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Original Article

Effects of ankle continuous passive motion on soleus hypertonia in individuals with cerebral palsy: A case series

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ABSTRACT

Background: Continuous passive motion device (CPM) provides repetitive movement over extended periods of time for those who have low functional ability. The purpose of this research was to evaluate the effects of a four-week program of continuous passive motion of the ankle joint on the changes in soleus hypertonia in individuals with cerebral palsy who suffered from life-long hypertonia.

Methods: A single group, repeated-measures study was conducted. Eight individuals (7 males and 1 female with a mean age of 21.8 ± 8.5 years) with spastic cerebral palsy underwent bilateral ankle CPM for 1 h a day, 5 days a week, for 4 weeks. The outcome measures included the Modified Ashworth Scale (MAS) score, passive range of motion (PROM) of the ankle, the ratio of maximum H reflex to maximum soleus M-response (H/M ratio), and post-activation depression (PAD). All outcomes were measured before and after the intervention. A paired t-test was used to examine treatment effects pre-versus post-intervention.

Results: Paired t-tests showed that the CPM program significantly decreased the MAS score ($p = 0.006$), decreased the maximum H/M ratio ($p=0.001$), improved PAD ($p = 0.003$, $p = 0.040$, and $p = 0.032$ at 0.2 Hz, 1 Hz, and 2 Hz, respectively), and increased the passive ankle range of motion ($p = 0.049$).

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Conclusion: Ankle CPM not only reduced soleus hypertonia but also improved the PROM in individuals with cerebral palsy. The results of this study show ankle CPM to be an effective intervention for individuals with cerebral palsy.

At a glance commentary

Scientific background on the subject

Improving ankle spasticity is a fundamental goal for individual with spastic cerebral palsy. Continuous passive motion devices provide repetitive training with less manpower for cyclic stretching. Nevertheless, there is no clear consensus on using the Modified Ashworth Scale and neurophysiological methods to determine the effectiveness of ankle continuous passive motion in individuals with spastic cerebral palsy.

What this study adds to the field

Ankle continuous passive motion device is a safe and effective intervention to reduce soleus hypertonia and increase the passive range of motion for individuals with cerebral palsy. It may employ before gait training to improve ankle joint performance in individuals with moderate or severe ankle spasticity.

Spasticity occurs in 70–80 % of individuals with cerebral palsy (CP) [1–3]. Ankle spasticity is one of the most common movement disorders in CP [4,5]. A spastic hypertonic ankle can adversely restrict the mobility and impair the independence of CP patients [6,7]. The muscle tone in ankle plantarflexors, as measured with the Modified Ashworth Scale (MAS), increases up to 4 years of age and decreases until 12 years of age; the spasticity level of CP then becomes stable with no significant change after age of 12 [1]. Although individuals with CP suffer from life-long spasticity, they are usually not offered sufficient opportunities for therapy due to high labor demands placed on therapists and limited benefits derived from current therapeutic strategies [8]. Therefore, it would be challenging and interesting to study novel interventions for improving hypertonia in individuals with spastic CP over 12 years of age.

Traditionally, slow and continuous stretching is used to prevent contracture and decreases spasticity resulting from CP [6]. Passive stretching places high demands on manpower and has a relatively short duration. Recently, continuous passive motion devices (CPM) have been proposed to address these barriers to therapy and to provide repetitive training with less effort [9]. CPM is usually applied by platform-based devices for cyclic stretching. CPM was first developed to increase passive ankle joint range of motion, ameliorate ankle stiffness, and increase the comfortable walking speed of neurologically impaired patients [10,11]. To ensure safe and effective gait training, a platform-based ankle CPM is usually employed before a wearable ankle robot to improve ankle joint performance before gait training in individuals with moderate or severe ankle spasticity [11]. CPM has demonstrated the potential to reduce spasticity for individuals with CP. The application of repetitive passive knee movement reduced lower extremity spastic hypertonia in children with CP and improved their ambulatory function [12]. Ankle CPM combined with active movement training significantly improved both active and passive range of motion (PROM) of ankle dorsiflexion, strength of ankle dorsiflexors, and functional activity in terms of balance maintenance and longer walking distance in children with CP [13]. However, knowledge of its effectiveness is still limited for older individuals with CP. The present study used the MAS and neurophysiological methods to determine the effectiveness of CPM in individuals with spastic CP.

Previous studies showed that the ratio of the maximum Hoffman reflex (H-reflex) to the maximal motor response of the soleus muscle (H/M ratio) in individuals with hypertonia was high [14,15]. Therefore, these neurophysiological measurements will complement the MAS in providing a complete picture of the hypertonia of individuals with spastic CP. The soleus H-reflex is increased in a spastic leg while the muscle response (M-wave) is stable. Therefore, a reduction in the H/M ratio in the spastic lower limb of individuals with CP can be interpreted as a reduction in spasticity [16]. Moreover, the severity of spasticity in adults with spastic CP was highly correlated to the diminished post-activation depression (PAD) of the H-reflex [4]. The purpose of this study was to investigate the effects of a CPM program on the changes in soleus hypertonia in individuals with CP. The hypothesis was that after 4 weeks of a CPM regimen, individuals with spastic CP would show improvements in their MAS score, the maximum H/M ratio, the PAD of H-reflex, and their ankle PROM.

Methods

Participants

Eight individuals with spastic CP (7 males; 1 female) with a mean age of 21.8 years (SD = 8.5), diagnosed with spastic diplegia or spastic quadriplegia or hemiplegia, were recruited from the community for this study [Table 1]. Only three of the participants walked independently without aids (Gross Motor Function Classification System Level I) [17]. The inclusion criteria were a clinical diagnosis of spastic CP, age 16–50 years, an MAS score of ankle plantar flexors greater than 0, ankle PROM greater than 10°, and stable medical conditions. None of the participants were on antispasticity medications during the period of the study. Participants were excluded if they had other neuromuscular disorders, severe ankle contracture (<10° PROM), a history of rhizotomy, fracture in the lower extremities, or recent (<6 months) Botox injections. The study protocol was approved by the Institutional Review Board of the Chang Gung Medical Foundation in accordance with the Helsinki Declaration before enrollment of the first participant.

Table 1 Clinical features of the participants (n = 8).

Participant	Gender	Age	Classification	Ambulation	GMFCS level
1	M	42	Spastic diplegia	A	I
2	M	22	Spastic diplegia	A	I
3	M	19	Spastic diplegia	NA	IV
4	M	22	Hemiplegia	A	I
5	M	16	Spastic quadriplegia	NA	IV
6	M	18	Spastic quadriplegia	NA	V
7	M	17	Spastic quadriplegia	NA	IV
8	F	18	Spastic quadriplegia	NA	IV
Mean/ratio	M:F 7:1	21.8 ± 8.5	Spastic diplegia : Hemiplegia : Spastic quadriplegia 3 : 1 : 4	A : NA 3 : 5	I : IV : V 3 : 4 : 5

Abbreviations: M: male; F: female; MAS: Modified Ashworth Scale; A: ambulant subject; NA: non-ambulant subject; GMFCS: Gross Motor Function Classification System.

Study design and procedures

This was a single group, repeated measures study. Pre-versus post-intervention changes were used to determine the effectiveness of the intervention. The authors registered the study at ClinicalTrials.gov under the number NCT02003755. Twenty individuals from the community volunteered to enroll in the study, eight of whom met the inclusion criteria [Fig. 1]. All participants gave their written informed consent before participating.

Intervention

Ankle CPM training was administered using a custom-made training system which has been previously utilized in patients with spinal cord injury [9,18,19]. This system provides CPM of the ankle joint [Fig. 2]. Behind the heel, there are two shallow cups on the platform, both 3 cm in height, to prevent the feet from sliding backwards. The cups were designed to be shallow so as not to interfere with the movement of the ankles during training.

The ankle was cycled between 5° plantarflexion and 5° dorsiflexion. A larger range of motion might have exceeded the participants' limitations, causing soft tissue injury and/or inducing more spasticity in individuals with CP who had long disease history and joint contracture. From a biomechanical point of view, the ankle joint displacement of 10° used in this study was similar to an ankle displacement of approximately 15° during cycling in a seated position [20]. The speed of the system was manually adjustable by an external controller. A potentiometer was built in to obtain the real-time degree of the platform. The frequency range of repeated PROM exercises used in previous studies was 24–72 revolutions per minute (RPM) [21,22]. Chang et al. used 60 RPM for robot-assisted CPM training of the ankle in individuals with chronic spinal cord injury to mimic the speed of ankle movement during ambulation and found it significantly decreased the MAS score [9,18]. Thus, in the current study, we used a constant speed of 60 RPM [9,18,19].

At the first training session, one of the researchers visited each participant's home and set up the system for them. The distance from the chair to the platform was adjusted to allow the participants' knees to be maintained at 70° flexion from full extension, while the hips were kept at 90° flexion and the ankles in a natural position. During each session, participants were seated with their feet positioned on the platform and the knee flexed at 70° from full extension to minimize the contribution of the gastrocnemius. The participant's foot was secured to the rigid footplate. Participants received 60 min per day of ankle CPM training of both legs, 5 days/week for 4 weeks at home. Except for ankle CPM, no other training or rehabilitation was given during the period of the experiment. A daily log of ankle CPM training sessions was kept and verified every week. All eight participants completed the 20 sessions of training in addition to the evaluations, which demonstrated good compliance to the training.

Outcome measures

The clinical examinations and the electrophysiological measurements were performed before and after 4 weeks of training. The post-training test was performed at least 24 h after the last training session to prevent observation of immediate short-term effects. To determine muscle tone of the ankle joint, the primary outcome measure was the MAS score, and the secondary outcome measures were PROM of the ankle, the maximum H/M ratio, and the PAD of the H-reflex. The MAS is a clinical assessment that measures the degree of hypertonia on a six-point scale. Alternatively, the neurophysiological methods are quantitative measurements of hypertonia. Although they are time-consuming and require special equipment and expertise, they are valuable tools for objective measurement [23].

Prior to electrophysiological testing, each participant underwent muscle tone evaluation of the ankle joint by a physical therapist using the MAS, as well as determination of the PROM. The MAS score was determined with the participant in a sitting position with knee flexion at 90° to minimize the influence of gastrocnemius muscle tone. The MAS score ranges from 0 (normal muscle tone) to 4 (fixed muscle contracture). The MAS was chosen because of its extensive use in clinical research. The intra-rater reliability of the MAS for measuring hypertonia of the gastrocnemius in children with spastic CP was moderate to good (intraclass correlation coefficients ranged from 0.56 to 0.79) [24,25]. The summed PROM of ankle dorsiflexion and plantarflexion was measured in the same position. The summed PROM was measured, rather than separate PROMs of dorsiflexion and plantarflexion, to avoid potential variations in defining the neutral ankle position before and after training. All measurements of

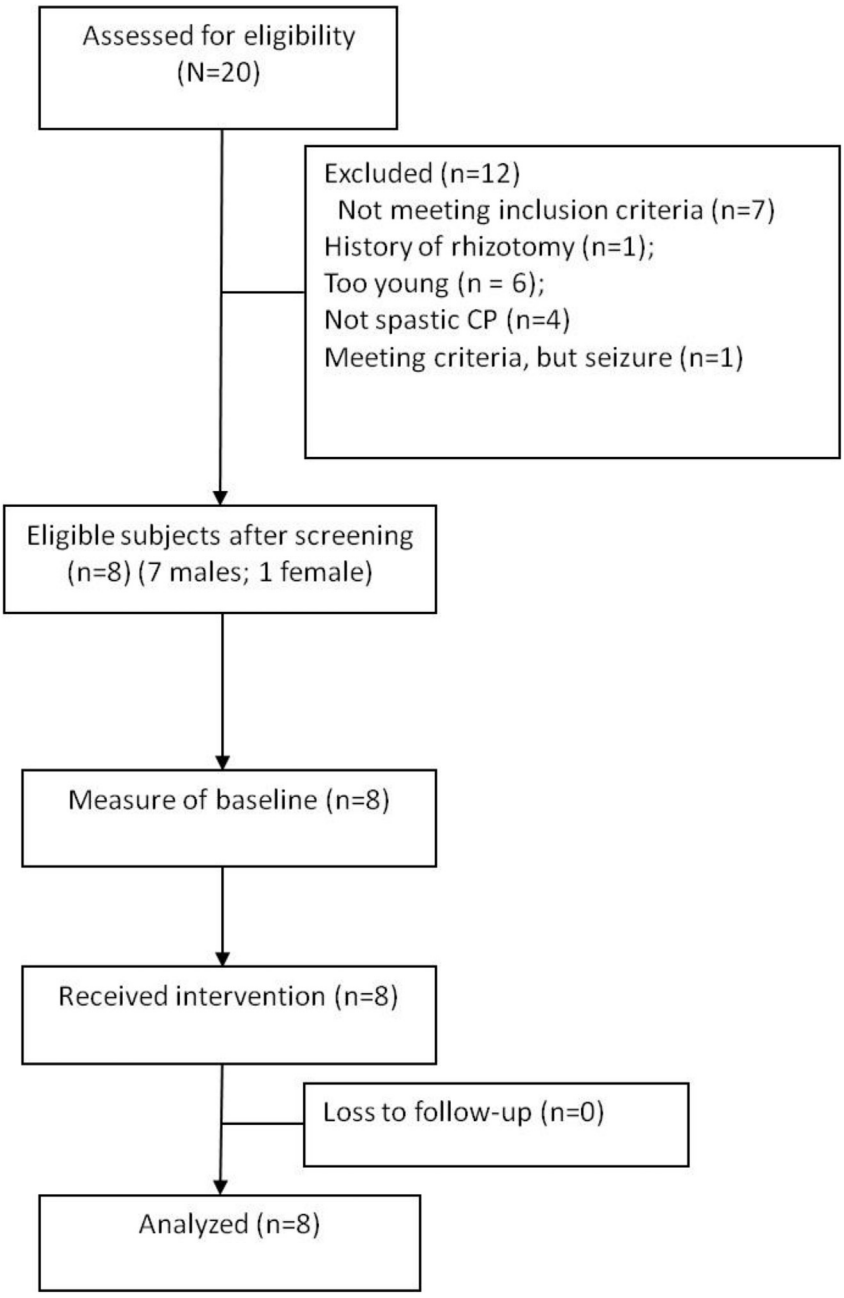

Fig. 1 Selection of the cerebral palsy subjects.

spasticity and joint range were performed under identical conditions for each test by a licensed physical therapist who was blinded to the purpose of the study.

For the electrophysiological tests, all participants were seated with back support. The test leg was determined in a randomized manner. The test leg was secured with hook and loop straps on a force-plate system [9,18] with the knee flexed at 70° from full extension to minimize the contribution of the gastrocnemius and the ankle at a neutral position (0° dorsiflexion or plantarflexion). The surface electromyographic recording electrode was positioned on the soleus muscle approximately 2 cm lateral to the midline of the distal calf and distal to the lateral head of the gastrocnemius [9,18,26–29]. A ground electrode was placed over the lateral malleolus. The muscle responses (M-waves) and H-reflexes of the soleus were elicited by transcutaneous electrical stimulation using a high voltage constant current stimulator (Stimulator model DS7A, Digitimer Ltd, Hertfordshire, UK) of the tibial nerve at the popliteal fossa with a fixed pulse width of 500 μs. The intensity of the electrical stimulation was adjusted higher until the maximal M-wave was found. The electrical stimulation was then set at an intensity which could elicit H-reflexes at an amplitude of 20 % of the maximal M-wave [30]. The timing and frequency control of the stimulation was provided by customized software that uses a programmable peripheral interface chip (PPI 8255) to send 5 V DC trigger pulses to the stimulator.

For the PAD recordings, five pairs of soleus H-reflexes were elicited in a randomized order at each of the following

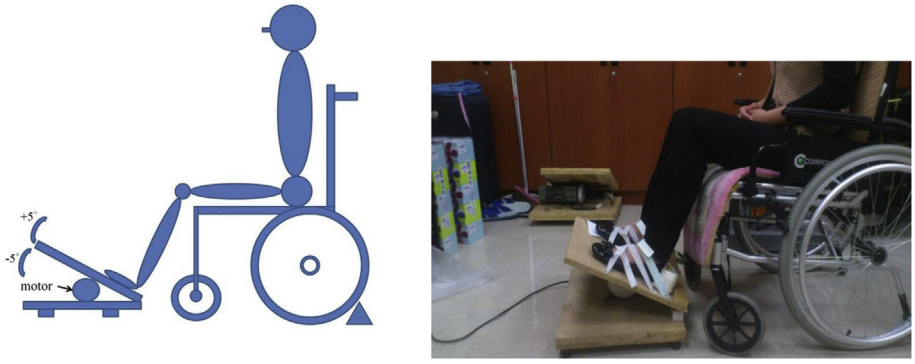

Fig. 2 Ankle continuous passive motion device provides continuous passive movements.

frequencies: 0.1 Hz, 0.2 Hz, 1 Hz, 2 Hz, 5 Hz, and 10 Hz, whereas the first H-reflex of each pair is the conditioning H-reflex and the second H-reflex is the testing H-reflex. We used the paired stimulation paradigm, instead of the train stimulation paradigm, to record PAD to avoid the potential contribution of muscular architecture changes induced by the train stimulation induced tetanic contractions [26,28,31]. The time interval between two consecutive pairs was set to 15 s to ensure complete recovery of the H-reflex [9,18]. All data were processed and analyzed off-line. Ten maximum M-waves (Mmax) and 10 maximum H-reflexes (Hmax) were recorded before CPM ankle training began. For calculating H/M ratio, the peak-to-peak amplitude Hmax was divided by Mmax. For PAD, the peak-to-peak amplitude of the testing H-reflex was divided by the peak-to-peak amplitude of the corresponding conditioning H-reflex in each pair of H-reflexes. The lower ratio indicated the stronger PAD.

Sample size calculation and statistical analysis

The sample size calculation was performed with G*Power 3 (a statistical power analysis program) [32]. Based on the MAS results of our previous study [9] and with the aim of showing clinically relevant differences, the Cohen's d effect size was set at 0.5. The program estimates that a total sample size of 7 was needed to reach 80 % power to detect an effect size of 0.5 at the 0.05 level of significance. With a potential 10 % attrition rate, a total of 8 subjects were targeted for this study. The data was presented using mean ± standard deviation. A paired t-test was performed to determine if the outcomes were different pre- versus post-intervention. The alpha level was set at 0.05.

Results

Descriptive and inferential statistics for outcome analyses are presented in Table 2. Significant differences were found in the MAS score and PROM after 4 weeks of ankle CPM. The MAS score was significantly decreased from 1.6 ± 0.3 to 1.1 ± 0.2 after CPM ($p = 0.006$), indicating decreased hypertonia. Also, the PROM of the ankle was significantly increased from 45.62° ± 8.63°–51.87° ± 9.23° ($p = 0.049$). It is apparent that only 4 of 8 participants improved ankle PROM after training and that of the other 4 participants remained unchanged.

Four weeks of ankle CPM significantly decreased the reflex excitability, as indicated by a significant decrease in the maximum H/M ratio ($p = 0.001$) [Table 2]. A lower ratio of test H-reflex/conditioning H-reflex represents a stronger PAD. After 4 weeks of ankle CPM training, the ratio of test H-reflex/conditioning H-reflex significantly decreased at 0.2 Hz, 1 Hz, and 2 Hz ($p = 0.003$, $p = 0.040$, $p = 0.032$, respectively) [Fig. 3]. Although there were no significant changes for the relatively higher frequencies of 5 Hz and 10 Hz; there was a trend to decrease the ratio after training. These results suggest that ankle CPM resulted in restoration of the PAD in adolescents and adults with CP.

Discussion

This study is the first to test the effects of a 4-week ankle CPM program on spastic hypertonia and ankle PROM in individuals with CP. Ankle CPM significantly improved the MAS score and ankle PROM, increased the maximum H/M ratio, and restored the PAD in individuals with CP, which supported our hypothesis.

Several factors may have contributed to the reduction of the MAS score and increased ankle PROM after ankle CPM. One is the normalization of spinal circuitry functions. Our study showed that the maximum H/M ratio decreased and PAD was restored after 4 weeks of ankle CPM. Another possible mechanism underlying the reduction in MAS score and increased ankle PROM could be the reduced sensitivity of muscle spindles [9]. In this case, passive motion causes the stretch reflex and provides continuing input to the spindle to decrease the reflex strength [33]. In spastic CP, stiffness and shortening of the soft tissues make the ankle joint resistant to stretching, limiting normal movement. McNair, Dombroski, Hewson, & Stanley (2001) found the dynamic nature of ankle CPM has a greater effect on decreasing the stiffness of the plantarflexor muscles than static holds [34]. An improvement in circulation resulting in the redistribution of the more mobile tissue constituents, such as the polysaccharides, and water redistributed within the collagen framework during continuous motion could also contribute to the improvements in MAS scores and ankle PROM [34].

CPM has been shown to decrease ankle hypertonia in individuals with chronic stroke [35], and spinal cord injury

Table 2 Descriptive and inferential statistics for outcome analyses.

Participant	MAS Pre	MAS Post	Ankle PROM (°) Pre	Ankle PROM (°) Post	Maximum H/M ratio Pre	Maximum H/M ratio Post
1	1	1	40	40	0.61	0.28
2	1.5	1	40	55	0.67	0.56
3	1.5	1	45	60	0.63	0.46
4	1.5	1	40	40	0.73	0.38
5	2	1	40	55	1.01	0.45
6	1.5	1.5	45	45	0.61	0.41
7	2.	1.5	50	55	0.73	0.52
8	1.5	1	65	65	0.66	0.30
Mean ± SD	1.6 ± 0.3	1.1 ± 0.2	45.62 ± 8.63	51.87 ± 9.23	0.70 ± 0.13	0.42 ± 0.09
p		0.006[b]		0.049[a]		0.001[b]

Abbreviations: MAS: Modified Ashworth Scale; PROM: passive range-of-motion; H/M ratio: Hoffman reflex to that of the maximal motor response of the soleus muscle.
The MAS score of 1.5 is being used in place of the category 1+.
[a] Significantly different from the pre-training ($p < 0.05$).
[b] Significantly different from the pre-training ($p < 0.01$).

[9,18]. However, using ankle CPM to reduce spasticity is a relatively new treatment concept. A study reported by Cheng et al. found that repetitive passive knee movement for 8 weeks significantly decreased the MAS score in children with CP [12]. Our study found that the MAS score significantly decreased after 4 weeks of ankle CPM training, suggesting that ankle CPM training is effective in reducing hypertonia in individuals with CP. However, it would be necessary to conduct MAS testing both before and after the intervention, as well as 4 weeks later, in participants not receiving the CPM therapy to validate this conclusion.

The 4-week ankle CPM training regimen significantly increased PROM of the ankle in 4 participants. It was noted that three were nonambulatory among these 4 participants. The PROM of the other subjects were maintained. This result was consistent with PROM improvement after postoperative shoulder CPM treatment in patients with arthroscopic rotator cuff repair [36] and after robot-assisted ankle CPM training in patients with chronic stroke [35]. Continuous passive movement of the ankle may enhance large fiber afferent input from muscle to spinal circuitries. The CPM exercises may induce more afferent inputs from muscle to spinal cord than static stretching treatments. Based on the gate-control theory of pain by Melzack and Wall, small noxious afferent inputs would be inhibited by competing afferent sensory stimulation, potentially increasing PROM while reducing pain [37].

Our study showed that the maximum H/M ratio before training was 70 ± 13 % for individuals with CP, which corresponds to a previous study which reported the maximum H/M ratio to be 62.81 ± 4.72 % for individuals with CP [4]. The maximum H/M ratio of individuals with CP, as reported in both our study and that of Achache et al., was higher than that in healthy adults (45.40 ± 3.79 %) [4]. This result was in line with previous studies indicated that high H/M ratio of the soleus muscle in individuals with hypertonia [14,15]. Four weeks of ankle CPM significantly decreased the mean maximum H/M ratio in the soleus muscle of individuals with CP from 70 % to 42 %, which complemented the reduction of the MAS score can provided a complete picture of a reduction in spasticity the hypertonia of individuals with spastic CP [16].

The mechanism for ankle CPM to cause restoration of PAD is not yet clear. A previous study showed that ankle CPM training restored the PAD in individuals with complete spinal cord injury, suggesting that the mechanism by which ankle CPM restored PAD was primarily through the plasticity of segmental spinal circuitry function rather than suprasegmental functions [9]. The possible mechanisms include alterations in presynaptic inhibition and interneuron activity as a result of passive training [31,38,39]. It has been suggested that exercise training was sufficient to cause plasticity in the spinal cord [39]. Passive exercise might mimic active exercise and provide recurrent signals that modify the hyperactive inhibition mechanisms within the spinal cord, restoring the PAD.

Our study showed that the PAD at 0.1 Hz, 1 Hz, and 2 Hz were successfully restored after four weeks of CPM training. A recent study using a novel human paradigm showed that the PAD is a pre-synaptic locus mechanism which was separately modulated from descending corticospinal influence [31]. This suggests that CPM may be modulated the presynaptic mechanism in the individuals with CP. The PAD at 0.1 Hz was usually served as a control condition, in which the interval between conditioning H reflex and testing H reflex was too long to produce PAD [9,31]. Our study showed that the PAD at 0.1 Hz was not changed after 4 weeks of training, suggesting that the recording was consistent. Although our study show that the changes of PAD 5 Hz, and 10 Hz were not significant, there was a trend to decrease the ratio after training. The observations of high frequency stimulations could be explained by a possible result of induced corticospinal contribution [40–43]. It probably due to that PAD at some frequency might require longer to restore. Our previous study showed that, in compared to 4 weeks of training, 12 weeks of training could further restore PAD at 5 Hz [9].

There are several common treatment strategies used in clinics for reducing spasticity and increasing PROM [44]. However, none of the common treatments have shown the ability to restore spinal circuitry functions in individuals with CP. This study is the first to show that ankle CPM might have the ability to cause plasticity in the spinal circuitry in adolescents and adults with CP, whose spinal circuitry is mature. Individuals with different upper motor neuron lesions exhibit

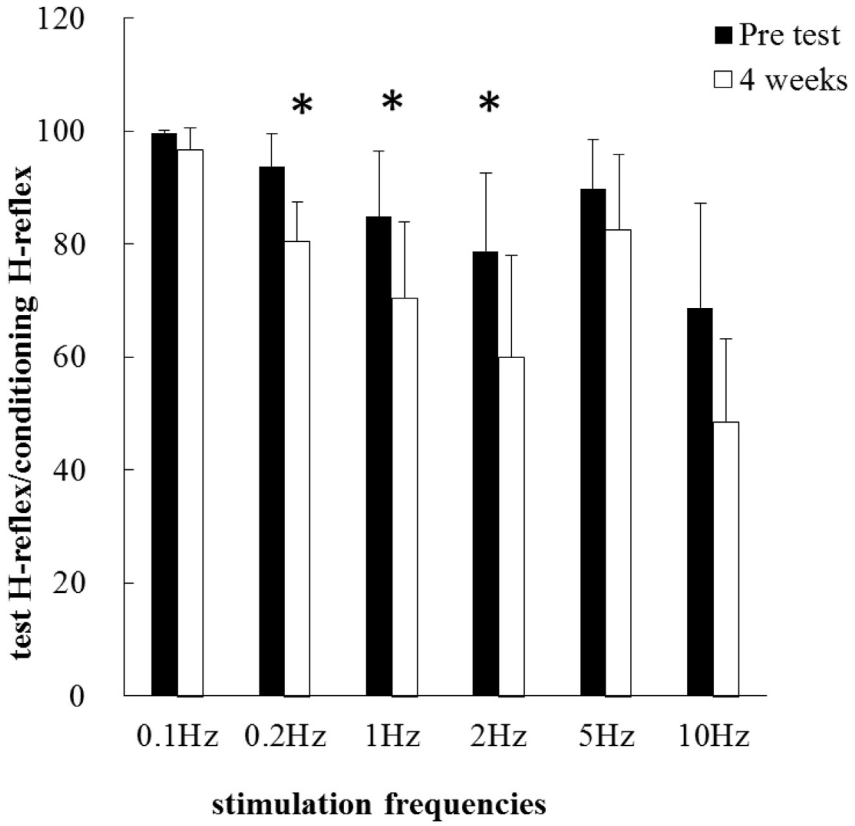

Fig. 3 Post-activation (PAD) depression prior to and after training in individuals with cerebral palsy. A lower ratio of test H-reflex/conditioning H-reflex indicates a stronger PAD depression. The bars show means and standard deviations of test H-reflex amplitude normalized to the respective conditioning H-reflexes in individuals with cerebral palsy. before (black bars) and after (white bars) the 4-week training regimen at stimulation frequencies of 0.1 Hz, 0.2 Hz, 1 Hz, 2Hz, 5 Hz, and 10 Hz *significant at $p < 0.05$.

decreased PAD [4,45–47], suggesting that impaired PAD might be the primary cause of spasticity after upper motor neuron lesions. Repeated PROM exercises may not only decrease neuron excitability in rats and humans with spinal cord injury but also improve the values of H-reflex, PAD and disynaptic Ia inhibition, which were demonstrated by a low frequency-dependent depression of the H-reflex [18,33].

Clinically, prolonged stretching is a common strategy to reduce spasticity [48], and relatively fast movement is avoided in spastic limbs. However, ankle CPM in our study successfully reduced spasticity in individuals with CP, with no reports of any harm in participants' daily logs. This suggests that ankle CPM could be applied safely in clinics for the treatment of spasticity. In addition, performing CPM exercises at home as a supplement to a clinical training regimen could be used to maximize the benefits of rehabilitation sessions [49–51]. Ankle CPM regimen like the one used in this study might increase the motivation and ankle function of individuals with spastic CP.

Study limitations

The design of this study has several limitations. First, the small sample size precludes generalizability to a larger CP population. Second, the participants had relatively low MAS scores at baseline, so this study cannot suggest how individuals with greater baseline spasticity might respond. Third, the study would be strengthened by adding a CP control group that does not go through the CPM training but is tested on the same measures at the same pre- and post-assessment time points as the group that went through the CPM training. Fourth, this study was conducted in non-pediatric CP patients whose spinal circuitry function was mature. The results cannot be extrapolated to pediatric CP patients. Fifth, the 4-week ankle CPM program was effective in non-pediatric CP patients, but the dose–response relationship requires further study. Finally, there was no follow-up assessment to examine whether the effects of ankle CPM training were maintained after the training regimen ceased.

Conclusions

As a preliminary result, a 4-week ankle CPM training program was able to effectively reduce spastic hypertonia in individuals with CP who were usually excluded from clinical rehabilitation programs, as measured by MAS scores. A parallel improvement of ankle PROM and restoration of spinal circuitry inhibition functions were also shown after ankle

CPM training. The ankle CPM rehabilitation could be a safe and appropriate clinical intervention for adults with CP.

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Conflicts of interest

The authors declare that they have no competing interests.

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