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ORIGINAL ARTICLE

The effect of training in an interactive dynamic stander on ankle dorsiflexion and gross motor function in children with cerebral palsy

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Abstract

Objective: To study the effect of active stretching of ankle plantarflexors using an interactive dynamic stander in children with cerebral palsy (CP).
Methods: Six children in Gross Motor Function Classification System classes I–III, aged 4–10 years, trained intensive active dorsiflexion in an interactive dynamic stander using ankle movement to play custom computer games following a 10-week control period. Gross Motor Function Measure Item Set, gait performance and passive and active dorsiflexion with extended and flexed knee were chosen as outcome parameters.
Results: Median active and passive ankle dorsiflexion increased significantly (5 and 10 degrees, respectively) with extended knee. There was a small but clinically significant increase in gross motor function. The intervention had no effect on temporospatial gait parameters.
Conclusion: In spite of the low number of participants, these results may indicate that intensive active stretching in an interactive dynamic stander could be an effective new conservative clinical treatment of ankle plantarflexor contracture in children with CP.

Keywords

Ankle, active stretching, cerebral palsy, gross motor function, rehabilitation device, range of motion

History

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Introduction

Reduced active and passive ankle dorsiflexion are common in cerebral palsy (CP). Limited dorsiflexion of the ankle during gait can lead to inappropriate loading of joints in the lower extremity with increased loading of active and passive structures limiting forefoot dorsiflexion, ankle dorsiflexion, knee extension and hip flexion. Incorrect loading associated with contracture, valgus deformities of the ankle and equinus have been correlated with pain [1]. Plantar flexor spasticity and contracture together with dorsiflexor weakness are common causes of reduced active dorsiflexion in CP. Physiotherapy has therefore traditionally focussed on passive stretching of the ankle plantarflexors together with strength training of the dorsiflexors to establish a balance in moments around the ankle allowing improved ankle coordination and range of movement.

The effects of passive stretching on muscle lengths and function in children with CP is uncertain [2]. Pin et al. concluded in their review that there was limited evidence that manual stretching can increase range of movements, reduce spasticity or improve walking efficiency in children with spasticity. They further concluded that more research was required given the lack of knowledge about treatment outcomes and the wide use of passive stretching. A more recent systematic review concluded that regular stretch does not produce clinically important changes in joint mobility, pain, spasticity or activity limitation in people with neurological conditions [3].

With limited evidence supporting the use of passive stretching, another approach to increasing active range of ankle dorsiflexion is the use of active stretching where the contract muscle is stretched during active movement training. A recent study by Zhao et al. [4] reported elongation of muscle fascicles, reduced pennation angle, reduced fascicular stiffness, decreased tendon length, increased Achilles tendon stiffness and increased Young's modulus in the calf muscle following a six-week program of passive stretching and active movement training in a study of seven children with CP. These physiological adaptations had a positive effect on length and improved the biomechanical function of the calf muscle. The challenge with active training of ankle dorsiflexors is to motivate children to many repetitions of the same movement. We speculated that the use of stimulating assistive devices, which made dorsiflexion purposeful and fun for the child, would be the best motivation for the training.

The purpose of this study was then to investigate whether it would be possible to implement an ankle-training program using an innovative interactive dynamic stander to facilitate active movement training actively stretching the plantarflexors and whether this form of training would increase the

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active and passive range of ankle dorsiflexion, temporospatial gait parameters and gross motor function in children with CP.

Methods

Ten children with a diagnosis of CP were included in the project. Details of the children's gender, age, dominant neurological symptom and gross motor function classification as defined by the Expanded and Revised Gross Motor Function Classification System (GMFCS E&R) [5] are presented in Table I.

Children were recruited through an announcement on the internet inviting interested physiotherapists or authorities to contact the project group. A preliminary meeting was held with all interested participants prior to inclusion in the project. Children from seven different municipalities spread throughout Denmark participated in the project.

The inclusion criteria were:

(1) CP diagnosis
(2) 4–18 years old
(3) Weight under 50 kg (equipment restraint)
(4) GMFCS E&R level I–III
(5) Indication for physiotherapy
(6) Cognitive ability adequate to understand the connection between ankle movements and the corresponding results in the computer games
(7) No botulinum toxin in the two months prior to inclusion or during inclusion
(8) No surgery six months prior to inclusion or during inclusion

The study was designed as a controlled study, where the children started with a control period of 10 weeks where they received therapy as usual followed by a 10-week intervention period. The intervention consisted of training in the training equipment 30 minutes per day, five days a week for a period of 10 weeks in addition to therapy as usual.

The training equipment utilized (HAPPY REHAB, InnovaidApS, Aarhus, Denmark) resembles a modified standing frame. The training equipment has two rotating foot pedals to allow for isolated plantar and dorsiflexion of each ankle. There are force sensors on the surface of the rotating foot pedals and moveable leg supports. These force sensor signals control small motors, which either resist or assist the child's ankle movements. The degree of resistance or assistance is calibrated to the child's strength, coordination and range of movement. Ankle movements are used to control computer games motivating the child to perform the desired movements repeatedly. The children trained without shoes and ankle foot orthoses were removed during the training to allow full ankle range of movement.

The training equipment is illustrated in Figure 1.

The children were tested three times during the study; at baseline, immediately after the control period and immediately after the intervention period. Testing comprised Gross Motor Function Measure Item Set (GMFM-IS), 10 m walk test recorded on video and measurement of their active and passive range of ankle dorsiflexion.

GMFM-IS tests for each child were carried out by their primary physiotherapist and video recorded. When the child had completed the final GMFM-IS test, the three tests were scored by a pediatric physiotherapist with experience in scoring GMFM-IS. The physiotherapist was blinded to the chronological order of the tests during scoring.

The 10-m walking test (10 MWT) was carried out by the child's primary physiotherapist following a detailed written and verbal instruction from a member of the project group. The time, number of steps taken and walking aid were recorded for each test.

Active and passive range of ankle dorsiflexion with flexed and extended knee were measured by the child's primary physiotherapist (tester) together with an assistant following a detailed written and verbal instruction from a member of the

Table I. Details of the participants' gender, age, dominant neurological symptom and gross motor function classification as defined by the expanded and revised Gross Motor Function Classification System.

Participant	Age (Year.month)	Gender	GMFCS E&R	Dominant neurological symptom (spastic, dyskinesia or ataxia)
1	6.2	Female	III	Spastic diplegia
2	11.5	Female	II	Spastic diplegia
3	4.8	Male	II	Spastic diplegia
4	5.4	Male	I	Spastic hemiplegia sin.
5	4.6	Male	I	Spastic hemiplegia dxt.
6	8.8	Male	II	Spastic diplegia
7	10.4	Female	I	Spastic hemiplegia dxt.
8	10.0	Male	II	Spastic diplegia
9	10.5	Female	III	Dyskinesia
10	4.1	Male	I	Spastic diplegia

GMFCS E&R, expanded and revised Gross Motor Function Classification System.

Figure 1. Child training in the HappyRehab dynamic stander.

project group. The measurement protocol was adapted from the Nordic CP follow-up programme protocol and was therefore well known to the participating physiotherapists. To help eliminate measurement bias, maximum passive and active ankle dorsiflexion were measured using a supplied modified goniometer (International Goniometer 1 degree precision, 12″ (30 cm), Procare, Roskilde, DK) where the tester was unable to see the result. The measurements were recorded by the assistant with the primary physiotherapist blinded to the results until the data collection was completed. The measurements were repeated three times during each test. The highest value of the three measurements was used in the statistical analysis. For children with hemiplegia, only the affected ankle was measured.

Friedman's analysis of variance by ranks was used to check for statistical significant changes in the outcome measures and post-hoc pairwise comparisons using Wilcoxon's signed-rank test were then performed if there was a significant result. Statistical analysis was performed using IBM© SPSS© Statistics Version 19 (New York, NY).

Procedures followed were in accordance with the Helsinki Declaration of 1975, as revised in 2008. The study was approved by the local ethical committee.

Results

Six of the included children completed the study. Table II details reasons for the individual children leaving the project.

Values for GMFM-IS, gait velocity, step length, 10-m walk test, active and passive ankle dorsiflexion for the children who completed the study are presented in Table III.

Active and passive dorsiflexion with a flexed knee showed no significant change ($p = 0.069$ and $p = 0.792$, respectively). There was, however, a highly significant increase in passive and active ankle dorsiflexion with extended knee ($p = 0.026$ and 0.001, respectively) with a mean increase in the median in the intervention period of 10 and 5 degrees, respectively. The present 10-week training intervention using active ankle training had a significant effect ($p = 0.048$) on the gross motor function of the children. Gait velocity and 10-m walk test show improvements during the study period, but there was no significant improvement in either the control or intervention period. There was no statistically significant change in step length.

Discussion

The outcome measures for this study were chosen because they are feasible in the clinical setting and have good reliability. The reliability of goniometry of the ankle in has

Table II. Children who did not complete the study with reasons for non-completion.

Participant	Reason for non-completion
1	The child did not have the adequate cognition to participate
4	Unable to comply in test situations
5	The family could not fit the training into their daily life and it could not be done in the kindergarten
10	The family could not fit the training into their daily life and it could not be done in the kindergarten.

Table III. Values for GMFM-IS, gait velocity, step length, 10-m walk test, active and passive ankle dorsiflexion for the children who completed the study.

Outcome	*N*	BL	Pre	Post	Friedman's test (*p*)	Wilcoxon's signed rank test (*p*)		
						BL→ Pre	Pre→Post	BL→Post
GMFM-IS	6	64.4 (56.9 to 88.0)	66.0 (53.9 to 89.7)	67.3 (55.9 to 96.0)	0.048*	0.655	0.014	0.138
Gait velocity, m/s	6	0.92 (0.26 to 1.89)	1.13 (0.52 to 1.92)	1.24 (0.78 to 2.38)	0.030*	0.074	0.207	0.028
Step length, m	6	0.51 (0.17 to 0.71)	0.52 (0.27 to 0.71)	0.56 (0.30 to 0.91)	0.154	–	–	–
10-m walk test, s	6	10.95 (5.3 to 39.1)	8.9 (5.2 to 19.3)	8.2 (4.2 to 12.9)	0.030*	0.075	0.463	0.028
Mean passive ankle dorsiflex flex knee**	9	0 (−10 to 45)	−6 (−20 to 45)	2 (−15 to 45)	0.069	–	–	–
Mean passive ankle dorsiflex ext knee**	9	−5 (−20 to 25)	−10 (−20 to 25)	0 (−10 to 30)	0.026*	0.655	0.017	0.027
Mean active ankle dorsiflex flex knee	11	−5 (−35 to 30)	−10 (−35 to 30)	−10 (−29 to 30)	0.792	–	–	–
Mean active ankle dorsiflex ext knee	11	−10 (−50 to 15)	−15 (−50 to 15)	−10 (−41 to 20)	0.001*	0.016	0.008	0.028

Outcomes are presented as median (range). Level of significance was set to $p < 0.05$.

n, number of subjects/number of tested legs; BL, baseline; Pre, pre-intervention; Post, post-intervention; GMFM-IS, Gross Motor Function Measure Item Set; Friedman's test, Friedman's analysis of variance by ranks.

*, Significant result; **, child number 8 had difficulties in relaxing during passive testing with passive dorsiflexion measuring less than active. We therefore chose to exclude data for passive dorsiflexion for this child from our analyses.

not been widely documented, but published studies show an intraclass correlation coefficient (ICC) of between 0.64, in a study of 38 patients with orthopedic problems [6], and 0.9, in a group of 38 children with CP [7]. We have in this study attempted to optimize reliability by using an established known protocol that has standard positioning and descriptions of anatomical landmarks and using multiple measurements by the same experienced pediatric physical therapist for the measurements [7]. The 10MWT has excellent test retest reliability in a study of 29 children with neuromuscular disease [8]. Gross motor function measure is a reliable and valid measure of gross motor function in children with CP [5, 9, 10]. The GMFM-IS was chosen for the study as it is much more efficient to administer than the GMFM-66 and has excellent agreement with the GMFM-66 [11].

The increase in ankle dorsiflexion seen in this study of 5 and 10 degrees is clinically significant, especially as it results from a 10-week intervention. The greater increase in active dorsiflexion in comparison with passive could also indicate that this type of training not only has an effect on the length-tension properties of the contract ankle plantarflexors but may also provide a training stimulus improving functional strength in the ankle dorsiflexors; however, this was not evaluated.

Differences between the effect of the training on ankle dorsiflexion with flexed and extended knee are to be expected. If we ignore the subtalar joint, then the gastrocnemii are two-joint muscles with their effect on the ankle joint dependent upon knee joint angle, whereas the soleus muscle is a single joint muscle. With a flexed knee, it is the soleus that limits dorsiflexion, and with an extended knee, it is the gastrocnemii muscles. Training in this intervention is carried out with an extended knee so that ankle dorsiflexion is resisted by active and passive force generation principally in the gastrocnemii muscles. It is therefore reasonable to expect greater adaptations in the gastrocnemii muscles as a result of the intervention producing a greater response in ankle dorsiflexion with an extended knee.

Gross motor function increased in both periods, but the statistical analysis revealed a significant change in GMFM only in the intervention period. This median change of 1.3 points in GMFM exceeds the minimum clinically important difference reported by Oeffinger et al. [12] for children in GMFCS levels II and III. A recent study by Park & Kim [13] used structural equation modelling to investigate the relationship between the various dimensions of the International Classification of Functioning, Disability and Health framework. This study of 98 children with CP developed a model identifying a causal effect between reduced range of movement and gross motor function. Our study corroborates this finding where we find an increase in range of movement together with an increase in gross motor function in these children.

The effect on gross motor function of another home-based neuro-rehabilitation program was reported by Mattern-Baxter et al. [14] in a quasi-randomized controlled trial of 12 children with CP who participated in a home-based locomotor treadmill training program. An intensive six-week training program produced an acceleration in the attainment of walking skills and decreased the amount of support used for walking in young children with CP. This would appear to corroborate the findings of this study that repeated, intensive and specific training has a positive effect on the gross motor function of children with CP.

Park & Kim [13] identified reduced selective motor control as a further motor impairment with a causal effect on gross motor function. The intensive coordinated activity at the ankle joint required to play the computer games might result in improved selective motor control at the ankle joint. Such an adaptation was reported by Reid et al. [15] following eccentric strength training of the spastic upper extremity of 14 children and adolescents with CP. This parameter could be an interesting focus for future studies, together with other neuromuscular measurements. Another possibility is to study cortical plasticity in response to this intervention using functional magnetic resonance imaging (fMRI). A pilot study by Phillips et al. [16] of six children with spastic CP used fMRI to measure cortical reorganization following a training intervention using body-weight-supported treadmill training. Similar measurements would be relevant in future studies to quantify the impact of this form for training on cortical reorganization. Such measurements will help to explain in greater detail the neurophysiological effects of the training.

Gait temporospatial measures appear to be affected by learning and are therefore difficult to interpret. Gait velocity and 10MWT times improve in both control and intervention periods and appear therefore to reflect learning effects. This also appears to apply to step length although this parameter did not show a statistically significant improvement. Future studies should examine the precise effects on gait of active stretching with a closer focus on parameters related to ankle kinematics, lower extremity kinetics and muscle activation patterns using instrumented gait analysis and electromyography.

The use of computer games appears to be a motivating factor, which secured good compliance among the participants in spite of the high training intensity and repetitive nature of the intervention. A recent study by Burdea et al. [17] reports a similar finding that game technology was appropriate for strength, motor control and function training in a group of three children with CP from 7 to 12 years of age. This same study reported that the 12-week intervention with game-based robotic training of the ankle led to a substantial improvement in gait speed, ankle kinematics and endurance together with GMFM improvements typical of other ankle strength training programs.

Our study corroborates that there is a considerable potential to motivate children to train repetitively using newer technology; however, more evidence of the effect is needed.

Five of the six children who participated in this study used ankle-foot orthoses (AFO) when they were not training. The use of AFO is widespread in children with plantar flexor contracture to reduce the risk of further contracture and to remove the risk of knee hyperextension due to the plantar flexor-knee extension couple in the stance phase of gait. Details of the individual AFO types was not recorded in this study, but this information may be relevant in future studies where a fixed AFO may inhibit the potential functional gain made possible by an increased active and passive mobility in the ankle joint.

Further work is required with a larger number of subjects and a longer intervention period to determine whether this form of training could produce larger significant gross motor functional improvements in children with CP by greater positive impact on their motor impairments.

Future studies documenting this area should include a greater number of participants who are representative of the population if the results are to be generalized. In spite of the low number of participants, these initial results may indicate that training in an interactive dynamic stander could be an effective new conservative clinical treatment of ankle plantarflexor contracture in children with CP.

Declaration of interest

One of the authors works as a consultant with InnovaidApS who produce and market the equipment used in the intervention. The authors alone are responsible for the content and writing of this paper.

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