

Review

Effects of music therapy on functional ability in people with cerebral palsy: a systematic review

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Abstract

Objective: This review aimed to investigate the effects of music therapy on functional ability in people with cerebral palsy.

Materials and Methods: An electronic search of the CENTRAL, MEDLINE, and EMBASE databases was conducted. Randomized controlled trials that examined the effects of music therapy in patients with cerebral palsy were included.

Results: Eight trials were eligible for inclusion in this study. We found a low risk of bias in random sequence generation and allocation concealment in all trials. The risk of bias in blinding of the outcome assessment was low in all studies. We found that music therapy had a significant effect on the Gross Motor Function Measure score (standardized mean difference [SMD] -0.42), Functional Independence Measure for Children score (SMD 0.38), and Goal Attainment Scale score (SMD -1.43). Music therapy had no significant effect on any of the other items.

Conclusion: There is limited evidence that music therapy improves gross motor function and activities of daily living in patients with cerebral palsy. However, this was insufficient to allow for generalizable conclusions. Further studies with larger sample sizes are required to confirm the effects of music therapy in this population.

Key words: music therapy, cerebral palsy, systematic review, disability, physiotherapy

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Introduction

Cerebral palsy (CP) comprises a heterogeneous group of early onset, non-progressive, neurodevelopmental disorders caused by an insult to the developing brain, most often before birth. CP is a leading cause of disability in children, with a prevalence of approximately 2 per 1,000 live births¹⁾. Globally, an estimated 17 million people living with CP¹⁾. In general, CP causes impaired movement associated with abnormal reflexes, floppiness or rigidity of the limbs and trunk, abnormal posture, involuntary movements, unsteady

gait, or a combination of these. The effect of CP on function varies widely. Some affected individuals can walk, whereas others require assistance.

The use of music in neurorehabilitation is grounded in neurophysiological theory and research on the influence of music on cognitive processes and motor learning principles and has been applied to neurological disorders, including CP^{2, 3)}. The therapeutic approach called neurologic music therapy (NMT) was established 20 years ago in the US and is known to be effective in neurorehabilitation⁴⁾. Conceptually, NMT is based on three main sensorimotor techniques, with the improvement of motor skills as the overall goal. The first technique is rhythmic auditory stimulation (RAS), which aims to develop and maintain physiological rhythmic motor activity (gait) using rhythmic auditory cues. The second is patterned sensory enhancement (PSE), the objective of which is to facilitate movements associated with the activities of daily living. PSE is not necessarily rhythmic in nature and uses complex elements of music, such as pitch, dynamics, harmony, meter, and rhythm, to enhance and organize movement patterns in time and space and to improve activity, muscle coordination, strength, balance, postural

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control, and range of motion⁵). The third technique is therapeutic instrumental music performance, in which playing a musical instrument is used as an orientation training task to simulate and facilitate functional movement. This technique most commonly uses percussion instruments played in a traditional or non-traditional manner to improve range of motion, limb coordination, postural control, dexterity, body perception, and sensation^{6, 7}). NMT has been shown to improve walking performance in patients with neurological diseases, and musical training with instruments allows interrelationships between movement, emotions, and cognition for task-based learning to improve motor control^{8–10}).

Several clinical trials have examined the use of music therapy in patients with CP. One randomized controlled trial investigated the effects of a combination of PSE music and resistance exercise in children with spastic diplegia and found that those who exercised with PSE music showed statistically significant improvements in gross motor capacity compared with controls who exercised without music, and that these effects lasted at least 3 months⁸). However, there have been no systematic reviews of the findings of available studies on the effect of music therapy in the CP population. It remains unclear whether music therapy is useful in patients with CP. Moreover, studies on music therapy tend to include small sample sizes. Therefore, a systematic review of trials is required to evaluate the effectiveness of music therapy. This study aimed to investigate the effect of music therapy on the functional ability of people with CP.

Materials and Methods

Search strategy

We searched the MEDLINE, Cochrane Central Register of Controlled Trials (CENTRAL), and EMBASE databases from January 1980 to December 2020. The review protocol, including the search strategy, was registered before starting the review¹¹). We also searched the reference lists of included studies to identify additional relevant trials.

Eligibility criteria

Participants

CP describes a group of permanent disorders of movement and posture that limit activity, and are attributed to non-progressive disturbances in the developing fetal or infant brain. Motor disorders in CP are often accompanied by disturbances in sensation, perception, cognition, communication, behavior, epilepsy, and secondary musculoskeletal problems.

Interventions

Music therapy is an evidence- and art-based discipline that uses musical experiences within a therapeutic relationship to address clients' physical, emotional, cognitive, and

social needs¹²). Music therapy includes the following interventions: listening and moving to live, improvised, or pre-recorded music, as well as RAS; therapeutic use of music and rhythm as a driving force to facilitate better quality of movement and PSE; performing or creating music using an instrument and therapeutic instrumental music performance; improvising music spontaneously using voice, instruments, or both; singing or vocal activities set to music; composing music; and music combined with other modalities.

Comparator

No intervention, usual care, or alternative intervention.

Study design

Randomized controlled trials, quasi-randomized trials, and cluster randomized trials were included. The Crossover trials were excluded.

Outcomes

The main outcome was gross motor function, which was assessed using the gross motor function measure (GMFM), energy expenditure index, gross motor performance measure (GMPM), or simple motor test for Cerebral Palsy.

Additional outcomes were as follows: muscle strength, assessed by grip strength or the Medical Research Council scale score; spasticity, assessed by the Modified Ashworth Scale or (Modified) Tardieu Scale score; respiratory function, evaluated by vital capacity or forced expiratory volume in 1 s; physical fitness, assessed by the 6-min walk test and cardiopulmonary exercise testing; lower limb function, measured by gait velocity (cm/s) and the Timed Up and Go test; upper limb function, assessed by the ABILHAND-kids Scale, Shriners Hospitals for Children Upper Extremity Evaluation, Melbourne Assessment of Unilateral Upper Limb Function, Assisting Hand Assessment, or Quality of Upper Extremity Skills Test (QUEST); activities of daily living, measured by the Functional Independence Measure for Children (WeeFIM), Pediatric Evaluation of Disability Inventory (PEDI), Goal Attainment Scaling (GAS) score, Canadian Occupational Performance Measure, Assessment of Motor and Process Skills, or School Function Assessment; participation, assessed by the Participation Survey/Mobility tool; quality of life, assessed by the Pediatric Outcomes Data Collection Instrument; and secondary complications. We explored the outcomes at the end of treatment and at the end of the scheduled follow-up.

Data extraction and quality assessment

Two researchers (SY and TY) independently selected the reviews included in the analysis. One researcher (SY) extracted the data and a second researcher (TY) independently checked the data extraction forms for accuracy and

completeness. Any discrepancy was resolved by discussion, with the final decision made by a third investigator (RM). Two authors independently assessed the risk of bias using the Cochrane risk of bias' tool¹³. Any disagreements concerning the risk of bias or quality of evidence in the included studies were resolved by discussion.

Statistical analysis

We performed a quantitative synthesis of the findings of the included studies and a summary of the effects of intervention for each study by calculating the risk ratio (RR) for dichotomous outcomes and standardized mean difference (SMD) for continuous outcomes. We anticipated that there would be a limited scope for meta-analysis because of the range of different outcomes measured across a small number of existing trials. However, when studies had used the same intervention, comparator, and outcome measure, we pooled the results using a random effects meta-analysis with SMDs for continuous outcomes and RRs for binary outcomes and calculated the 95% confidence intervals and two-sided *P*-values for each outcome. Heterogeneity in the measures of effect between the studies was assessed using the χ^2 test and *I*² statistic. An *I*² value >60% indicated substantial heterogeneity.

Results

After screening the 314 records, 25 potentially relevant studies were identified. Eight of these 25 studies met the

study inclusion criteria^{8, 14–20} (Figure 1). The details of each study are presented in Table 1.

Study characteristics

Two of the eligible studies included patients with a Gross Motor Function Classification System score of I–IV^{8, 14, 15}. None of the studies included participants with conditions other than CP. In almost all studies, the participants were under 20 years of age. The sample sizes ranged from 11¹⁴ to 120¹⁵. Three studies used RAS provided by music therapists^{14, 18, 20}. The intervention involved listening to music in two studies^{15, 19}, a music therapist using PSE in two studies^{8, 17}, and a patient playing an instrument in one study¹⁶.

Almost all studies included some form of gross motor function as an outcome^{8, 14–20}. Three studies included limb function measures^{14, 16, 20}, three included activities of daily living^{8, 15, 19}, one study included WeeFIM¹⁵, another included PEDI⁸, and another included the GAS T-score as an outcome¹⁹. Only one study assessed upper limb function (using QUEST). Three studies included lower limb function as an outcome^{8, 18, 20}, three studies included velocity, two included cadence^{18, 19}, and one study included grip strength¹⁹.

Study quality

Six trials included the appropriate sequence generation (Table 2). Two studies that did not report the methods used for random sequence generation were classified as unclear^{16, 18}. Three studies did not report allocation concealment^{16–18}. Fifty percent of the trials included blinding of

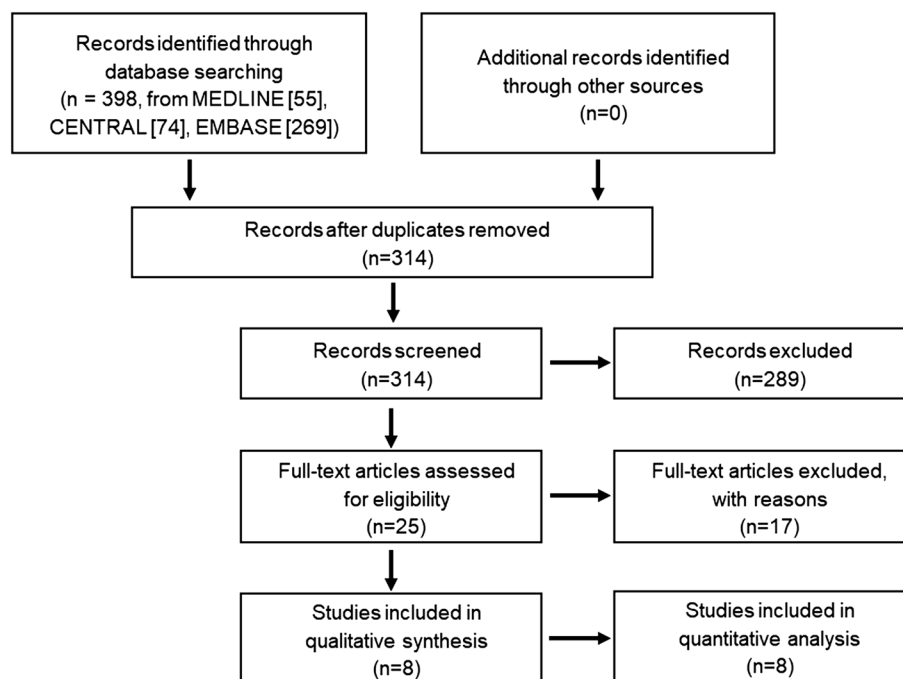


Figure 1 Study flow diagram.

Table 1 Characteristics of included studies

Study	Country	Participant characteristics	Number (I/C)	Intervention group	Control group	Outcomes
López-Ortiz 2016 ¹⁴⁾	USA	GMFCS score of II–IV Age 6–15 years Ability to follow two-step directions Medically stable No history of surgery or seizures within past 6 months	11 (5/6)	TDC for 1 h three times weekly for 4 weeks + physical therapy and occupational therapy	Physical and occupational therapy	Changes in clinical balance Upper limb function
Duymaz 2018 ¹⁵⁾	Turkey	Age 5–11 years Diagnosis of spastic-type CP Able to understand and cooperate with commands Not using technical or mobility aids No hearing problems GMFCS level I–III	120 (60/60)	NDT while listening to a classical music disc for 45 minutes	NDT only	GMFM-88 WeeFIM FACES
Wang 2013 ⁸⁾	Taiwan	Age 5–13 years Spastic diplegia, GMFCS score I–III Able to stand independently without falling Able to follow and cooperate with verbal instructions Parental commitment to supervise the training program without altering current therapy or activities	36 (18/18)	PSE music combined with sit-to-stand exercise	Exercise with no music	GMFM dimensions D and E Daily mobility and self-care functions Functional strength Walking speed
Yu 2009 ¹⁶⁾	China	Diagnosis of CP Age <14 years No severe organic disease, acute or chronic infection or coagulopathy, severe visual or hearing impairment, or progressive brain disease (brain tumor, moyamoya disease, etc.)	60 (30/30)	Playing a musical instrument for children	Needle therapy	Scoring for comprehensive functions Scoring for gross motor function
Teixeira–Machado 2017 ¹⁷⁾	USA	Diagnosis of CP Age 15–29 years Increased muscle tone No physical activity during the study protocol No cardiopathy or neoplasia	26 (13/13)	Global range of motion with coordinated and rhythmic dynamic floor exercises Motor coordination Body image Skill and agility	Traditional kinesiotherapy exercises	FIM WHODAS GMFCS
Kwak 2007 ¹⁸⁾	USA	Spastic-type CP Age 6–20 years	18 (9/9)	Conventional gait training enhanced by RAS provided by both a physical therapist and a music therapist	Conventional gait training by a physical therapist while a music therapist observed	Neurological damage How RAS could affect gait training
Ben–Pazi 2018 ¹⁹⁾	USA	Age 2–18 years Hypertonia interfering with daily functions Ability to use headphones for at least 10 min	18 (9/9)	Exposure to audio stimulation for at least 10 min; each session lasting preferably for 30 min four times a week	Music alone	CCHQ GAS GMFM-88 QUEST
Kim 2012 ²⁰⁾	Korea	No discernible hearing deficit Able to walk at least 10 m without a walking aid or a helper Able to understand the command to walk following rhythmic auditory stimulation	28 (15/13)	Rhythmic auditory stimulation using a combination of a metronome beat set to the individual's cadence and rhythmic cueing from a live keyboard	NDT	Gait pathology Kinematic data for the pelvis, hip joint, knee, ankle, and foot

C: control; CCHQ: Care and Comfort Hypertonicity Questionnaire; CP: cerebral palsy; FACES: Wong-Baker Faces Pain Rating Scale; FIM: Functional Independence Measure; GAS: Goal Attainment Scale; GMFM: Gross Motor Function Measurement; GMFCS: Gross Motor Function Classification System; I: intervention; NDT: neurodevelopmental treatment; PSE: patterned sensory enhancement; QUEST: Quality of Upper Extremity Skills Test; RAS: rhythmic auditory stimulation; TDC: Training Dance Control; WHODAS: World Health Organization Disability Assessment Schedule.

Table 2 Risk of bias summary

López–Ortiz 2016 ¹⁴⁾	Duymaz 2018 ¹⁵⁾	Wang 2013 ⁸⁾	Yu 2009 ¹⁶⁾	Teixeira–Machado 2017 ¹⁷⁾	Kwak 2007 ¹⁸⁾	Ben–Pazi 2018 ¹⁹⁾	Kim 2012 ²⁰⁾	
Low	Low	Low	Unclear	Low	Unclear	Low	Low	Random sequence generation
Low	Low	Low	Unclear	Unclear	Unclear	Low	Low	Allocation concealment
Low	Low	Low	High	High	High	Low	High	Blinding of participants and personnel
Low	Unclear	Low	Unclear	Unclear	Unclear	Low	Low	Blinding of outcome assessment
Low	Low	Low	Low	Low	Low	Low	Low	Incomplete outcomes data
Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Low	Unclear	Selective reporting
Low	Low	Low	Low	Low	Low	Low	Low	Other bias

Table 3 Summary of results of meta-analysis for outcomes

Outcome	Studies, n	Participants (intervention/control), n	SMD (95% CI)	Inconsistency value I^2 (%)
QUEST	1	18 (9/9)	-0.07 (-0.99, 0.86) $P=0.87$	-
WeeFIM	1	120 (60/60)	0.38 (0.01, 0.74) $P=0.04$	-
WeeFIM (after follow-up)	1	120 (60/60)	0.39 (0.03, 0.75) $P=0.04$	-
GMFM	3	174 (87/87)	0.42 (0.12, 0.72) $P=0.01$	0
GMFM (after follow-up)	2	156 (78/78)	0.58 (0.26, 0.90) $P=0.0004$	42
Velocity	3	82 (42/40)	0.29 (-0.16, 0.74) $P=0.21$	71
Velocity (after follow-up)	1	36 (18/18)	0.17 (-0.49, 0.82) $P=0.62$	-
Cadence	2	46 (24/22)	0.20 (-0.39, 0.79) $P=0.51$	40
PEDI	1	36 (18/18)	0.05 (-0.61, 0.70) $P=0.89$	-
PEDI (after follow-up)	1	36 (18/18)	0.14 (-0.52, 0.79) $P=0.68$	-
Grasp	1	18 (9/9)	0.08 (-0.84, 1.01) $P=0.86$	-
GAS T-score	1	18 (9/9)	-1.43 (-2.49, -0.36) $P=0.01$	-

CI: confidence interval; GAS: Goal Attainment Scale; GMFM: Gross Motor Function Measurement; PEDI: Pediatric Evaluation of Disability Inventory; QUEST: Quality of Upper Extremity Skills Test; SMD: standardized mean difference; WeeFIM: Functional Independence Measure for Children.

both study participants and observers, and were judged to have a high risk of bias^{16–18, 20}. Half of the studies did not include blinding of the outcome assessors^{15–18}. Incomplete outcome data bias was judged to be low in all studies. Study protocols were not available for any of the included studies. No other potential sources of bias were identified in the available data. Outcome data were missing for three studies and could not be obtained. Therefore, only five studies were included in the quantitative analysis (Table 3).

Effectiveness

Gross motor function

Three trials (including 174 participants) included GMFM data^{8, 15–19}. Meta-analysis showed that music therapy had a significant effect on the GMFM score (SMD, -0.42; 95% CI, -0.12, 0.72; $P=0.01$; $I^2=0\%$).

Activities of daily living

Only one trial (120 participants) reported the WeeFIM data¹⁵. It was found that music therapy had a significantly positive effect on the WeeFIM score (SMD, 0.38; 95% CI

0.01, 0.74; $P=0.04$). Only one trial (with 36 participants) assessed the PEDI⁸, and no significant effect of music therapy was found (SMD 0.05; 95% CI -0.61, 0.7; $P=0.89$). Another trial (with 18 participants) assessed the GAS T-score¹⁹ and found a significant effect of music therapy (SMD, -1.43; 95% CI, -2.49, -0.36; $P=0.01$).

Upper limb function

One trial (with 18 participants) assessed upper limb function using QUEST¹⁹ and did not find any significant effect of music therapy (SMD, -0.07; 95% CI, -0.99, 0.86; $P=0.87$).

Lower limb function

Three trials (including 82 participants) reported data on velocity^{8, 18, 20}. Meta-analysis showed that music therapy had no significant effect on walking velocity (SMD: 0.29; 95% CI: -0.16, 0.74; $P=0.21$; $I^2=71\%$). Two trials (including 46 participants) included data on cadence^{18, 20} and found no significant effect of music therapy (SMD 0.20; 95% CI -0.39, 0.79; $P=0.51$; $I^2=40\%$).

Muscle strength

Only one trial (with 18 participants) investigated grip strength¹⁹ and found that music therapy had no significant positive effect (SMD 0.08; 95% CI -0.84, 1.01; $P=0.86$).

Discussion

Eight trials (with 317 participants) were eligible for inclusion in this review on the effects of music therapy in patients with CP. We found a low risk of bias for random sequence generation and allocation concealment in all the trials. However, the risk of bias in the blinding of the study participants was high in half of the studies. However, the risk of bias in the blinding of the outcome assessment was low in all studies. This meta-analysis found that music therapy had a significant effect on WeeFIM and GMFM scores and on the GAS T-score but not on any other items in participants with CP. No adverse events were observed.

In terms of study heterogeneity, the music therapy content varied widely, and one study¹⁷ included adults with CP. The sample sizes in the included studies were small, ranging from 11 to 120 participants, which may have resulted in inadequate statistical power to detect the significant effects of music therapy in the CP population.

We did not find a high risk of selective reporting, other biases, or any major problems with the overall certainty of the evidence. However, due to insufficient sample size, the results of this meta-analysis may come into question when the results of larger-scale studies become available in the future.

To the best of our knowledge, there has been no comprehensive review of the effects of music therapy in patients with CP. We found only one limited systematic review of the effects of music therapy in neurorehabilitation of children and adults with CP²⁰. This review showed that dance and RAS have a potentially positive impact on body function, emotional expression, social participation, and attitudinal change. However, only RAS and dance have been investigated, and no meta-analysis has been performed.

Although music therapy for CP is used in actual clinical practice, the main reason for the lack of randomized controlled trials is probably the lack of researchers study-

ing both CP and music therapy. However, research on music therapy has been increasing in recent years²², future scholars are focusing on the clinical significance of music therapy. In the rehabilitation of children with CP, music therapy is an alternative tool for making the process more enjoyable and improve exercise adherence²³. Further studies are needed to investigate the effects of music therapy on adherence to exercise in patients with CP. In addition, music therapy does not require special equipment or skills, which makes it easy to introduce in rural areas where human medical resources are limited. The recent pandemic of coronary infections has made face-to-face interventions more difficult, and music therapy using virtual reality (VR) technologies has been implemented²⁴. Since virtual reality technology can intervene remotely, we believe that it can easily be utilized in rural areas.

Finally, despite our literature search strategy, the possibility of a selection bias cannot be excluded. This review did not include unpublished data, articles in the press, studies published in languages other than English, or studies for which only an abstract has been published.

Conclusions

We found limited evidence that music therapy could improve gross motor function and activities of daily living in the CP population. However, we could not find sufficient data to allow generalizable conclusions regarding the effects of music therapy in this population. No randomized controlled trials have used singing or vocal activities in music therapy for CP. Furthermore, there have been no randomized controlled trials on music therapy for spasticity, physical fitness, participation restrictions, or quality of life as outcomes of CP. Future studies with larger sample sizes are needed to determine the effects of music therapy in patients with CP.

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