IJIRR

International Journal of Information Research and Review
Vol. 05, Issue, 10, pp.5753-5757, October, 2018

RESEARCH ARTICLE

THE EFFECTS OF PROGRESSIVE FUNCTIONAL TASK TRAINING EXERCISES ON MUSCLES STRENGTH AND MOBILITY IN CHILDREN WITH CEREBRAL PALSY

***[1]Dr. Ankita Pathak, [2]Dr. Singh, K.K., [3]Dr. Manoj Kumar Mathur and [4]Dr. Anuja Chaudhary**

[1]MPT (Ortho), Maharaja Vinayak Global University, Jaipur Physiotherapy College, Jaipur Rajasthan, India
[2]MPT (Ortho), PHD, Principal Dept of Physiotherapy, Maharaja Vinayak Global University, Jaipur Physiotherapy College, Jaipur Rajasthan, India
[3]MPT (Musculoskeletal Disorders). Assist professor, Dept of Physiotherapy, Maharaja Vinayak Global University, Jaipur Physiotherapy College, Jaipur Rajasthan, India
[4]MPT (Neurological Disorders). Assist professor, Dept of Physiotherapy, Maharaja Vinayak Global University, Jaipur Physiotherapy College, Jaipur Rajasthan, India

ARTICLE INFO

Article History:
Received 14th July, 2018
Received in revised form
17th August, 2018
Accepted 10th September, 2018
Published online 30th October, 2018

Keywords:

Evaluate,
Mobility,
Children.

ABSTRACT

Study Objectives: the objective of this study was to evaluate the effectiveness of a sufficiently long and intensive 12-week functional PRE strength training programme, by applying current guidelines. Design: Single-blind randomized controlled design. **Methods:** This study was conducted at Physiotherapy OPD of Jaipur Physiotherapy College, Mahraj vinayak global University, Jaipur on spastic unilateral or bilateral Cerebral Palsy children with physical disabilities. Full written informed consent was obtained from all parents and 12-year-old children before enrolment. The total 51 children were pre-stratified according to three stratification variables: sex, GMFCS level (I, II–III), age (youngest: 6–9y; oldest 10–13y), and subsequently randomized to one of two groups using sealed envelopes. Assessments were at baseline (T0), during training (T1; a subset of outcomes), directly after training (T2), and at the 6-week follow-up (T3). The measurement points of main interest were T2 and T3. The control group continued their conventional physical therapy programme. Children in the control group received one to three sessions a week. The training group followed a 12-week progressive functional task training programme for the lower extremities. This replaced their conventional physical-therapy programme. Training was given three times a week for 45 to 60 minutes at Neuro Physioherapy unit of OPD in small groups (four or five children). Each training session consisted of one exercise on a child-adapted leg-press (Enraf Nonius, Delft, the Netherlands) and three functional exercises (sit-to-stand, lateral step-up, half knee-rise), loaded with a weighted vest. During the training, intensity progressively increased, based on repeated estimation of the eight-repetition maximum. **Outcome Measure:** Gross Motor Function Measure (GMFM- 66), the Sit-To-Stand (STS) test, and the Lateral Step-Up (LSU) test**. Result**: Sixty-one patients were asked to participate; four turned out to be ineligible. Of the remaining 57 eligible patients, six did not give consent. The remaining 51 children were randomized. Of these, one dropped out before T0 (GMFCS level III, female, age 13y 1m, training) owing to a hip injury that made pretesting and training impossible, and one was lost to follow up at T1 (GMFCS level II, female, age 12y 1mo, training) owing to an unexpected long-term stay abroad. Analyses were performed for 49 participants. There was no statistically significant difference between the groups for personal characterstics. Training compliance and intensity of the 36 scheduled training sessions, three to six sessions were cancelled because of school-related activities. After correction for these, the mean compliance was 92.3% (range 71– 100%). A mean of 32 training sessions (range 30–33) were attended. Reasons for absence were illness (41.4%), medical appointment (8.6%), vacation (6.9%), or other ⁄ unknown (43.1%). Every 2 weeks, starting from week four, eight-repetition maximum tests were performed for leg-press and loaded STS exercises. Based on these eight-repetition maximum test values the training load progressed. During the last 8 weeks of training, the mean eight-repetition maximum strength increased from 116 to 149% on the leg-press, and from 28 to 41% on the loaded STS. Each week, one to six children reported mild to moderate muscle soreness. **Conclusion**: In conclusion, 12 weeks of functional PRE strength training is effective in increasing isometric muscle strength of the knee extensors and hip abductor by 11 to 12%, and increasing six repetition maximum leg-press strength by 14%. However, this does not result in an increase in mobility. Consequently, functional PRE strength training is indicated for a child with CP when the aim is to improve leg muscle strength. It could also be included in a more extensive treatment regime, addressing several elements of fitness including muscle strength, or used as a target treatment specifically anticipating temporary muscle weakness, such as before or after botulinum-toxin-A or surgical treatment.

****Corresponding author: Dr. Ankita Pathak,***
MPT (Ortho), Maharaja Vinayak Global University, Jaipur Physiotherapy College, Jaipur Rajasthan, India

INTRODUCTION

Cerebral palsy (CP) refers to a group of permanent disorders of the development of movement and posture, causing activity limitations, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain (Bax, 2005). Damage to the central nervous system cause disorders in neuromuscular, musculoskeletal and sensorial systems. These disorders result in posture and movement deficiencies. It is the most common cause of movement disability in childhood. Children with CP may experience a variety of impaired muscle functions, such as spasticity, muscle weakness, and loss of selective motor control. Although all impaired muscle functions limit the performance of daily life activities and participation in a child with CP, a recent study has shown that muscle weakness showed a stronger association with mobility limitations in children with CP than spasticity (Ross, 2007).

Strength training for these children is, therefore, expected to improve or maintain their mobility. Until recently, strength training in children with CP was discouraged as it was assumed that it would increase spasticity. However, this was not supported by the results of earlier uncontrolled studies which showed that strength training can improve lower-limb muscle strength in children with CP without increasing spasticity (Morton, 2005 and Eek, 2008). Although studies have shown there to be sufficient evidence for its effectiveness on muscle strength, these effects are probably overestimated because of the low methodological quality of the studies (Dodd, 2002 and Taylor, 2005). The few uncontrolled studies that have evaluated the effectiveness of strength training on mobility outcomes in children with CP have reported limited effectiveness (Morton, 2005 and Damiano, 1998). Three recently published randomized clinical trials8–10 evaluated both muscle strength and mobility in children with CP, but conflicting results were reported (Scianni, 2009 and Mockford, 2008). One of the explanations for these conflicting results could be the high variability in training characteristics, such as type of training, training intensity, and training period. To be successful, strength training must be individualized, and involve a progressive increase in intensity, thereby stimulating strength gains that are greater than those associated with normal growth and development (i.e. ‘overload’).

This is known as progressive resistance exercise (PRE). According to the US National Strength and Conditioning Association, children should be trained at an 8- to 15-repetition maximum, which is the number of repetitions that can be completed before fatigue (Faigenbaum, 2009). To stimulate strength progression, the amount of resistance should be increased as strength increases. Most previous strength training studies did not present full details of their training programme, or used insufficient intensities. Adequate interpretation of study results is not possible when key principles of the PRE are not followed. Further, earlier reported training programmes lasted 5 to 8 weeks but some have argued that a 6-week training programme may not be long enough to induce functional adaptations (Patikas, 2006). Therefore, the purpose of this study was to evaluate the effectiveness of a sufficiently long and intensive 12-week functional PRE strength training programme, by applying current guidelines (Faigenbaum, 2009). We hypothesize that this training would improve muscle strength and, subsequently, lead to improved mobility in children with CP, without increasing muscle spasticity.

METHODS

This study was conducted at Physiotherapy OPD of Jaipur Physiotherapy College, Mahraj vinayak global University, Jaipur on spastic unilateral or bilateral Cerebral Palsy children with physical disabilities. Full written informed consent was obtained from all parents and 12-year-old children before enrolment. The total 51 children were pre-stratified according to three stratification variables: sex, GMFCS level (I, II–III), age (youngest: 6–9y; oldest 10–13y), and subsequently randomized to one of two groups using sealed envelopes. Assessments were at baseline (T0), during training (T1; a subset of outcomes), directly after training (T2), and at the 6-week follow-up (T3). The measurement points of main interest were T2 and T3. Single-blind randomized controlled design. The control group continued their conventional physical therapy programme. Children in the control group received one to three sessions a week. The training group followed a 12-week progressive functional task training programme for the lower extremities. This replaced their conventional physical-therapy programme. Training was given three times a week for 45 to 60 minutes at Neuro Physioherapy unit of OPD in small groups (four or five children).

Each training session consisted of one exercise on a child-adapted leg-press (Enraf Nonius, Delft, the Netherlands) and three functional exercises (sit-to-stand, lateral step-up, half knee-rise), loaded with a weighted vest. During the training, intensity progressively increased, based on repeated estimation of the eight-repetition maximum. The primary outcome, ‘mobility’, was measured with the 66- item version of the Gross Motor Function Measure (GMFM- 66), the Sit-To-Stand (STS) test, and the Lateral Step-Up (LSU) test. The GMFM-66 is a standardized observational instrument, based on interval scaling (Russell, 2002). It reports on the child’s level of capacity, and has been internationally validated for evaluating change in the gross motor activities of children with CP. The STS test assesses the number of repetitions (full movement from standing up to sitting down) that the child can perform in 30 seconds on a child-sized chair with a height-adaptable seat (no backrest, no armrest). The LSU test assesses the number of repetitions (full movement from stepping up to stepping down) that the child can perform in 30 seconds on a 21cm (GMFM I–II) or 11cm (GMFCS III) step. Subjectively reported mobility was assessed with the 28- item version of the Dutch mobility questionnaire (MobQues- 28), which measures caregiver-reported mobility limitations.

It was found to be valid and reliable in a recent study in aDutch population (van Ravesteyn, 2010). The secondary outcome, ‘muscle strength’, was measured with (1) a six-repetition maximum test on a leg-press and (2) isometric strength tests of the hip flexor / abductor, knee flexor / extensor, and ankle plantar flexor muscles of the most affected leg, using a hand-held dynamometer (MicroFet; Biometrics, Almere, the Netherlands). At T1 only a subset of these outcomes were tested: the STS, LSU, and six-repetition maximum. The control outcome measure was spasticity, quantified on a scale of 0 to 5, calculated as the sum of a catch in response to a fast muscle stretch (0, no catch; 1, catch) in five leg muscles (adductor, rectus femoris, hamstrings, soleus, gastrocnemius). Scholtes et al.15 give detailed descriptions of all outcome measures and measurement protocols. Muscle soreness was scored weekly on a Likert scale (no; mild; moderate; severe; extremely severe).

Data Analysis

Baseline characteristics were analysed with the Student's t-test for continuous normally distributed data, a v2 test for dichotomous data, and a Mann–Whitney U test for ordinal data. A two-tailed value of $p<0.05$ was considered to be statistically significant. To investigate the effect of the strength training, we used generalized estimating equations for longitudinal analysis of continuous outcomes and log linear generalized estimating equation analysis for ordinal outcomes. This method takes into account the dependency of within-patient observations and the fact that not all children are assessed at each measurement point (missing data). All analyses were performed in SPSS, version 15 (SPSS Inc., Chicago, IL, USA). In the primary generalized estimating equation model, the outcome variable (e.g. GMFM-66, LSU, STS, hand-held dynamometer, six-repetition maximum, MobQues-28, or spasticity) was analysed as a dependent variable, with treatment allocation (1, training; 0, control) as the key independent variable adjusted for time. To evaluate whether the two groups differed in change over time, the interaction term (group*time) was added as an independent variable. We had four measurement points, so time was included as a dummy variable (reference was baseline), and three interaction terms were analysed (group 1*T1; group 1*T2; group 1*T3). All models were corrected for the school attended. We also analysed whether sex (1, male; 0, female), age, body mass index, limb distribution (1, unilateral; 0, bilateral), GMFCS level (dummies for levels I–III; reference, I), puberty onset (Tanner stages, dummies for early [stages 1–2], middle [stages 3–4], or late adolescence [stage 5]; reference, early), and problem behaviour (Strength and Difficulties Questionnaire,18 dummies for normal [scores 0–10], borderline [scores 11–13], or abnormal behaviour [scores 14–40]; reference, normal) were confounders of the training effect. A two-tailed value of $p<0.05$ was considered statistically significant.

RESULTS

Sixty-one patients were asked to participate; four turned out to be ineligible. Of the remaining 57 eligible patients, six did not give consent. The remaining 51 children were randomized. Of these, one dropped out before T0 (GMFCS level III, female, age 13y 1m, training) owing to a hip injury that made pretesting and training impossible, and one was lost to follow up at T1 (GMFCS level II, female, age 12y 1mo, training) owing to an unexpected long-term stay abroad. Analyses were performed for 49 participants. There was no statistically significant difference between the groups for personal characterstics. Training compliance and intensity of the 36 scheduled training sessions, three to six sessions were cancelled because of school-related activities. After correction for these, the mean compliance was 92.3% (range 71– 100%). A mean of 32 training sessions (range 30–33) were attended. Reasons for absence were illness (41.4%), medical appointment (8.6%), vacation (6.9%), or other / unknown (43.1%). Every 2 weeks, starting from week four, eight-repetition maximum tests were performed for leg-press and loaded STS exercises. Based on these eight-repetition maximum test values the training load progressed. During the last 8 weeks of training, the mean eight-repetition maximum strength increased from 116 to 149% on the leg-press, and from 28 to 41% on the loaded STS. Each week, one to six children reported mild to moderate muscle soreness.

DISCUSSION

This study hypothesized that a 12-week functional PRE strength training programme, in which appropriate guidelines for increasing muscle strength are applied, 13 improves muscle strength in children with CP, and subsequently leads to improved mobility, without any adverse effect on muscle spasticity. This 12-week functional PRE strength training resulted in statistically significant larger improvements (8%) in total hand-held isometric muscle strength in the training group than in the control group. Isolated significant improvements were found in the knee extensors (12%) and hip abductors (11%), but not in the knee, hip, and ankle plantar flexors. This seems to underline the importance of specificity in strength training; i.e. the exercise must be specific to the working of the muscle. Because the circuit training specifically targeted strengthening knee and hip extensors (with the leg-press, STS, forward step-up, and half-knee-rise exercises) and hip abductors (with the lateral step-up exercise), improvements in these target muscles were, therefore, to be expected. Unfortunately, isometric hip extension strength was not assessed, owing to this assessment's low reliability, as found in a pre-pilot study.

Two other randomized studies have evaluated isometric muscle strength, and found similar results, either in total muscle strength9 or isolated (targeted) muscle strength.8 The training also resulted in a statistically significant improvement (14%) in six-repetition maximum leg-press strength, compared with the control group. To our knowledge, no other study has assessed this outcome measure in a similar patient group, so it is impossible to compare these results with others. However, one randomized study10 did use the one-repetition maximum STS as an outcome. When adjusted for body weight, the strength increase in favour of the intervention group was comparable (approximately 15%). The results of this study show that training with a sufficient intensity, e.g. with a training load at which no more than 8 to 12 repetitions can be completed before muscular fatigue, results in increases in muscle strength. With this result we hope to fill the gap that was recently addressed in a literature discussion19 on the effectiveness of strength training on muscle strength, and stressing the need for new randomized controlled trials that apply appropriate guidelines. We feel that our study adds to this discussion and the current evidence 20 that PRE strength training is effective in increasing leg muscle strength in this group of patients. There was a detraining effect, resulting in loss of muscle strength at 6 weeks after the training ended, which was expected, and has also been observed in healthy children.21 Surprisingly, this effect was not observed in the few comparable (mostly uncontrolled) studies in children with CP3,9,12 with a follow-up assessment.

Based on the findings of our randomized controlled trial, we recommend that strength training should be included in a regular exercise routine to maintain increased strength levels.22 Unexpectedly, the 8 to 14% strength improvements were not accompanied by mobility improvements, according to the GMFM-66, the functional strength tests, and the mobility questionnaire. Two out of three comparable randomized controlled trials showed that their strength training resulted in small, but significant, functional improvements on the GMFM-66 in favour of the training group.8–10 So, despite the current popularity of strength training, the evidence from current randomized controlled trials is inconclusive on the effectiveness of strength training to improve mobility in children with CP. Possible explanations for the lack of mobility improvement in our study might be that the 8 to 14% improvements were too small to improve mobility, or that the number of individual muscles gaining strength was too limited. Therefore, future studies should focus on optimizing the training programme to reach maximum strength gains in children with CP. Besides this, we need to gain a better understanding of the longitudinal relation between muscle strength and mobility. Daily activities require only a certain amount (e.g. lowest threshold) of muscle strength. Increases above these lowest threshold values may be accompanied by increases in mobility, (Bohannon, 2007) but there might also be a point (e.g. highest threshold) at which further strength increases provide no additional advantage in mobility improvements (Bohannon, 2007). In that case, strength training would not be the appropriate choice of treatment when the aim is to improve mobility. Here, other components, such as balance and coordination, might influence mobility improvement to a greater degree than muscle strength alone. This should also be a subject for future research. In addition, the lack of effectiveness on mobility improvement found in this study might also be explained by the lack of variation in task contexts and individually tailored exercises. It is said that functional improvement is unlikely to occur unless the task to be learned is practised under various different contexts (Carr, 2003).

Even though our training contained functional exercises that resembled daily mobility activities (e.g. rising from a chair), the actual context of these exercises (i.e. chair height) remained unchanged throughout the training period. The lack of effectiveness on mobility improvement might also be explained by the non-individual specificity of the exercises. Selection of individually tailored mobility exercises with a more goal-oriented approach might improve the effectiveness of strength training on mobility outcomes (Papavasiliou, 2009). There was no change in spasticity (in either group) during, directly after, or 6 weeks after the training, confirming the results of similar studies (Morton, 2005) This supports the current belief that strength training for patients with spasticity is not contra-indicated. Children were informed that they could experience a mild to moderate degree of muscle soreness the day after the training. Muscle soreness was occasionally reported. This muscle soreness, rated as mild or moderate, was transient and never interfered with their training or daily life. Therefore, this cannot be interpreted as a contra-indication. Moreover, it might confirm that training intensity was sufficient to generate muscle overload. This study has some limitations. First, the groups differed slightly statistically, although non-significantly, in personal characteristics, but none of these were found to be confounders. Secondly, the intervention group performed better at baseline on all outcome measures, reaching statistical significance in the number of STS repetitions, isometric ankle plantarflexion, and total muscle and six-repetition maximum strength. Therefore, outcome measures were controlled for baseline differences. Another limitation was the unknown frequency and content of the usual physiotherapy of the control group. However, the therapists of the control group did not know the PRE training principles.

In conclusion, 12 weeks of functional PRE strength training is effective in increasing isometric muscle strength of the knee extensors and hip abductor by 11 to 12%, and increasing six repetition maximum leg-press strength by 14%. However, this does not result in an increase in mobility. Consequently, functional PRE strength training is indicated for a child with CP when the aim is to improve leg muscle strength. It could also be included in a more extensive treatment regime, addressing several elements of fitness including muscle strength, or used as a target treatment specifically anticipating temporary muscle weakness, such as before or after botulinum-toxin-A or surgical treatment.

Acknowledgments

We would like to thank all our well wishers who help us in this study and give us support and courage do this.

Funding: There was no funding source for this study.

Conflict of Interest: Nil.

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