Cochlear Implants Int. 2015 Sep;16 Suppl 3:S13-21.
20. Innes-Brown H, Marozeau JP, Storey CM, Blamey PJ. Tone, rhythm, and timbre perception in school-age children using cochlear implants and hearing aids. J Am Acad Audiol. 2013 Oct;24(9):789-806.
21. Di Nardo W, Schinaia L, Anzivino R, De Corso E, Ciacciarelli A, Paludetti G. Musical training software for children with cochlear implants. Acta Otorhinolaryngol Ital. 2015 Oct;35(4):249-57.
22. Fu QJ, Galvin JJ 3rd, Wang X, Wu JL. Benefits of music training in mandarin-speaking pediatric cochlear implant users. J Speech Lang Hear Res. 2015 Feb;58(1):163-9.
23. Yucel E, Sennaroglu G, Belgin E. The family oriented musical training for children with cochlear implants: speech and musical perception results of two year follow-up. Int J Pediatr Otorhinolaryngol. 2009 Jul;73(7):1043-52.
24. Kosaner J, Kilinc A, Deniz M. Developing a music programme for preschool children with cochlear implants. Cochlear Implants Int. 2012 Nov;13(4):237-47.
25. Cheng X, Liu Y, Shu Y, Tao DD, Wang B, Yuan Y, et al. Music training can improve music and speech perception in pediatric mandarin-speaking cochlear implant users. Trends Hear. 2018 Jan-Dec;22:2331216518759214.
26. Dennis M, Spiegler BJ, Juranek JJ, Bigler ED, Snead OC, Fletcher JM. Age, plasticity, and homeostasis in childhood brain disorders. Neurosci Biobehav Rev. 2013 Dec;37(10 Pt 2):2760-73.
27. Kinugawa K, Schumm S, Pollina M, Depre M, Jungbluth C, Doulazmi M, et al. Aging-related episodic memory decline: are emotions the key? Front Behav Neurosci. 2013 Feb;7:2.
28. Gfeller K, Jiang D, Oleson JJ, Driscoll V, Knutson JF. Temporal stability of music perception and appraisal scores of adult cochlear implant recipients. J Am Acad Audiol. 2010 Jan;21(1):28-34.
29. George EM, Coch D. Music training and working memory: an ERP study. Neuropsychologia. 2011 Apr;49(5):1083-94.