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ORIGINAL ARTICLE

Neurologic music therapy in upper-limb rehabilitation in children with severe bilateral cerebral palsy: a randomized controlled trial

Eugenio MARRADES-CABALLERO [1], Clara S. SANTONJA-MEDINA [2], Jose M. SANZ-MENGIBAR [3, 4], Fernando SANTONJA-MEDINA [5, 6] *

[1]Superior Music Conservatory, Valencia, Spain; [2]Professional Music Conservatory, Carcaixent, Valencia, Spain; [3]Clinic of Rehabilitation, Madrid, Spain; [4]Balancephysio Vojta Therapy, London, UK; [5]Faculty of Medicine, University of Murcia, Murcia, Spain; [6]Department of Traumatology, V. de la Arrixaca University Hospital, Murcia, Spain

*Corresponding author: Fernando Santonja Medina, Calle Francisco Jimenez Ruiz 27, Murcia, Spain. E-mail: fernando@santonjatrauma.es

ABSTRACT

BACKGROUND: After receiving neurologic music therapy, functional improvements in children with severe bilateral cerebral palsy have not been found in the literature. Musical training with instruments allows interrelationships between movement, emotions and cognition for task-based learning, in order to improve motor control.
AIM: To understand whether neurologic music therapy has an impact on the functionality of children with severe cerebral palsy.
DESIGN: A randomized controlled assessor-blind trial was carried out.
SETTING: Children were recruited and treated in their own community center.
POPULATION: Eighteen children with severe bilateral cerebral palsy between 4 and 16 years old were studied.
METHODS: The intervention group (N.=18) received music therapy for 16 weeks, in addition to its usual physiotherapy input. Two music therapists implemented a neurologic music therapy program of therapeutic instrumental music performance. The control group (N.=9) received its usual therapeutic input, similar to the intervention group, but not neurologic music therapy. Overall and specific "Chailey levels of Ability" were quantified, as well as the locomotor stages.
RESULTS: Significant improvements in the overall and specific "arm and hand position" as well as "activities" from the Chailey Levels of Ability and the locomotor stages were observed ($P<0.05$) in the group which received the music therapy. All these improvements persisted after 4 months. The control group showed no improvements after a four-month follow-up.
CONCLUSIONS: Optimized intervention of neurologic music therapy can improve the functionality of children with severe bilateral cerebral palsy.
CLINICAL REHABILITATION IMPACT: Music therapy is a useful tool in rehabilitation and its positive effects remain four months after completing the treatment.

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KEY WORDS: Cerebral palsy - Rehabilitation - Upper extremity - Music therapy.

Neurologic music therapy (NMT)[1] has been successfully used in children with cerebral palsy (CP) with Gross Motor Function Classification System (GMFCS) levels I, II and III. Improvements in gait using Rhythmic Auditory Stimulation[2] and Resistance Exercise Effects through Patterned Sensory Enhancement[3, 4] have been found in the literature. Also, positive effects in neuroplasticity by active training with musical instruments[5, 6] have been observed due to the consistent bidirectional transmission of information between sensory and motor areas of the brain of people with CP. NMT active techniques have been used for therapeutic purposes,[7] however, research on people with poor gross motor levels (GMFCS-E&R= IV-V)[8] and severely limited manual ability (MACS=IV-V),[9] has not been previously published. Therapeutic intervention is complex in these patients due to severe communicative, cognitive and psychological difficulties.[10]

Music connects motor and cognitive aspects, facilitates learning and provides emotional experiences.[11, 12] These processes are used in clinical intervention in neurorehabilitation through therapeutic instrumental music performance (TIMP).[1] Promoting neuroplasticity by motor control learning and functional recovery[13] is possibly due to the effect of hearing and visual feedback.[14]

The rhythm produced with musical instruments acts as a force that optimizes all aspects of motor control through neurophysiological processes such as priming and entrainment[15, 16] by synchronization of movement, modulating muscle activation patterns, and controlling movement in space.[1] Specificity and intensity (repetition, level of difficulty) of the required intervention in order to optimize neurorehabilitation[17-19] are obtained by modifying the different musical features. For instance, music activities can develop communication skills[20] and self-expression mechanisms,[21] target regions where volition and motivation originate,[22] and improve movement planning and coordination.[23]

Our hypothesis was that TIMP administered by two musical therapists using interactive activities is a task-specific functional training that optimized learning and motor control in children with severe bilateral CP.

This study aimed to explore the improvements in gross motor and upper-limb function in patients with severe bilateral CP (GMFCS=IV-V) using TIMP.

Materials and methods

Study design

This analytic experimental study was designed as a controlled assessor-blind cross-over trial. Patients were randomized into two groups by the recruiting center team who did not participate in the research. The study was approved by the Research Ethics Committee of the University of Murcia ID: 1186/2015.

Participants

Twenty-four children were recruited from "La Cruz Roja Cerebral Palsy Center," Valencia, Spain. Inclusion criteria were: being between 4 and 18 years old, a diagnosis of bilateral CP, severe levels of gross motor function (GMFCS-E&R),[8] severe to moderate levels on the Manual Ability Classification System (MACS),[9] severe levels on the Communication Function Classification System (CFCS),[24] hearing preservation, interest in music and no risk of self-harming. In addition, participants' parents or guardians gave informed consent for participation in the study and video recording. Exclusion criteria were: severe behavioral problems, poor attention due to secondary effects of medication, non-attendance to more than 20% of the sessions. Only 18 patients met the inclusion criteria (12 males and six females) who were randomized to receive NMT. A blinded pediatric physical therapist assessed all the children before (T0) and after the following periods. The first 16 weeks, a group of 9 children received physiotherapy treatment once a week only (control group= T0-T1), while another group of 9 children received NMT in addition to the usual physiotherapy treatment (First 9 subjects of the intervention group= T1-T2 A). After this period, the groups were crossed over for the following 16 weeks. The children who previously received only physiotherapy treatment, would receive NMT at this point (second 9 subjects of the intervention group= T1-T2 B). The carry-over effect of *NMT* was assessed in 9 children after a washout period

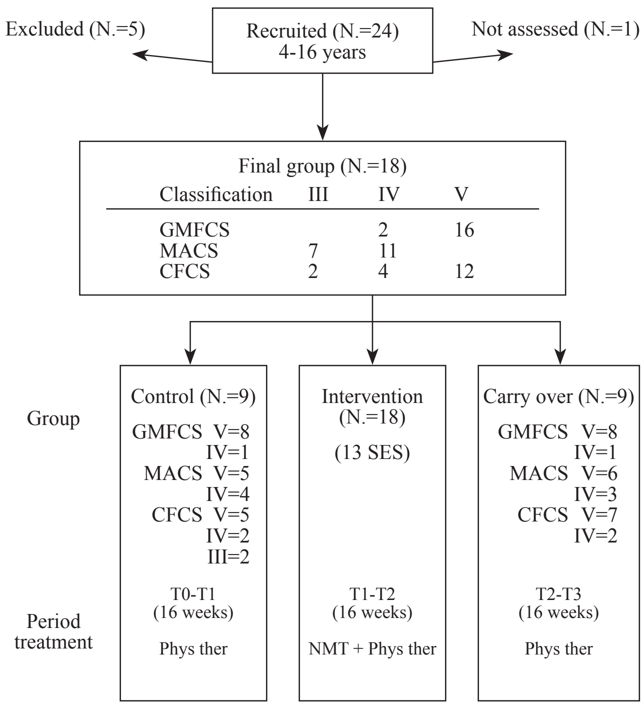

Figure 1.—Study design. Baseline features.
T: evaluation time; NMT: neurologic music therapy; SES: sessions; GMFCS: gross motor classification system; MACS: manual ability classification system; CFCS: communication function classification system; phys ther: physical therapy.

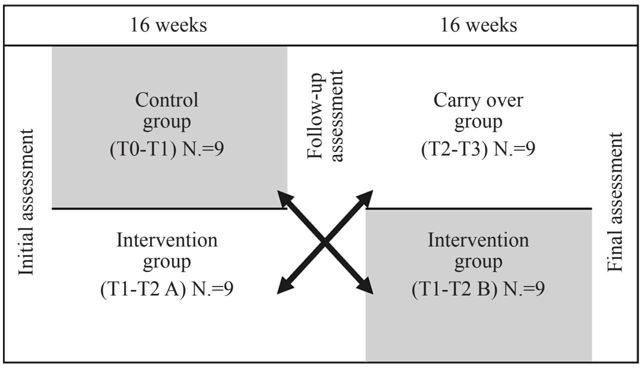

Figure 2.—Cross-over intervention timing.

when they again received physiotherapy only (carry-over effect group= T2-T3). This cross-over design allowed us to increase our intervention group (N.=18), and to treat all the children equally for ethical reasons (Figure 1, 2). Inter and intra-rater reliability of the examiner were studied in previous research and they were higher that 0.9 ($P<0.05$) for GMFCS and locomotor stages quantification.[25]

Intervention

Thirteen NMT sessions were carried out over 16 weeks. Intervention was performed once a week and simultaneously by a team of two music therapists in order to improve motor learning. The music was always live and customized according to each patient's needs and preferences. Small percussion instruments, Spanish guitar, keyboard (Casio Tone Bank-6) and drums (e-drum 150) were played.

Two types of activities were performed during the sessions. At the beginning, patients chose and played the musical instruments using their own movement strategies (hitting, rubbing, crashing) to generate rhythmic music patterns (10% of the session). Music therapists played along in order to contribute to the structure and cosmetic function to the baseline pattern. Children integrated their own musical creativity, thereby strengthening their intrinsic motivation, as well as expression and communication skills.

Afterwards, therapeutic instrumental music performance activities included repetitive, task-specific training with varied and incremental levels of difficulty (80% of the session). The tasks required children's attention and concentration, and were organized to stimulate, sign and guide movement. Music therapists composed small pieces of music testing different music parameters trying to trigger forearm pronation and supination, elbow flexion and extension and shoulder flexion. Most of the activities were performed in a prone position or sitting in their wheelchairs and were aimed at challenging upper-limb movements and head and trunk control.

The sequencing process was open and under continuous evaluation in order to define the functional goals. All the sessions lasted 40 minutes, were video recorded from front and lateral views, and the specific tasks to train through the different music interventions were defined after assessing each child's needs and the required movement components. The sessions were divided into 4 parts: 1) introduction: updated information was collected from the patients; 2) "Warm-up": rhythm and emotional adaptation with preparatory exercises (10%); 3) Treatment (80%, 25-30 minutes); 4) Recap (10%), summary and preparation for the next session.

Assessment of outcomes

The assessment before and after the intervention was carried out by the same pediatric physiotherapist,[25] who was blinded to the intervention group.

"Chailey Levels of Ability"[26] was selected for its quantitative and qualitative features assessing mild changes of the functional skills in children with severe CP (high correlation with GMFM and AIMS). All the parameters described in this scale were scored, as well as the locomotor stages which classify gross motor and locomotion functions.[25]

Statistical analysis

The Wilcoxon Test was used for all continuous variables. Baseline characteristics were compared between groups using Mann-Whitney U tests. Data analysis was carried out using SPSS 21.0. Only P values less than 0.05 were considered statistically significant.

Results

No significant differences between intervention and control groups in any of variables assessed were found at the beginning of the study.

Average values and standard deviation before and after the NMT intervention of the whole sample (N.=18) are shown in Table I. Data included the summaries of significant differences before and after the intervention: overall scoring of Chailey Levels of Ability ($P=0.002$); "activities" section ($P=0.007$); "arm and hand position" section ($P=0.027$) and locomotor stages ($P=0.008$). Average values increased after NMT intervention in these variables, while no significant differences were observed in the rest of the parameters studied. Nevertheless, a progressive in-

TABLE I.—*Average values and standard deviation of the studied variables before and after music therapy intervention (groups T1-T2 A and T1-T2 B).*

Variables in intervention group	Average T1	Average T2	P value
Symmetry	8.22±3.59	8.22±3.59	1.000
Loadbearing	16.61±6.05	16.67±6.05	0.317
Shoulder girdle position	14.44±4.03	14.44±4.03	1.000
Pelvic girdle position	14.11±3.46	14.11±3.46	1.000
Lateral spinal profile	11.06±4.96	11.06±4.96	1.000
Head movement	11.67±3.87	11.89±4.00	0.157
Chin position	12.06±4.29	12.06±4.29	1.000
Arm and hand position	13.56±5.38	15.22±6.12	0.027*
Leg position	7.17±3.11	7.39±3.35	0.180
Activities	11.00±6.73	13.17±7.33	0.007*
Ability to be positioned	9.11±2.30	9.11±2.30	1.000
Trunk position and movement	4.39±0.92	4.39±0.92	1.000
Hip position	1.50±1.76	1.50±1.76	1.000
Loadshifts	1.94±0.54	1.94±0.54	1.000
Width of standing base	2.00±0.59	2.00±0.59	1.000
Total Chailey Test	138.83±42.41	143.17±43.44	0.002*
Locomotor stages	1.17±0.86	1.56±0.62	0.008*

P-value column defines significant differences between these measurements.
Pre-intervention = Average T1; Post-intervention = Average T2.

TABLE II.—*Average values and standard deviation of the studied variables in both assessments of the control group (T0-T1).*

Variables in control group	Average T0	Average T1	P value
Symmetry	8.22±3.35	8.22±3.35	1.000
Loadbearing	16.78±8.06	16.78±8.06	1.000
Shoulder girdle position	14.33±4.90	14.33±4.90	1.000
Pelvic girdle position	14.22±4.41	14.22±4.41	1.000
Lateral spinal profile	10.22±5.14	10,22±5.14	1.000
Head movement	11.56±4.28	11.56±4.28	1.000
Chin position	12.56±4.42	12.56±4.42	1.000
Arm and hand position	12.00±5.00	12.00±5.00	1.000
Leg position	7.11±3.22	7.11±3.22	1.000
Activities	10.33±6.30	10.67±6.00	0.317
Ability to be positioned	9.78±2.73	9.78±2.73	1.000
Trunk position and movement	4.33±1.00	4.33±1.00	1.000
Hip position	1.67±2.00	1.67±2.00	1.000
Loadshifts	1.89±0.78	1.89±0.78	1.000
Width of standing base	1.89±0.78	1.89±0.78	1.000
Total Chailey Test	136.9±48.9	137.2±48.6	0.317
Locomotor stages	1.22±0.83	1.33±0.87	0.317

P value column defines significant differences between these measurements.
Average T0: initial assessment; average T1: second assessment after 16 weeks of initial assessment (control group).

creasing of "load-bearing," "head movement" and "leg position" values can be observed after NMT.

Baseline values for all variables remain stable in the control group (Table II), showing no changes with conventional physiotherapy.

Improvements persisted 16 weeks after the NMT treatment was discontinued and only usual physiotherapy treatment was performed. Table III compares measurements taken in the intervention group after a 16-week follow-up without NMT (average T3). No significant differences in sex or age were observed.

Table IV shows that there were no statistical differences between intervention groups T1-T2 A and T1-T2 B at assessment time previous to NMT intervention.

TABLE III.—*Average values at three assessment times (groups T1-T2 A and T2-T3).*

Variables in carry over group	Average T1	Average T2	Average T3	P value T1-T2	P value T2-T3
Symmetry	8.22±4.02	8.22±4.02	8.22±4.02	1.000	1.000
Loadbearing	16.44±3.57	16.56±3.57	16.56±3.57	0.317	1.000
Shoulder girdle position	14.56±3.24	14.56±3.24	14.56±3.24	1.000	1.000
Pelvic girdle position	14.00±2.45	14.00±2.45	14.00±2.45	1.000	1.000
Lateral spinal profile	11.89±4.94	11.89±4.94	11.89±4.94	1.000	1.000
Head Movement	11.78±3.67	11.78±3.67	11.78±3.67	1.000	1.000
Chin position	11.56±4.36	11.56±4.36	11.56±4.36	1.000	1.000
Arm and hand position	15.11±5.58	15.33±6.08	16.44±7.09	0.317	0.465
Leg position	7.22±3.19	7.33±3.08	7.67±2.96	0.317	0.317
Activities	11.67±7.45	13.56±8.41	13.56±10.25	0.109	1.000
Ability to be positioned	8.44±1.67	8.44±1.67	8.44±1.67	1.000	1.000
Trunk position and movement	4.44±0.88	4.44±0.88	5.44±2.96	1.000	0.317
Hip position	1.33±1.58	1.33±1.58	1.33±1.58	1.000	1.000
Loadshifts	2.00±0.00	2.00±.00	2.00±0.00	1.000	1.000
Width of standing base	2.11±0.33	2.11±0.33	2.11±.33	1.000	1.000
Total Chailey Test	140.7±37.6	143.1±38.2	145.5±41.0	0.027*	0.917
Locomotor stages	1.11±0.93	1.44±0.73	1.56±1.01	0.083	0.564

P value defines significant differences among the 3 scoring times.
Average T1 shows average values and standard deviations at the first assessment before music therapy intervention. Average T2 includes similar data taken after 16 weeks of music therapy intervention. Average T3 includes the carry over quantification assessed at a 16 weeks follow-up after music therapy intervention was discontinued.

TABLE IV.—*Statistical differences in the studied variables at initial assessment between intervened groups (T1-T2 A and T1-T2 B).*

Variables	Average T1-T2 A	Average T1-T2 B	P value
Symmetry	8.22±4.02	8.22±3.35	1.000
Loadbearing	16.56±3.57	16.78±8.06	0.941
Shoulder girdle position	14.56±3.24	14.33±4.90	0.911
Pelvic girdle position	14.00±2.45	14.22±4.41	0.897
Lateral spinal profile	11.89±4.94	10,22±5.14	0.493
Head movement	11.78±3.67	12.00±4.53	0.910
Chin position	11.56±4.36	12.56±4.42	0.636
Arm and hand position	15.33±6.08	15.11±6.53	0.941
Leg position	7.33±3.08	7.44±3.78	0.946
Activities	13.56±8.41	12.78±6.57	0.830
Ability to be positioned	8.44±1.67	9.78±2.73	0.229
Trunk position and movement	4.44±0.88	4.33±1.00	0.806
Hip position	1.33±1.58	1.67±2.00	0.700
Loadshifts	2.00±0.00	1.89±.78	0.675
Width of standing base	2.11±0.33	1.89±0.78	0.444
Total Chailey Test	143.1±38.2	143.2±50.4	0.996
Locomotor stages	1.44±0.73	1.67±0.50	0.461

P value column defines significant differences between these measurements
Initial assessment of intervention group A: average T1-T2 A; initial assessment of Intervention group B: average T1-T2 B.

Discussion

No other paper has previously shown our positive outcomes using NMT in the functionality of children with severe bilateral CP (GMFCS V= 88.88%).

The results of this study confirmed our hypothesis that an optimized music therapy program including active music techniques (TIMP) improves upper-limb function in children with severe bilateral CP.

Our results adjust coherently to task-specific training that could be repeated more while progressively increasing the level of difficulty. Therefore, the "arm and hand position" and "activities" fields could be trained more intensely. Improvements in "load-bearing," "leg position," and "head movement" have also been observed, even if not statistically significant. Proprioceptive feedback and feedforward when playing the musical instruments could support the positive outcomes of these variables. The other variables remained at their baseline values. This is self-explanatory since these tasks could not be trained as much due to patients' limitations. This leads us to believe that NMT could improve sensorimotor control in different neurological disorders with severe motor, cognitive and communication difficulties.

It could be possible that physiotherapy treatment helped to maintain the effects of NMT on the carry-over group; however, it is unlikely that the improvements observed in group T1-T2 B were due to 16+16 weeks of physiotherapy treatment. Firstly, no improvements were observed when this group was treated only with physiotherapy as with control group T0-T1 (Table II). Secondly, Table IV showed no statistical differences between intervention groups T1-T2 A and T1-T2 B at assessment time previous to NMT intervention. Thirdly, usual physiotherapy treatment was carried out with the same regularity in the whole sample before this study was started; therefore, both groups performed from similar baseline conditions.

These findings prompt us to think that patients with different neurological conditions including severe motor, cognitive and communication difficulties could take advantage of the feedback, entrainment and priming effects with NMT techniques. That is, patients could improve with a therapeutic instrumental music performance (TIMP) program.

Research on children with both poor gross motor function and manual ability, as well as lack of verbal communication, has not been found. Previous studies included children with GMFCS levels I to III only because walking ability was a requirement.[27] Other inclusion criteria were being able to stand without support,[2] being able to transfer from sitting to standing,[3, 4] being able to sit at the keyboard unaided[7] and, being able to follow verbal instructions.

Our therapeutic approach was eclectic, flexible and aiming at overall functional outcome in order to face the severe motor, sensory, communication, cognitive and behavioral difficulties. Motor improvement can be obtained targeting cognitive and emotional aspects because rhythm determines movement[1] and music quality (subjective to each patient) increases cognition and feelings.[11, 28, 29]

Therapists observed and assessed musical preferences in order to understand subjective peculiarities and design specific tasks to facilitate motor control according to cognitive and communication skills. Live music was required to achieve this, creating and adapting the pieces to immediate needs. To complete this process, the comprehension of the proposed activities was also required. Both therapists used music to communicate and interact with each patient.

Sessions were customized individually according the patients' abilities in order to target objectives through a combination of manual tasks, sensory intervention, and postural challenges. This malleability provided freedom to increase motivation and attention, to improvise, and to optimize the therapy process.[22, 30] Improvisation techniques allowed the patients to repeat and automate tasks without stereotypical movements and to progressively use more complex activities. Moreover, repetitive movements during TIMP helps to guide and give structure to the movement,[1] taking advantage of the neurophysiological effects through "priming" and "entrainment" of this technique[11, 16, 23, 31] and the benefits due to immediate feedback[6, 14] when playing the instruments. Furthermore, TIMP also helps the development of manual function by handling musical instruments.

In this way, important requirements in neurorehabilitation fulfilled[13, 18, 19] active organization, participation, reward and motivation of the patient, as well as repetition and increasing task difficulty.

The control group was composed of 9 subjects (T0-T1), selected randomly from the same sample of children who had been and previously assessed before the intervention. This reduced the group selection bias and controlled the confusion variables.[32] The quantification of 9 patients (T2-T3) helps us to describe the carry-over effect of NMT, 16 weeks after discontinuing the treatment.

Limitations of the study

Limitations of this research include the heterogeneous nature of our sample. Our patients had mixed types of CP with unpredictable movements and emotional reactions. The cooperation between both therapists was important in order to make the handling of the children and percussion possible, and also to guide and contain the behavior and the emotions.

Further classification of these types of patients also according to their cognitive, psychological and emotional aspects (together with the current motor and communication classifications) would provide greater objectivity and uniformity in the results of future research. Reliability would also increase if a larger sample was studied; however, this would imply a significant timeline increase due to the customized intervention.

Clinical guidelines suggested in neurorehabilitation[33] may need further investigation, for instance, how to increase the number and frequency of the sessions, as well as, therapy intensity without reducing attention level. Relations with other parameters such a sleep may also need to be studied.

Future research lines could include an objective quantification of the upper-limb range of motion and communication improvements. This could be obtained using kinematic motion analysis.

Conclusions

Optimized and combined intervention of therapeutic instrumental music performance can improve the upper-limb function in children with severe bilateral CP. These positive effects remained 16 weeks after completing the treatment.

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