Functional power-training in young children with cerebral palsy

Liesbeth van Vulpen

The work presented in this thesis was conducted at Amsterdam Rehabilitation Research Center | Reade, Amsterdam, The Netherlands and Amsterdam Movement Science, Department of Rehabilitation Medicine, VU University Medical Center, Amsterdam, The Netherlands.

The project presented in this thesis was funded by Duyvensz-Nagel Stichting, Stichting Mitialto, Revalidatiefonds, Dokter Izak Wessel Stichting, Koninklijk Nederlands Genootschap voor Fysiotherapie and Reade. The funders had no influence on the project content, the interpretation of the results, the final conclusions and their publication.

The printing of this thesis was financially supported by the Scientific College Physical Therapy (WCF) of the Royal Dutch Society for Physical Therapy (KNGF), the Nederlandse Vereniging voor Kinderfysiotherapeuten, Reade, CrossFit Aan 't IJ, ProCare, Phelps Foundation for Spastics, OIM Orthopedie, and Kamer Orthopedie.

Cover	Dorota Kozerska
Layout	Renate Siebes | Proefschrift.nu
Printed by	ProefschriftMaken
ISBN	978-94-90791-65-0

VRIJE UNIVERSITEIT

Functional power-training in young children with cerebral palsy

ACADEMISCH PROEFSCHRIFT

ter verkrijging van de graad Doctor
aan de Vrije Universiteit Amsterdam,
op gezag van de rector magnificus
prof.dr. V. Subramaniam,
in het openbaar te verdedigen
ten overstaan van de promotiecommissie
van de Faculteit der Geneeskunde
op dinsdag 26 juni 2018 om 13.45 uur
in de aula van de universiteit,
De Boelelaan 1105

door

Liesbeth Francien van Vulpen

geboren te Utrecht

promotor:	prof.dr. J.G. Becher
copromotoren:	dr. S. de Groot dr. A.J. Dallmeijer

Contents

Introduction

Ability to walk and run (i.e. walking capacity) is important for daily life activities of children, especially the short-term high explosive sprinting activities that are used in playing. Children with cerebral palsy (CP) are often integrated in community schools and recreational facilities but have difficulties with walking and sprinting activities because of their motor impairments. As a result, children with CP have problems in daily life with keeping up with peers in these activities. A key aspect of the decreased walking capacity in children with CP is the lower-limb muscle weakness that results from CP (1;2). Strength training programs, that have been developed to improve walking capacity, showed inconclusive evidence for improving walking, despite improvements in muscle strength (3-5). Recent insights have suggested that strength training with high movement velocity exercises might be more effective for improving walking than traditional strength training programs with low movement velocity exercises (6;7). A specific training program meeting this condition has been developed for children with CP, suitable from the age of 4 years.

Aim of this thesis is to evaluate the effectiveness of a functional high-velocity resistance training (further referred to as power-training) to improve muscle strength, walking capacity and parent-reported mobility performance of children with CP.

Cerebral palsy

Cerebral palsy (CP) is the most common cause of physical disability in pediatric rehabilitation, affecting about 1 in 500 children with an estimated prevalence of 17 million people worldwide (8). CP is the umbrella diagnosis for a group of permanent disorders in the development of movement and posture, attributed to non-progressive disturbances that have occurred in the developing fetal or infant brain (8;9). There are multiple causes for the brain disturbance, which can occur during prenatal (e.g. intoxication, infection), perinatal (e.g. hypoxic-ischemia), or postnatal (e.g. infection, trauma) period until the first birthday (8). CP is diagnosed based on its clinical manifestation as a motor disorder that causes limitations in activities and/or participation. The motor disorders are often accompanied by other impairments in body function, like disturbances of sensation, cognition, communication, behavior, by epilepsy, and by secondary musculoskeletal problems. The severity of the motor impairment, as well as the secondary impairments, are different for each person with CP, resulting in a large variability in functioning in all domains.

According the description of The Surveillance of Cerebral Palsy in Europe (SCPE), there are three main types of motor disorder — spastic, dyskinetic and ataxic CP (10). The three types of motor disorders with abnormal (pattern of) posture and/or movement are

defined as follows: 1) spastic CP is characterized by increased tone and/or pathological reflexes. *Bilateral* spastic CP involves limbs on both sides of the body, whereas in *unilateral* spastic CP, only the limbs on one side of the body are affected. 2) Dyskinetic CP is characterized by involuntary repetitive movement of body segments during rest or activities. 3) Ataxic CP is characterized by problems with muscle coordination, resulting in movements executed with abnormal force, rhythm, and accuracy (10). Almost always, there are mixed motor disorders with one dominant motor disorder. When there is no dominant motor disorder, it is classified as mixed. In about 80% of the children with CP, the spastic form is the predominant form (11;12). Children with unilateral or bilateral spastic CP were included in the studies described in this thesis.

The severity of the limitations in mobility can be classified by the Gross Motor Function Classification System (GMFCS) which has become the gold standard for classification of mobility level in children with CP. The GMFCS is an ordinal classification with a 5 level scale, in which different descriptors are used according to the age of the child (0–2, 2–4, 4–6, 6–12, 12–18 years). The descriptors for children 6–12 years of age are shown in Figure 1.1. (13;14). In the group of children with a spastic CP, 60 to 70% can walk with or without assistive mobility devices at the age of 6 to 12 years, which is GMFCS level I–III (15;16). Children with GMFCS level I and II were included in the studies described in this thesis.

Consequences of spastic cerebral palsy

To categorize the impact of the CP on the child's functioning, the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) is used (17). The ICF-CY is the current framework used in pediatric rehabilitation medicine for measuring health and disability (17). This framework serves to describe the consequences of a disorder at the domains of body functions and structures, activity and participation (Figure 1.2). The consequences of CP can be present at all domains of the ICF-CY model. The impaired muscle activation and impaired muscle strength are consequences at the domain body function and structures. The impairments in muscle function causes limitations in activities such as walking and running. Consequently, the ability to participate in daily life situations — like playing soccer with classmates in the schoolyard or joining classmates on foot in class trips — can be reduced.

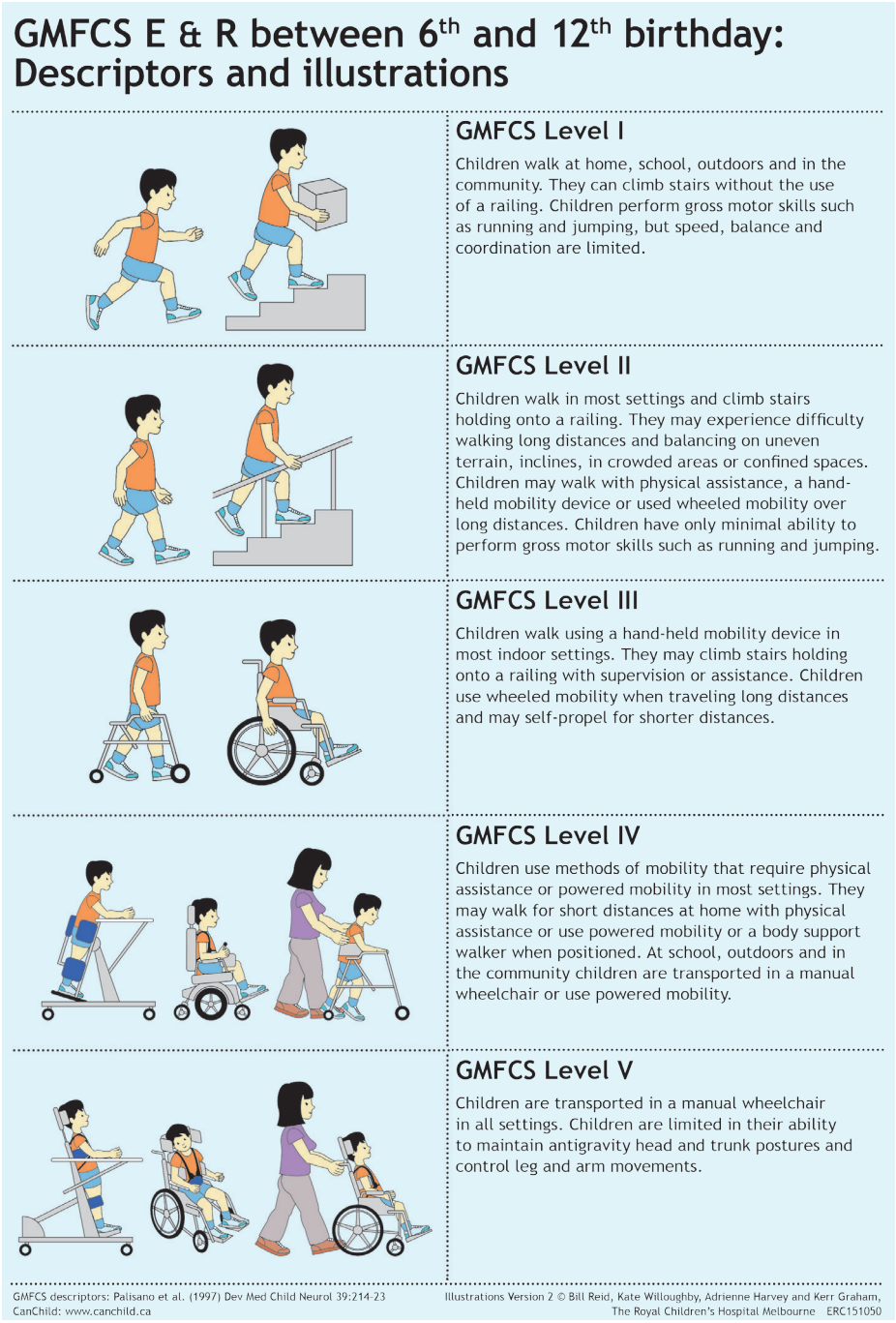

Figure 1.1 Gross Motor Function Classification System for children with cerebral palsy, 6–12 years of age.
Image is courtesy of B. Reid and A. Harvey, The Royal Children's Hospital, Melbourne, Victoria, Australia.

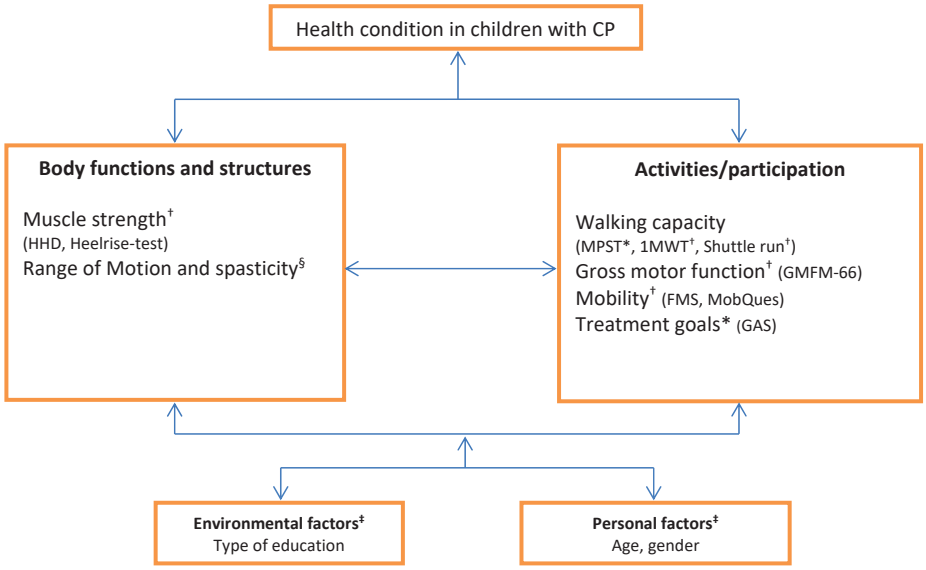

Figure 1.2 Study outcomes used on the different ICF levels: * primary outcomes; † secondary outcomes; § adverse outcomes; ‡ control outcomes.
Abbreviations: CP, cerebral palsy; HHD, hand-held dynamometer; GMFM, Gross Motor Function Measure; MPST, Muscle Power Sprint Test; 1MWT, 1-minute walk test; FMS, Functional Mobility Scale; MobQues, Mobility Questionnaire; GAS, Goal Attainment Scaling.

Decreased walking ability in cerebral palsy

We distinguish walking ability as a term for: A) the capacity to walk and run (i.e., at a certain speed and endurance); and B) the walking performance of the child (i.e., what a child actually walks in her/his daily environment) (18) (see Panel 1.1). Furthermore, we define three factors in walking capacity: 1) sprint capacity; 2) walking speed; and 3) walking or running endurance (Panel 1.1). Children with CP, GMFCS level I and II, have significant lower walking speed, sprint capacity, time duration and significant lower walking performance in comparison with their typically developing (TD) controls (8;19-23). These children are often integrated in community schools and recreational facilities, and thus are required to perform the same activities alongside their TD peers. They have problems with keeping up with peers in these activities because of their impaired walking ability (24).

In almost all childhood playing activities, such as games on the playground or sports such as football and racket sports, the child is more involved in short-term high-intensity sprint activities than in long-term running activities (25;26). In short-term playing activities,

sufficient sprint capacity and agility are extremely important for children, while children with CP have significant lower sprint capacity than their TD peers (22).

To be able to walk at a certain velocity makes it easier to integrate in a daily live school environment. Many distances in their school life are walked together with all the children in their class, like walking from classroom to playground, from classroom to gym class, and walking in school outings. As children with CP have a lower walking velocity than their peers special arrangements have to be made by teachers, parents and therapists, like an extra teacher or teacher-assistant in the class room that accompanies the child with CP to the gym class and/or stairs. Small increases in walking velocity can make a big difference in daily life situations, like making it possible to walk with all their classmates to the playground without any extra assistants.

In addition to the lower walking and sprinting velocity, children with CP have lower aerobic capacity compared with TD children (19;21;27). Aerobic capacity is needed to be able to walk and run longer distances. Children with CP use significant more energy for walking (i.e., higher energy cost) than TD children (28-32). When having a higher energy cost during walking with a reduced aerobic capacity, the physical strain of walking is consequently higher for children with CP (33). A high physical strain in walking activities leads to faster exhaustion, which is often a complaint of individuals with CP (34).

Panel 1.1 Definitions of terms used in this thesis

Walking ability
We define walking ability as a term for:
- the capacity to walk and run at a certain speed and time duration
- the walking performance of the child (i.e. what a child actually walks and runs in her/his daily environment)

Walking capacity
We define walking capacity as:
- walking speed
- sprint power
- walking / running endurance

Muscle strength
We define muscle strength as:
- the ability to exert force in an isometric or dynamic condition with a defined body position

Muscle weakness

Muscle weakness is reported as an important factor affecting walking capacity in children with CP (8). Walking children with CP only have about 36% to 82% of the lower-limb muscle strength of TD children (2;35). Previous studies have reported moderate to strong relationships between lower-limb muscle strength and walking capacity in children and young adults with CP, showing better walking capacity in those with stronger muscles (35-38).

Whereas CP results from a primary injury in the central nervous system, clinical symptoms are observed in the peripheral neuromuscular system — skeletal muscles in particular (8). Insufficient force generation has been attributed to decreased central activation or neuronal drive (2;39-42), inappropriate co-activation of antagonist muscle groups (39;42), and altered muscle architecture (42-45).

Greater weakness has been reported in the distal lower-extremity musculature, as compared with the proximal lower-extremity musculature (2;35). Especially the plantar flexor muscles show strength levels as low as 48% of the force of those in matched TD children (2). Likewise, children with CP produce less muscle power in the distal muscle groups (the plantar flexors) than in the more proximal muscle groups (39;42). Power production of plantar flexor muscles in the push-off phase in walking is important for step length and walking speed and these could, therefore, be decreased during gait in children with CP (46). Dallmeijer et al. (2011) showed that, in comparison with TD children, ankle power generated at push-off is indeed reduced in children with CP by more than 40% during gait (1).

Functional power-training

Several interventions have been studied to increase muscle strength to enhance the walking capacity of children with CP; the most commonly used method is progressive resistance exercise training (PRE) (4). However, despite an increase in strength in most lower-limb muscles, PRE did not improve the walking capacity of children with CP (3;4;47;48). Apparently the newly gained muscle strength does not transfer to functional improvements in walking. Moreau et al. suggested that to get functional improvements, the strength training velocity has to be at a higher, more functional movement velocity than generally used in PRE training (7). This suggestion is based on their findings that children with CP have a reduced capacity to rapidly generate forces (7). This capacity is particularly needed in playing activities such as running and sprinting games on the playground and in sports as pointed out before (22).

Another possible reason for the limited effects of PRE on walking capacity is lack of targeted training of the plantar flexor muscles (3;4;47;48). As mentioned above, this muscle group tends to weaken more than the proximal muscle groups and is important for generating power at push-off during walking. Therefore, addressing the plantar flexors in a strength training program could be important to improve in walking capacity.

Specificity of training is, according to the guidelines of the National Strength and Conditioning Association, one of the most basic concepts to incorporate in strength training programs (49). The term refers to the method in which a person is trained in a specific manner to produce a specific adaptation or training outcome (49). The acronym *SAID*, which stands for *Specific Adaptation to Imposed Demands*, is sometimes used to explain specificity in training. The underlying principle is that the type of demand placed on the body dictates the type of adaptation that will occur. Training at high-speed movements like sprinting, activates and recruits the same motor units required by the sprint (49).

The body's system must be overloaded beyond their normal levels for training adaptations (49). Overload refers to assigning a workout of greater intensity (the amount of resistance used) and training volume (total amount of work performed in a training session) than the person is accustomed to. Without the stimulus of overload, even an otherwise well-designed program greatly limits a person's ability to make improvements. The obvious application of this principle in the design of resistance training programs involves increasing the loads applied in the exercises (49). Other overload stimuli include for instance the number of sessions per week (see Figure 1.3) and the number of exercises or sets per training session (49). If these stimuli are applied over a period of time the body will adapt and this becomes its new norm.

To strengthen the plantar flexor muscles and to improve walking capacity in children with CP, we developed a functional power-training program that consists of resistance training with a high specificity to sprinting and walking and with a progressive intensity by using loaded functional exercises like running while dragging a loaded box and pushing a loaded chair as fast as possible. To control the progressiveness in the exercises we kept the distance (derived from the 70% of their maximal speed) and the exercise time (25s) the same and increased the load by 10% when the child became faster, i.e., was able to cover the distance in less than 25s.

We hypothesized that with increasing the specificity of strength training — i.e., a higher movement velocity in the progressive loaded strength training exercises that are incor-

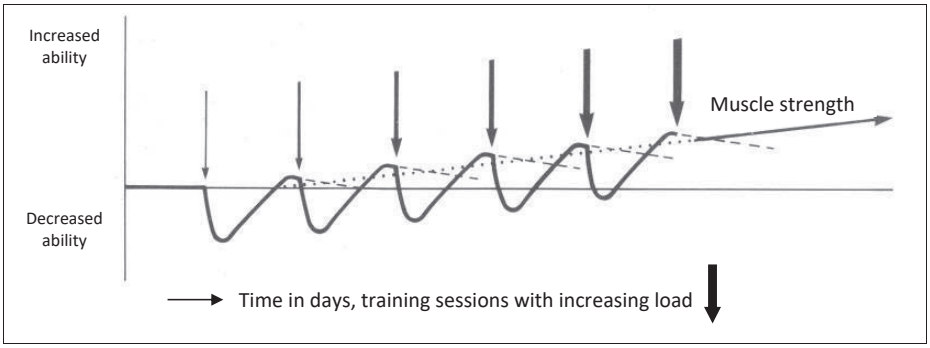

Figure 1.3 Principle of progressive overload in training – optimal improvement.

porated in functional movements like walking and sprinting — we could increase muscle strength and walking ability and subsequently participation in children with CP.

Aims and hypothesis of this thesis

Primary aim of this thesis is to evaluate the effectiveness of functional power-training to improve muscle strength, walking capacity (walking speed, sprint power, endurance) and parent-reported mobility performance of children with CP. First, we aim to investigate the feasibility and test-retest reliability of measuring lower-limb strength in young children with CP. Secondly, we evaluate the effectiveness of functional power-training to improve muscle strength, walking capacity and parent-reported mobility performance of children with CP. In addition, we investigate whether changes in lower-limb muscle strength can explain changes in walking capacity in young children with CP.

Our hypothesis is that functional power-training improves lower-limb muscle strength, walking capacity and parent-reported mobility performance in children with CP.

Outline of this thesis

Chapter two describes the feasibility and test-retest reliability of the measurement of lower-limb muscle strength and walking speed in young children with CP. Chapter three provides a detailed description of the design and methodology of the study to evaluate the effectiveness of the functional power-training on muscle strength and walking ability in children with CP. The effects of functional power-training on lower-limb muscle strength, walking capacity, and parent-reported mobility performance are provided in chapters four and five. In chapter six the relationships between the changes in lower-limb muscle

strength and the changes in walking capacity during the 14-weeks periods of usual care, functional power-training and follow-up is evaluated and the clinical implications are described. Finally, in chapter seven, the findings and conclusions of the studies described in the previous chapters are summarized and integrated in a discussion regarding clinical implications and the use of strength training in the treatment of children with CP. In addition, suggestions for further research are made.

References

(1) Dallmeijer AJ, Baker R, Dodd KJ, Taylor NF. Association between isometric muscle strength and gait joint kinetics in adolescents and young adults with cerebral palsy. Gait Posture 2011 Mar;33(3):326-32.

(2) Wiley ME, Damiano DL. Lower-extremity strength profiles in spastic cerebral palsy. Dev Med Child Neurol 1998 Feb;40(2):100-7.

(3) Franki I, Desloovere K, De CJ, Feys H, Molenaers G, Calders P, et al. The evidence-base for conceptual approaches and additional therapies targeting lower limb function in children with cerebral palsy: a systematic review using the ICF as a framework. J Rehabil Med 2012 May;44(5):396-405.

(4) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014 Feb;35(2):239-49.

(5) Reedman S, Boyd RN, Sakzewski L. The efficacy of interventions to increase physical activity participation of children with cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol 2017 Oct;59(10):1011-8.

(6) Moreau NG, Falvo MJ, Damiano DL. Rapid force generation is impaired in cerebral palsy and is related to decreased muscle size and functional mobility. Gait Posture 2012 Jan;35(1):154-8.

(7) Moreau NG, Holthaus K, Marlow N. Differential adaptations of muscle architecture to high-velocity versus traditional strength training in cerebral palsy. Neurorehabil Neural Repair 2013 May;27(4):325-34.

(8) Graham HK, Rosenbaum P, Paneth N, Dan B, Lin JP, Damiano DL, et al. Cerebral palsy. Nat Rev Dis Primers 2016;2:15082.

(9) Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl 2007 Feb;109:8-14.

(10) Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). Dev Med Child Neurol 2000 Dec;42(12):816-24.

(11) Himmelmann K, Beckung E, Hagberg G, Uvebrant P. Bilateral spastic cerebral palsy--prevalence through four decades, motor function and growth. Eur J Paediatr Neurol 2007 Jul;11(4):215-22.

(12) Himmelmann K, McManus V, Hagberg G, Uvebrant P, Krageloh-Mann I, Cans C. Dyskinetic cerebral palsy in Europe: trends in prevalence and severity. Arch Dis Child 2009 Dec;94(12):921-6.

(13) Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol 1997 Apr;39(4):214-23.

(14) Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. Dev Med Child Neurol 2008 Oct;50(10):744-50.

(15) Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. Pediatrics 2008 Jan;121(1):e187-e192.

(16) Westbom L, Hagglund G, Nordmark E. Cerebral palsy in a total population of 4-11 year olds in southern Sweden. Prevalence and distribution according to different CP classification systems. BMC Pediatr 2007 Dec 5;7:41.

(17) International Classification of Functioning, Disability and Health for Children and Youth. Geneva: World Health Organisation WHO; 2008.

(18) Holsbeeke L, Ketelaar M, Schoemaker MM, Gorter JW. Capacity, capability, and performance: different constructs or three of a kind? Arch Phys Med Rehabil 2009 May;90(5):849-55.

(19) Balemans AC, van WL, De Heer SJ, Van den Brink J, De Koning JJ, Becher JG, et al. Maximal aerobic and anaerobic exercise responses in children with cerebral palsy. Med Sci Sports Exerc 2013 Mar;45(3):561-8.

(20) van WL, Dallmeijer AJ, Balemans AC, Zhou C, Becher JG, Bjornson KF. Walking activity of children with cerebral palsy and children developing typically: a comparison between the Netherlands and the United States. Disabil Rehabil 2014;36(25):2136-42.

(21) Verschuren O, Takken T. Aerobic capacity in children and adolescents with cerebral palsy. Res Dev Disabil 2010 Nov;31(6):1352-7.

(22) Verschuren O, Maltais DB, Douma-van RD, Kruitwagen C, Ketelaar M. Anaerobic performance in children with cerebral palsy compared to children with typical development. Pediatr Phys Ther 2013;25(4):409-13.

(23) Wilson NC, Mackey AH, Stott NS. How Does the Functional Mobility Scale Relate to Capacity-Based Measures of Walking Ability in Children and Youth with Cerebral Palsy? Phys Occup Ther Pediatr 2013 May 8.

(24) Bax MC, Flodmark O, Tydeman C. Definition and classification of cerebral palsy. From syndrome toward disease. Dev Med Child Neurol Suppl 2007 Feb;109:39-41.

(25) Bailey RC, Olson J, Pepper SL, Porszasz J, Barstow TJ, Cooper DM. The level and tempo of children's physical activities: an observational study. Med Sci Sports Exerc 1995 Jul;27(7):1033-41.

(26) Van PE, Dore E. Short-term muscle power during growth and maturation. Sports Med 2002;32(11):701-28.

(27) Rose J, Gamble JG, Medeiros J, Burgos A, Haskell WL. Energy cost of walking in normal children and in those with cerebral palsy: comparison of heart rate and oxygen uptake. J Pediatr Orthop 1989 May;9(3):276-9.

(28) Bolster EAM, Balemans ACJ, Brehm MA, Buizer AI, Dallmeijer AJ. Energy cost during walking in association with age and body height in children and young adults with cerebral palsy. Gait Posture 2017 May;54:119-26.

(29) Brehm MA, Becher J, Harlaar J. Reproducibility evaluation of gross and net walking efficiency in children with cerebral palsy. Dev Med Child Neurol 2007 Jan;49(1):45-8.

(30) Dallmeijer AJ, Brehm MA. Physical strain of comfortable walking in children with mild cerebral palsy. Disabil Rehabil 2011;33(15-16):1351-7.

(31) Johnston TE, Moore SE, Quinn LT, Smith BT. Energy cost of walking in children with cerebral palsy: relation to the Gross Motor Function Classification System. Dev Med Child Neurol 2004 Jan;46(1):34-8.

(32) Thomas SS, Buckon CE, Russman BS, Sussman MD, Aiona MD. A comparison of the changes in the energy cost of walking between children with cerebral palsy and able-bodied peers over one year. J Pediatr Rehabil Med 2011;4(3):225-33.

(33) Balemans AC, Bolster EA, Brehm MA, Dallmeijer AJ. Physical Strain: A New Perspective on Walking in Cerebral Palsy. Arch Phys Med Rehabil 2017 Dec;98(12):2507-13.

(34) Berrin SJ, Malcarne VL, Varni JW, Burwinkle TM, Sherman SA, Artavia K, et al. Pain, fatigue, and school functioning in children with cerebral palsy: a path-analytic model. J Pediatr Psychol 2007 Apr;32(3):330-7.

(35) Dallmeijer AJ, Rameckers EA, Houdijk H, De GS, Scholtes VA, Becher JG. Isometric muscle strength and mobility capacity in children with cerebral palsy. Disabil Rehabil 2015 Nov 25;1-8.

(36) Eek MN, Beckung E. Walking ability is related to muscle strength in children with cerebral palsy. Gait Posture 2008 Oct;28(3):366-71.

(37) Ferland C, Lepage C, Moffet H, Maltais DB. Relationships between lower limb muscle strength and locomotor capacity in children and adolescents with cerebral palsy who walk independently. Phys Occup Ther Pediatr 2012 Aug;32(3):320-32.

(38) Ross SA, Engsberg JR. Relationships between spasticity, strength, gait, and the GMFM-66 in persons with spastic diplegia cerebral palsy. Arch Phys Med Rehabil 2007 Sep;88(9):1114-20.

(39) Elder GC, Kirk J, Stewart G, Cook K, Weir D, Marshall A, et al. Contributing factors to muscle weakness in children with cerebral palsy. Dev Med Child Neurol 2003 Aug;45(8):542-50.

(40) Fowler EG, Kolobe TH, Damiano DL, Thorpe DE, Morgan DW, Brunstrom JE, et al. Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings. Phys Ther 2007 Nov;87(11):1495-510.

(41) Rose J, McGill KC. Neuromuscular activation and motor-unit firing characteristics in cerebral palsy. Dev Med Child Neurol 2005 May;47(5):329-36.

(42) Stackhouse SK, Binder-Macleod SA, Lee SC. Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. Muscle Nerve 2005 May;31(5):594-601.

(43) Barber L, Barrett R, Lichtwark G. Passive muscle mechanical properties of the medial gastrocnemius in young adults with spastic cerebral palsy. J Biomech 2011 Sep 2;44(13):2496-500.

(44) Gough M, Shortland AP. Could muscle deformity in children with spastic cerebral palsy be related to an impairment of muscle growth and altered adaptation? Dev Med Child Neurol 2012 Jun;54(6):495-9.

(45) Willerslev-Olsen M, Lorentzen J, Sinkjaer T, Nielsen JB. Passive muscle properties are altered in children with cerebral palsy before the age of 3 years and are difficult to distinguish clinically from spasticity. Dev Med Child Neurol 2013 Jul;55(7):617-23.

(46) Gage J, Schwartz M, Koop S, Novacheck T. The identification and treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009.

(47) Scholtes VA, Becher JG, Janssen-Potten YJ, Dekkers H, Smallenbroek L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. Res Dev Disabil 2012 Jan;33(1):181-8.

(48) Taylor NF, Dodd KJ, Baker RJ, Willoughby K, Thomason P, Graham HK. Progressive resistance training and mobility-related function in young people with cerebral palsy: a randomized controlled trial. Dev Med Child Neurol 2013 Sep;55(9):806-12.

(49) Baechle TR, Earle RW. Essentials of Strength training and conditioning. third ed. Champaign, Il 61825-5076: Human Kinetics; 2008.

1

Feasibility and test-retest reliability of measuring lower-limb strength in young children with cerebral palsy

Liesbeth F. van Vulpen
Sonja de Groot
Jules G. Becher
Sander G. de Wolf
Annet J. Dallmeijer

2

Eur J Phys Rehabil Med 2013 Dec;49(6):803-13

Abstract

Background Quantifying leg muscle strength in young children with cerebral palsy (CP) is essential for identifying muscle groups for treatment and for monitoring progress.

Aim To study the feasibility, intratester reliability and the optimal test design (number of test occasions and repetitions) of measuring lower-limb strength with handheld dynamometry (HHD) and dynamic ankle plantar flexor strength with the standing heel-rise (SH) test in 3–10 year aged children with CP.

Design Test-retest design.

Setting Rehabilitation centre, special needs school for children with disabilities, and university medical centre.

Methods Knee extensor, hip abductor and calf muscle strength was assessed in 20 ambulatory children with spastic CP (3–5 years (n=10) and 6–10 years (n=10)) on two test occasions. Intraclass correlation coefficients (ICC) and Smallest Detectable Differences (SDD) were calculated to determine the optimal test design for detecting changes in strength.

Results All isometric strength tests had acceptable SDDs (9–30%), when taking the mean values of 2–3 test occasions (separate days) and 2–3 repetitions. The one-leg SH test had large SDDs (40–128% for younger group, 23–48% for older group).

Conclusion Isometric strength (improvements) can only be measured reliably with HHD in young children with CP when the average values over at least 2 test occasions are taken. Reliability of the SH test is not sufficient for measuring individual changes in dynamic muscle strength in the younger children.

Clinical rehabilitation impact Results of this study can be used to determine the optimal number of test occasions and repetitions for reliable HHD measurements depending on expected changes, muscle group and age in 3–10 year old children with CP.

Introduction

Cerebral palsy (CP) comprises of a group of disorders in the development of movement and posture, attributed to non-progressive disturbances that have occurred in the developing fetal or infant brain (1). Prevalence rates of CP are about 2 per 1000 births in Europe (2) of which 82% has a spastic CP (3;4). Motor impairment in CP is multi-factorial and includes problems such as spasticity, coordination problems, loss of selective motor control and muscle weakness (5). A Canadian study of gross motor development showed improvement of motor function at early ages until around 5 years, depending on Gross Motor Function System (GMFCS) level, when an average of 90% of gross motor capacity has been reached (4;5).

The ability to walk is of major concern for the families of children with CP (4). In the group of children with a spastic CP, 60–70% has achieved walking ability with or without assistive mobility devices between 6–12 years, which is level I-III according to the GMFCS (4;6). In order to walk without assistive devices, stability in the stance phase and propulsive power in the ankle are important factors. In able-bodied persons, the highest generation of power during the entire gait cycle is produced by the plantar flexor muscles during the end of push-off, which is the period between heel rise and toe off (7;8). The plantar flexors account for about 50% of the propulsive force in walking in able-bodied persons (7). In children with CP it is important to establish an optimal gait pattern within the constraints of their motor impairments (9). With the muscle weakness around the ankle, knee and hip, and the important role of these muscles in relation to gait pattern and walking ability (7), there is a need to understand the role of muscle strength when planning interventions such as physical training, orthotic treatment, botulinum toxin treatment and orthopaedic surgery for children with CP. This especially applies to children below 7 years who are still developing their gait pattern (10;11).

Quantifying muscle strength is essential for identifying muscle groups that need to be targeted in treatment, determining intensity of training based on the maximum capacity of the identified muscle group, monitoring progress and adjusting exercise programs. It is therefore necessary to have reliable and feasible methods to measure muscle strength, which can be used in clinical practice for young children with CP.

A hand-held dynamometer (HHD) is a practical way for clinicians to measure isometric muscle strength. Limited research has been done on test-retest reliability in children with CP (12-14). Only two studies examined the test-retest reliability of ankle plantar flexors (13;14). Overall, results vary by muscle group and test-retest reliability in children with CP below 6 years is unknown.

Where ICC reflects the test ability to differentiate among children, the standard error of measurement (SEM) quantifies the precision of individual test scores within the subject (15). For monitoring clinical progress, a change in strength needs to be larger than the smallest detectable difference (SDD), which is directly derived from the SEM (16). Two out of the three studies on test-retest reliability with HHD studied both the ICC and SEM. Despite good reliability coefficients these above-mentioned studies found high SEM (12;13), indicating that only large changes in muscle strength can be measured. SEM can be reduced by taking the average value over repeated measures. When using HHD in research and clinical practice it is now common practice to take the average over two or three repetitions in one test occasion. Crompton et al. (2007) showed that ICC values for different test occasions were lower and 95% confidence intervals wider compared with ICC-values for repeated measures (repetitions) in one test occasion in children with CP from 6 years and older (13). In the current study the most optimal test design (number of occasions and repetitions) for minimizing measurement error was assessed, taking into account the age of the children and the muscle group to be tested, to be able to detect changes in strength in children with CP.

Although isometric strength testing with the HHD is often used in studies it is only indicative of the capacity to produce force in an isometric condition and at that particular muscle length. These results can not be extrapolated to dynamic conditions (17). To measure ankle plantar flexor strength in a dynamical way a standing heel-rise test can be used (18;19). In a plantar flexor strength training study with children with CP the unilateral standing heel-rise test was used to measure the effect of the training (19). The standing heel-rise test on one leg has a good reliability in healthy adults (20;21), but reliability has not been investigated in children with CP.

The first purpose of this study was to investigate the feasibility and test-retest reliability of: a) isometric muscle strength of the plantar flexor, hip abductor and knee extensor with HHD, and b) dynamic plantar flexor strength assessed with standing heel-rise test, in ambulatory children with spastic CP in the age of 3–5 and 6–10 years old. The second purpose was to determine the optimal test design (number of measurements) based on the SEM and SDD, by conducting a Decision-study for 3x3 hypothetical test designs with 1 to 3 test occasions on separate days and 1 to 3 repetitions at one occasion.

Method

Participants

A convenience sample of 10 participants in the age of 3–5 years (mean age 4.9 (SD 0.7)) and 10 participants in the age of 6–10 years (mean age 8.2 (SD 1.4)), with a diagnosis of unilateral (n=11) or bilateral (n=9) spastic CP were recruited from a rehabilitation centre, a special needs school for children with disabilities, and a university medical centre. The inclusion criteria were: diagnosis of predominantly spastic types of CP, able to follow simple instructions, being ambulant without assistive devices (GMFCS level I and II) and aged between 3–10 years. Exclusion criteria were: serial casting of lower limb less than 3 months before the test, treatment with botulinum toxin A in lower limb less than 6 months before the test or lower-limb orthopaedic surgery in the last 18 months before the test. The Medical Ethics Committee of the VU University Medical Center in Amsterdam approved this study and written informed consent was obtained from the parents of each participant. Characteristics of the participants are shown in Table 2.1.

Procedure

Participants were assessed on two test occasions, within three weeks with the same conditions and time of day. The muscle strength tests (HHD, standing heel-rise tests) were performed on both test occasions to determine test-retest reliability. The examiner (LvV) was a paediatric physiotherapist with 15 years working experience with children with CP and trained in using HHD. In addition, tests to assess motor selectivity, spasticity and range of motion (ROM) were performed once.

Measurements

Isometric muscle strength was measured of the hip abductors, knee extensors and ankle plantar flexors with knee extended and knee in 90 degree flexion, with a hand-held dynamometer (microFET Hand-held Dynamometer, Biometrics BV, Almere, The Netherlands). The make-method, where the child gradually builds up force against the dynamometer for about 5 s, was used. The make-method was found more reliable than the break-method in an inter-tester reliability study of strength measurement using HHD in older CP children (22).

Participants were allowed one or two practice trials for each test until the investigator was confident that the child understood the task. Each child performed subsequently

Table 2.1 Characteristics of the participants

		3–5 years	6–10 years
Sex, n	Boy	1	5
	Girl	9	5
Age, y	Mean (SD)	4.9 (0.7)	8.2 (1.4)
	Range	3.7–5.7	6.0–10.7
Body mass, kg	Mean (SD)	19.1 (2.8)	27.6 (7.5)
	Range	15.0–24.5	20.0–42.0
Height, m	Mean (SD)	1.1 (0.1)	1.3 (0.1)
	Range	1.0–1.2	1.2–1.6
GMFCS[1]	I	5	8
	II	5	2
SCPE[2] classification	Bilateral spastic CP	5	4
	Unilateral spastic CP	5	6
Most impaired side[3]	Right side	6	6
	Left side	4	4
Ankle ROM[4],°	Most impaired, mean(SD)	17.2 (7.1)	14.0 (9.9)
	Range	10–30	0–25
	Less impaired, mean(SD)	26.7 (8.3)	23.0 (4.8)
	Range	15–45	15–30

[1] GMFCS Gross Motor Function Classification System (32).
[2] SCPE Surveillance of Cerebral Palsy (2).
[3] Most impaired side was determined by using the modified Trost SMC test for selective motor control for the ankle dorsal flexion possibility (7;25).
[4] ROM Range Of Motion ankle dorsal flexion with knees in extension in degrees °.

three repetitions for each muscle group and the maximum force (peak force) for each repetition was registered. When the concentration and/or motivation of the child were not optimal a fourth or fifth repetition was performed. Strong verbal encouragement during the repetitions was given to produce maximal effort. Each child was tested on a second occasion, 4 to 20 days later, by the same examiner, blinded from the strength values from the first test occasion. Test positions were standardized according to procedures used in previous studies (12;14;23) (Table 2.2). Lever arm was measured between standardized landmarks (Table 2.2) with a hard tape measure. Torque (Nm) is calculated by multiplying force (Newton) by the length (meter) of the lever arm (24).

Standing heel-rise; the number of repetitions (with a maximum of 20) for standing heel-rise on both limbs, and one limb, was measured based on a standardized protocol. An infra-red beam was placed lateral to the heels of the child (Figure 2.1). The infra-red

Table 2.2 Strength testing procedure using hand-held dynamometer

Muscle group	Position	Stabilization	HHD placement	Distance from axis of rotation to HHD
Hip abductor (HA)	Supine, hips at 45 degrees, knees extended	Pelvis stabilized using a belt, Contra lateral lower limb held in neutral manually	5 cm proximal from joint axis femoral epicondyl	Distance from trochanter major to location HHD
Knee extensor (KE)	Sitting, hips and knees at 90 degrees	Pelvis stabilized in the chair using a belt, lumbar stabilization by adjusting back of the chair	Anterior tibia, 5 cm proximal from bimalleolar line	Distance from knee joint line, lateral side, to location HHD
Ankle plantar flexor (GSTR)	Supine, hips at 45 degrees, knees extended, ankle in plantar grade position	Pelvis stabilized using a belt, manual stabilization on lower limb	Metatarsal heads	Distance lateral malleolus to location HHD
Ankle plantar flexor (SOL)	Supine, hips and knees at 90 degrees resting on bench, ankle in plantar grade position	Pelvis stabilized using a belt, manual stabilization on lower limb	Metatarsal heads	Distance lateral malleolus to location HHD

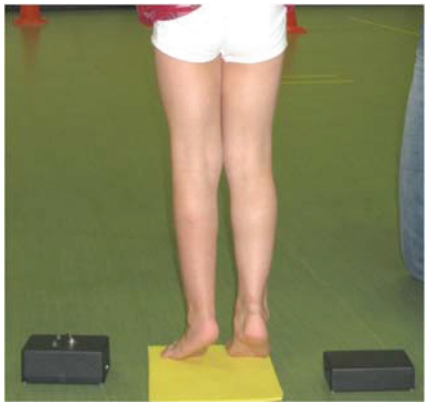

Figure 2.1 Standing heel-rise both limbs.

light barrier was 1.7 cm from the ground. When the child made a standing heel-rise the beam connected to the receiver and gave an auditory signal. The examiner counted the number of signals. The children were allowed to touch the examiner only with a single finger for balance. The test was terminated when the child leaned or pushed down on

the examiner, the child's knees flexed, or when the child gave up or asked to stop despite encouragement.

Selective motor control of knee extension and ankle dorsal flexion were tested by the muscle selectivity grading scale with three levels of control: 0, no ability to isolate movement; 1, partial ability to isolate movement; and 2, complete ability to isolate movement (7;25). Test positions are described by Gage et al. (7). The outcome of the test was used to determine the most impaired side of the child.

Range of motion (ROM) of the ankle included maximal dorsal flexion with knee flexed and extended to differentiate between gastrocnemius and soleus muscle contracture as demonstrated in the Silverskiöld test (7). Severe contractures of the ankle dorsiflexion (<0 degree of dorsal flexion) were expected to decrease the feasibility of the measurement.

Body mass and height; Each child was weighed on a mechanical scale and height was measured with a height scale while standing against the wall.

Data analysis

Feasibility

Feasibility of the measurements was evaluated by studying the convenience of the tests for the child, operationalized as the time necessary to complete all tests per test occasion, the number of times and reasons that a child was not able to perform the test, and whether more than 3 repetitions were necessary for the measurement with HHD.

Test-retest reliability

Test-retest reliability was assessed in a Generalization study (G-study) and a Decision study (D-study), which is based on analysis of variance (ANOVA) (15;16).

G-study; Variance component analyses, with a random-effects design and the method of restricted maximum likelihood, was carried out to obtain the variance components. Variance components were calculated for subjects (var_s), test occasion (separate days) (var_o), repetitions (on same test occasion) (var_r), interaction between subject-occasion (var_{so}), subject-repetition (var_{sr}), occasion-repetition (var_{or}), and the residual error variance (varsor,e) for all HHD outcome measures. For the standing heel-rise test, variance components were calculated for var_s, var_o, and their interactions only. ICC was calculated as the ratio of variance between subjects to the variance between subjects plus error variance according to: ICC = vars / (var_s + var_o + var_r + var_{so} + var_{sr} + var_{or} + $var_{sor,e}$) (15;16). SEM was calculated according to: SEM = $\sqrt{}$(var_o + var_r + var_{so} + var_{sr} +

var_{or} + $var_{sor,e}$) and SDD was calculated as SDD = 1.96*SEM*√2 (15). ICCs above 0.75 are indicative of good reliability, and those below 0.75 of poor to moderate reliability (26).

D-study; A D-study was performed to examine the effect of test design, by taking the average values over repeated test occasions on separate days and repetitions at 1 occasion, on SEM and SDD (15;16). Based on this method, SEM and SDD values were calculated for 3 x 3 hypothetical study designs with 1–3 test occasions on separate days and 1–3 repetitions at one occasion, by dividing the variance components by the number of occasions or repetitions (15;16). The SEM for a study design with 2 occasions and 3 repetitions, for example, was calculated by dividing the variance related to occasion by 2, variance related to repetition by 3, and the interaction terms are divided by 2 x 3. The SDDs were also expressed as a percentage of the average group value (15).

Results

One child of the 6–10 years group dropped out. Due to illness and family problems this child could not be tested a second time. This child was excluded from the analysis. Mean isometric strength values, averaged over 3 repetitions, and dynamic plantar flexor strength measured with the standing heel-rise test (SH) are listed in Table 2.3. The average torque varied from 3.6 Nm for the soleus muscle of the most impaired limb (SOL_{most}) to 25.8 Nm for the knee extensor muscles of the less impaired limb (KE_{less}) in the age group 3–5 years, and 4.3 Nm (SOL_{most}) to 54.0 Nm (KE_{less}) in the age group 6–10 years.

Feasibility

The isometric muscle strength instructions were easily understood by the children except for one child in the younger age group. The children were eager to perform the tests and were interested in how strong they were. There was, however, one boy in the older age group who was not willing to perform all the strength tests and it was, therefore, not possible to measure the soleus muscle on the second test occasion. There were no children with severe contractures in the ankle joints (<0 degree dorsal flexion) and the tester had therefore no problems to get the ankle in the required test position.

In the group of 3–5 years old there was a need for a fourth repetition (for two muscle groups) in 50% of the cases. This was 40% in the older age group. Reasons were a lack of concentration or not performing the movement correctly. After practice, the tests were easy to conduct for the assessor, although practice sessions are required to get used to handling and stabilizing the HHD during the tests.

Table 2.3 Mean values of Torque (Nm) measured with HHD and standing heel-rise

		3–5 y		6–10 y	
		Occasion 1 (n=10)	Occasion 2 (n=10)	Occasion 1 (n=10)	Occasion 2 (n=9)
HA_{most}[1] (Nm)	Mean[2] (SD)	9.6 (3.7)	9.7 (3.0)	24.8 (8.9)	23.3 (8.4)
	Range	5.7–17.4	6.2–15.5	11.4–39.3	12.1–37.9
HA_{less} (Nm)	Mean (SD)	9.1 (3.3)	9.1 (3.2)	24.7 (8.6)	24.0 (9.4)
	Range	4.1–15.5	4.3–15.7	12.0–39.3	11.8–40.0
KE_{most} (Nm)	Mean (SD)	14.0 (5.5)	13.3 (4.0)	33.3 (8.7)	32.5 (10.6)
	Range	8.6–24.6	9.3–21.7	22.1–48.5	19.3–49.4
KE_{less} (Nm)	Mean (SD)	14.7 (5.7)	16.0 (4.3)	39.9 (8.8)	40.2 (11.2)
	Range	8.6–25.8	10.7–23.0	25.8–53.4	25.1–54.0
$GSTR_{most}$ (Nm)	Mean (SD)	7.9 (3.5)	8.5 (3.5)	20.1 (5.7)	17.5 (5.8)
	Range	4.7–14.0	5.4–16.8	9.5–27.9	8.6–24.8
$GSTR_{less}$ (Nm)	Mean (SD)	12.2 (3.8)	11.9 (4.4)	25.9 (6.5)	22.6 (7.3)
	Range	8.3–19.2	6.7–18.5	15.2–33.8	11.9–35.9
SOL_{most} (Nm)	Mean (SD)	7.1 (3.8)	6.7 (3.0)	12.5 (5.6)	11.0 (4.5)[3]
	Range	3.6–15.8	4.0–14.5	5.9–20.0	4.3–18.0
SOL_{less} (Nm)	Mean (SD)	9.6 (4.7)	8.4 (4.4)	17.8 (5.7)	13.9 (4.5)
	Range	4.3–21.0	4.7–19.9	6.2–24.7	5.0–19.9
SH BothLimbs[5] (number)	Median	18	17.5	20	20
	Range	4–20	7–20	10–20	10–20
	Floor–ceiling[6]	n=1–n=4	n=1–n=5	n=1–n=9	n=1–n=8
SH most[7] (number)	Mean (SD)	2.33 (3.1)	1.62 (2.2)	4.5 (5.6)	4.44 (4.8)
	Range	0–8	0–6	0–16	0–20
	Floor–ceiling	n=5–n=1	n=4–n=1	n=4–n=1	n=3–n=1
SH less (number)	Mean (SD)	9.33 (6.7)	6.88 (5.9)	13.1 (6.2)	15.56 (6.7)
	Range	0–20	0–14	0–20	0–20
	Floor–ceiling	n=1–n=1	n=2–n=1	n=1–n=3	n=1–n=5

[1] HA, hip abductors; KE, knee extensors; GSTR, ankle plantar flexors knees extended (gastrocnemius); SOL, ankle plantar flexors knees 90° flexed (soleus); HAmost, most impaired limb; HAless, less impaired limb.
[2] Mean of 3 repetitions per muscle group.
[3] n=8 for ankle plantar flexors with knees flexed for most impaired and less impaired limb (SOLmost and SOLless).
[4] SH both limbs = number of repetitions for standing heel-rise on both limbs.
[5] Floor and ceiling effect = number of children who scored the minimum (floor) and who scored the maximum (20, ceiling).
[6] Standing heel-rise on the most impaired limb.

The Standing heel-rise test was easy to understand for all the children. The auditory signal was a good motivation for the children to give their best performance. It was, however, difficult for the children to keep their balance, i.e. not to lean on the examiner and to keep their knees extended. In the SH on both limbs all children of the older age group had the maximum score of 20 repetitions on both test occasions, except one child that scored 10 repetitions on both occasions.

Reliability

Results of the reliability analysis of the HHD assessments of the most impaired limb are listed in Table 2.4a and Table 2.4b, showing ICC, SEM and SDD for 1 single measurement (row 1) and for different test designs when the average is taken over 2 or 3 repetitions, in combination with 1–3 test occasions on separate days. The results of the less impaired limb (not shown) were similar to the results of the most impaired limb.

The ICC values were all above 0.77 for a single measurement, which indicates good reliability. The SEM and SDD for a single measurement were, however, large for all muscle groups (SDD: 19–63%). The SEM and SDD for the hip abductors (HA) and knee extensor (KE) were smaller (SDD: 19–45%) compared to the calf muscles (GSTR and SOL, SDD: 42–63%). All SEM and SDD values were lower in the older age group compared to the younger children for all assessed muscle groups.

The results of the different test designs for HHD measurements show that the SEM and SDD reduced more by taking the average over more occasions on separate days than by taking the average over more repetitions in 1 test occasion. In addition, results show that SEM and SDD improved only slightly when the average over 3 instead of 2 repetitions were taken.

There was a ceiling effect for the SH on both limbs and, therefore, no analysis was carried out with these data. ICC values of the unilateral SH ranged from 0.86 to 0.89 in the younger age group and from 0.87 to 0.98 in the older age group, which are indicative for good reliability (Table 2.5) when tested in only 1 test occasion. The SEM and SDD are high in both groups, especially in the younger age group (SDD: 81% for less impaired limb and 128% for most impaired limb).

Results in the 3–5 year group showed that the SEM and SDD remain high even after increasing the number of test occasions from 1 to 3 and taking the mean over these test occasions (SDD reduction from 128% to 74%).

Table 2.4a Reliability ordered by number of test occasions (Occ) and repetitions (Rep), of isometric muscle strength measurement for the most impaired limb (D-study)

Age group 3–5 years																	
		HA_{most}				KE_{most}				$GSTR_{most}$				SOL_{most}			
Occc	Rep	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD* %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %
1	1	0.85 (0.76–0.91)	1.4	3.8	39%	0.80 (0.69–0.88)	2.2	6.1	45%	0.86 (0.78–0.91)	1.36	3.8	46%	0.81 (0.71–0.88)	1.57	4.4	63%
1	2	0.89 (0.82–0.93)	1.1	3.1	32%	0.82 (0.72–0.89)	2.0	5.7	41%	0.88 (0.81–0.93)	1.2	3.3	40%	0.87 (0.79–0.92)	1.24	3.4	50%
1	3	0.91 (0.85–0.95)	1.0	2.8	29%	0.83 (0.73–0.90)	2.0	5.5	41%	0.89 (0.82–0.93)	1.14	3.2	38%	0.90 (0.84–0.94)	1.11	3.1	44%
2	1	0.91 (0.85–0.95)	1	2.8	29%	0.89 (0.82–0.93)	1.6	4.3	32%	0.91 (0.85–0.95)	1.06	2.9	36%	0.90 (0.84–0.94)	1.11	3.1	44%
2	2	0.94 (0.90–0.96)	0.8	2.2	23%	0.9 (0.84–0.94)	1.4	4.0	29%	0.93 (0.88–0.96)	0.91	2.5	31%	0.93 (0.88–0.96)	0.88	2.4	35%
2	3	0.95 (0.92–0.97)	0.7	2.0	21%	0.91 (0.85–0.95)	1.4	3.9	29%	0.94 (0.90–0.96)	0.85	2.4	29%	0.95 (0.92–0.97)	0.78	2.2	31%
3	1	0.94 (0.90–0.96)	0.8	2.3	24%	0.92 (0.87–0.95)	1.3	3.5	26%	0.92 (0.87–0.95)	0.94	2.6	32%	0.93 (0.88–0.96)	0.91	2.5	36%
3	2	0.96 (0.93–0.98)	0.7	1.9	19%	0.93 (0.88–0.96)	1.2	3.3	24%	0.95 (0.92–0.97)	0.79	2.2	27%	0.95 (0.92–0.97)	0.72	2	29%
3	3	0.97 (0.95–0.98)	0.6	1.7	17%	0.94 (0.90–0.96)	1.2	3.2	23%	0.95 (0.92–0.97)	0.73	2.0	25%	0.96 (0.93–0.98)	0.64	1.8	26%

* SDD in % = SDD / mean of the group Nm. Change that can be detected reliable (95% certainty to be true change).

Table 2.4b Reliability ordered by number of test occasions (Occ) and repetitions (Rep), of isometric muscle strength measurement for the most impaired limb (D-study)

Age group 6–10 years																	
		HA_{most}				KE_{most}				$GSTR_{most}$				SOL_{most}			
Occ	Rep	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %	ICC (95% CI)	SEM (Nm)	SDD (Nm)	SDD %
1	1	0.94 (0.90–0.96)	2.17	6.0	25%	0.94 (0.90–0.96)	2.23	6.2	19%	0.78 (0.66–0.86)	2.83	7.8	42%	0.79 (0.68–0.87)	2.43	6.7	57%
1	2	0.96 (0.93–0.98)	1.78	3.1	20%	0.96 (0.93–0.98)	1.94	5.4	16%	0.79 (0.68–0.87)	2.72	7.5	40%	0.82 (0.72–0.89)	2.26	6.3	53%
1	3	0.96 (0.93–0.98)	1.63	4.5	19%	0.96 (0.93–0.98)	1.83	5.1	15%	0.79 (0.68–0.87)	2.68	7.4	39%	0.82 (0.72–0.98)	2.2	6.1	52%
2	1	0.97 (0.95–0.98)	1.6	4.4	18%	0.97 (0.95–0.98)	1.63	4.5	14%	0.87 (0.79–0.92)	2.02	5.6	30%	0.88 (0.81–0.93)	1.75	4.9	41%
2	2	0.98 (0.97–0.99)	1.3	3.6	15%	0.98 (0.97–0.99)	1.4	3.9	12%	0.88 (0.81–0.93)	1.93	5.4	28%	0.9 (0.84–0.94)	1.61	4.5	38%
2	3	0.98 (0.97–0.99)	1.19	3.3	14%	0.98 (0.97–0.99)	1.31	3.6	11%	0.88 (0.81–0.93)	1.9	5.3	28%	0.9 (0.84–0.94)	1.56	4.3	36%
3	1	0.98 (0.97–0.99)	1.37	3.8	16%	0.98 (0.97–0.99)	1.38	3.8	12%	0.91 (0.85–0.95)	1.67	4.6	25%	0.91 (0.85–0.95)	1.45	4.0	34%
3	2	0.98 (0.97–0.99)	1.1	3.1	13%	0.98 (0.97–0.99)	1.17	3.2	10%	0.92 (0.87–0.95)	1.59	4.4	23%	0.93 (0.88–0.96)	1.33	3.7	31%
3	3	0.99 (0.98–0.99)	0.99	2.7	11%	0.99 (0.98–0.99)	1.09	3.0	9%	0.92 (0.87–0.95)	1.56	4.3	23%	0.93 (0.88–0.96)	1.29	3.6	30%

* SDD in % = SDD / mean of the group Nm. Change that can be detected reliable (95% certainty to be true change).

Table 2.5 Reliability ordered by number of test occasions (Occ) of standing heel-rise (SH) tests (D-study)

	3–5 years							
	SH most impaired limb[1]				SH less impaired limb			
Occ[2]	ICC (95% CI)	SEM (No.)	SDD[3] (No.)	SDD %	ICC (95% CI)	SEM (No.)	SDD (No.)	SDD %
1	0.89 (0.79–0.94)	0.92	2.55	128	0.86 (0.74–0.93)	2.38	6.6	81
2	0.94 (0.88–0.97)	0.65	1.8	90	0.93 (0.86–0.96)	1.68	4.66	57
3	0.96 (0.93–0.98)	0.53	1.47	74	0.95 (0.91–0.97)	1.38	3.83	47

	6–10 years							
	SH most impaired limb[1]				SH less impaired limb			
Occ[2]	ICC (95% CI)	SEM (No.)	SDD (No.)	SDD %	ICC (95% CI)	SEM (No.)	SDD (No.)	SDD %
1	0.98 (0.96–0.99)	0.78	2.16	48	0.87 (0.74–0.94)	2.36	6.54	46
2	0.99 (0.98–1.00)	0.55	1.52	34	0.93 (0.86–0.96)	1.67	4.67	32
3	0.99 (0.98–0.99)	0.45	1.25	28	0.95 (0.91–0.97)	1.36	3.77	26

[1] SH most impaired limb = standing heel-rise of the most impaired limb.
[2] Number of test occasions.
[3] SDD in % = SDD / mean of the group No. Change that can be detected reliable (95% certainty to be true change).

Discussion

In this study the feasibility and reliability of leg strength measurements in a group of young children with CP were assessed. The results of this study showed that the measurement error reduced more by taking the average over more occasions on separate days than by taking the average over more repetitions in 1 test occasion. Our data showed that it is possible to monitor muscle strength in the lower extremities in children in the age of 3–5 years, when taking the average over 2 or 3 test occasions and 2 repetitions. This is feasible in clinical practice when a physical therapist is treating a child on a regular basis. For the older age group small changes (<20%) in strength of the HA and the KE muscles can be detected with only 1 test occasions and 2 repetitions. For each age, muscle group

and expected changes the optimal test design can be derived from the SDDs reported in Table 2.4a and Table 2.4b. That makes it possible for a clinician to choose the best test design fitting their purpose.

The feasibility of the isometric muscle tests was good, although most of the younger age group could not concentrate long enough to measure all the muscle groups in 1 occasion or they needed more time. Even the 3-years-old children were able to understand that they had to push as hard as they could and gave their best performance. It is however recommended to perform a limited number of tests at a time in the younger age group.

Reliability parameters, like the ICC, are highly dependent on the heterogeneity of the study sample, while the agreement parameters, based on SEM and SDD, are more specific for the measurement (27). ICC values of all HHD measurements in this study were indicative for good reliability, although we divided the children in a younger and older group and reduced heterogeneity in that respect. More important in this study was the SDD, derived from the SEM, enabling interpretation of a decline or an increase in strength. Variance components were higher regarding the factor "occasion" than in the factor "repetition", as shown in the D-study (Table 2.4a+b). Therefore there is more advantage in reducing the measurement error in the isometric strength tests by taking the average of more test occasions instead of more repetitions (Table 2.4a+b).

The SEM and SDD of the isometric strength tests in the hip abductor and knee extensor were not as high as in the ankle plantar flexor strength measurements. This is in agreement with other studies in children with CP (12;13). Previous studies (28;29) have shown that in children with CP strength training can lead to 30 to 39% changes in knee extensor strength (28;29). Therefore, the reported measurement error in the current study appears to be of an acceptable level for evaluating the isometric strength in individuals in the older age group, even with only 1 test occasion (SDD≥19%). In the 6–10 yrs group a 16% change can be detected in the KE with 1 occasion and 2 repetitions (Table 2.4b). To detect smaller changes in KE the number of test occasions can be increased. In younger children it is necessary to take the average of 2 test occasions (SDD≥29%) to detect a 30% change in knee extensor strength (Table 2.4a). This example shows that the younger age group needs another test design for monitoring change in for example knee extensor strength than the older children.

Despite standardization and stabilisation procedures, ankle plantar flexor muscle strength demonstrated large variability. The largest measurement error was found for the soleus muscles. Strength of the plantar flexor muscles is difficult to measure since the high

forces and short lever arm (distance from lateral malleolus to location HHD) makes it difficult to stabilize the dynamometer manually. We expected that it would be easier to measure strength of the plantar flexors in the younger children than in the older age group because of lower strength levels. The measurement errors are, however, equally large in the two age groups. It is only possible to measure changes of 23 to 30% or larger in strength in the calf muscles in both age groups when choosing a test design with 3 test occasions and 3 repetitions depending on age, muscle group (GSTR or SOL) and change to be expected. Organising 3 test occasions, however, is not always feasible for clinicians and children. An increase of 51% in ankle plantar flexor strength (SOL) was reported in a strength training intervention in children with CP from 8 to 18 years old (30), indicating that this test could also be used in interventions when large improvements are expected. Choosing the most optimal design is a trade off between the expected changes in strength and the feasibility of performing repeated measures on separate days to reduce the SDD.

Plantar flexor muscles in the most impaired limb in independently ambulant children with CP, produce as little as 48% of the force of those in matched typically developing children measured with HHD (23). The distal lower-limb musculature is weaker than the proximal muscles (23). Whereas in normal human walking, the plantar flexors provide much of the force to support the body and accelerate the lower limbs in push-off, particularly in mid- and late stance (8). Knowing the important role of the plantar flexor strength in walking it is important for clinicians to monitor muscle strength of the calf muscles and know the SDDs of the HHD measurements of these muscle groups.

The SDDs found in this study were smaller in the proximal muscle groups than in the calf muscles. We expect, however, larger changes in muscle strength after strength training in the distal muscle groups than in the proximal muscle groups, knowing that the weakest muscles develop more in strength than the stronger muscles after the same amount of training.

The results of this study showed that two or more test occasions are necessary in isometric strength testing with HHD in young children with CP, and in the calf muscles in older children. This can be problematic due to time constraints and it is, therefore, necessary to analyse thoroughly which muscle groups you want to monitor. It is not feasible and too time consuming to monitor several muscle groups with HHD. We implemented the HHD measurements in the clinical setting by testing on 2 or 3 test occasions before and after an intensive training program, which is feasible in our setting.

The standing heel-rise test has been proposed as an alternative test for measuring plantar flexor strength instead of dynamometry (18). The standing heel-rise test was not easy to perform for all children and they had many ways to compensate for their poor muscle strength, balance and/or coordination by flexing their knees in preparation or leaning on the examiner. Standardisation of the test was therefore difficult which could be an important explanation of the poor reliability of the test.

The good ICCs (ICC 0.86–0.98) and large SEM (0.78 to 2.38 heel-rises) for the standing heel-rise test found in this study are consistent with those of Yocum et al. (2003) in children with plantar flexion weakness and typically developing children (ICC: 0.85–0.88, SEM: 2.6 and 3.2 heel-rises) (31). In our study we found the highest SDD (2.55, 128%) in the heel-rise test with the most impaired limb in the younger age group. In the D-study we found that when increasing the number of test occasions to three, it would be possible to measure individual changes of 74% in the most impaired limb and 47% in the less impaired limb in the younger age group (Table 2.5). It does not seem feasible and useful to use the single limb heel-rise test in clinical practice when SDD is so large. In a plantar flexor strength training study, however, with children with CP in the age of 6 to 16 years, gains in heel rises of 122% and 450% after 5–12 weeks of training were found (19). When expecting such large increase in strength after training the standing heel-rise test is sensitive enough to detect change even in the younger age group.

The SH test on both limbs was stopped when the children reached 20 heel rises. Especially in the older age group almost all children reached 20 repetitions, which yielded a ceiling effect, therefore, we were not able to perform a reliability analysis on these data. In further reliability research with the SH test on both limbs, we recommend that there is no maximum set in the test to avoid a ceiling effect.

The results of this study are limited to ambulatory children with spastic CP (GMFCS I-II) in the age of 3–10 years old. Results cannot be generalized to other clinical sub-types of CP or muscle groups.

Conclusion

Results of this study can be used to determine a test design for HHD measurement in young children with CP, depending on expected changes in muscle strength, muscle group and age of the participants. Choosing the most optimal design is a balance between the smallest detectable changes and the feasibility of performing repeated measures on separate days. The single limb SH test can only be used to determine large changes in

dynamic plantar flexor strength when the average is taken over 3 test occasions in young children with CP. The SH test on both limbs needs further development to avoid ceiling effects.

Acknowledgements

This study was supported by the Duyvensz-Nagel stichting, Amsterdam, The Netherlands and Amsterdam Rehabilitation Research Centre | Reade, Amsterdam, The Netherlands. The authors are very grateful to all children and their parents for their participation. We also thank paediatric physiotherapists Cindy Warntjes for her help with the assessments and Susan Aylott for correcting the English text of this article.

References

(1) Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl 2007 Feb;109:8-14.

(2) Prevalence and characteristics of children with cerebral palsy in Europe. Dev Med Child Neurol 2002 Sep;44(9):633-40.

(3) Andersen GL, Irgens LM, Haagaas I, Skranes JS, Meberg AE, Vik T. Cerebral palsy in Norway: prevalence, subtypes and severity. Eur J Paediatr Neurol 2008 Jan;12(1):4-13.

(4) Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. Pediatrics 2008 Jan;121(1):e187-e192.

(5) Gormley ME, Jr. Treatment of neuromuscular and musculoskeletal problems in cerebral palsy. Pediatr Rehabil 2001 Jan;4(1):5-16.

(6) Gorter JW, Rosenbaum PL, Hanna SE, Palisano RJ, Bartlett DJ, Russell DJ, et al. Limb distribution, motor impairment, and functional classification of cerebral palsy. Dev Med Child Neurol 2004 Jul;46(7):461-7.

(7) Gage J, Schwartz M, Koop S, Novacheck T. The identification and treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009.

(8) Neptune RR, Kautz SA, Zajac FE. Contributions of the individual ankle plantar flexors to support, forward progression and swing initiation during walking. J Biomech 2001 Nov;34(11):1387-98.

(9) Ledebt A, Bril B, Breniere Y. The build-up of anticipatory behaviour. An analysis of the development of gait initiation in children. Exp Brain Res 1998 May;120(1):9-17.

(10) Rosenbaum PL, Walter SD, Hanna SE, Palisano RJ, Russell DJ, Raina P, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. JAMA 2002 Sep 18;288(11):1357-63.

(11) Scholtes VA, Dallmeijer AJ, Becher JG. Can we identify predictors of multilevel botulinum toxin A injections in children with cerebral palsy who walk with a flexed knee pattern? J Child Neurol 2008 Jun;23(6):628-34.

(12) Berry ET, Giuliani CA, Damiano DL. Intrasession and intersession reliability of handheld dynamometry in children with cerebral palsy. Pediatr Phys Ther 2004;16(4):191-8.

(13) Crompton J, Galea MP, Phillips B. Hand-held dynamometry for muscle strength measurement in children with cerebral palsy. Dev Med Child Neurol 2007 Feb;49(2):106-11.

(14) Taylor NF, Dodd KJ, Graham HK. Test-retest reliability of hand-held dynamometric strength testing in young people with cerebral palsy. Arch Phys Med Rehabil 2004 Jan;85(1):77-80.

(15) Streiner LD, Norman GR. Health Measurement Scales, a practical guide to their development and use. fourth edition ed. New York: Oxford University Press Inc.; 2008.

(16) de Vet HC, Terwee CB, Mokkink LB, Knol DL. Measurement in Medicine. Cambridge; 2011.

(17) Damiano DL, Dodd K, Taylor NF. Should we be testing and training muscle strength in cerebral palsy? Dev Med Child Neurol 2002 Jan;44(1):68-72.

(18) Lunsford BR, Perry J. The standing heel-rise test for ankle plantar flexion: criterion for normal. Phys Ther 1995 Aug;75(8):694-8.

(19) McNee AE, Gough M, Morrissey MC, Shortland AP. Increases in muscle volume after plantarflexor strength training in children with spastic cerebral palsy. Dev Med Child Neurol 2009 Jun;51(6):429-35.

(20) Haber M, Golan E, Azoulay L, Kahn SR, Shrier I. Reliability of a device measuring triceps surae muscle fatigability. Br J Sports Med 2004 Apr;38(2):163-7.

(21) Moller M, Lind K, Styf J, Karlsson J. The reliability of isokinetic testing of the ankle joint and a heel-raise test for endurance. Knee Surg Sports Traumatol Arthrosc 2005 Jan;13(1):60-71.

(22) Verschuren O, Ketelaar M, Takken T, Van BM, Helders PJ, Gorter JW. Reliability of hand-held dynamometry and functional strength tests for the lower extremity in children with Cerebral Palsy. Disabil Rehabil 2008;30(18):1358-66.

(23) Wiley ME, Damiano DL. Lower-extremity strength profiles in spastic cerebral palsy. Dev Med Child Neurol 1998 Feb;40(2):100-7.

(24) Le Veau B. Biomechanics of Human Motion. 3rd ed. Philadelphia: WB Saunders; 1992.

(25) Smits DW, van Groenestijn AC, Ketelaar M, Scholtes VA, Becher JG, Gorter JW. Selective motor control of the lower extremities in children with cerebral palsy: inter-rater reliability of two tests. Dev Neurorehabil 2010;13(4):258-65.

(26) Portney LG, Watkins MP. Foundations of Clinical Research Application to Practice. Third Edition ed. New Jersey: Pearson education; 2009.

(27) de Vet HC, Terwee CB, Knol DL, Bouter LM. When to use agreement versus reliability measures. J Clin Epidemiol 2006 Oct;59(10):1033-9.

(28) Damiano DL, Abel MF. Functional outcomes of strength training in spastic cerebral palsy. Arch Phys Med Rehabil 1998 Feb;79(2):119-25.

(29) Morton JF, Brownlee M, McFadyen AK. The effects of progressive resistance training for children with cerebral palsy. Clin Rehabil 2005 May;19(3):283-9.

(30) Dodd KJ, Taylor NF, Graham HK. A randomized clinical trial of strength training in young people with cerebral palsy. Dev Med Child Neurol 2003 Oct;45(10):652-7.

(31) Yocum A, McCoy SW, Bjornson KF, Mullens P, Burton GN. Reliability and validity of the standing heel-rise test. Phys Occup Ther Pediatr 2010 Aug;30(3):190-204.

(32) Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol 1997 Apr;39(4):214-23.

Effectiveness of functional power training on walking ability in young children with cerebral palsy: study protocol of a double-baseline trial

Liesbeth F. van Vulpen
Sonja de Groot
Eugene A.A. Rameckers
Jules G. Becher
Annet J. Dallmeijer

Pediatr Phys Ther 2017 Jul;29(3):275-282

Abstract

Purpose To evaluate the effect of functional high-velocity resistance (power) training, to improve walking ability of young children with cerebral palsy (CP).

Methods Twenty-two children with bi- or unilateral spastic CP, Gross Motor Functiom Classification System I and II, aged 4–10 years will be recruited. A double-baseline design will be used to compare a 14-weeks functional power training (3 times a week) with a 14-weeks usual care period and a 14 weeks follow-up period. The power exercises will be loaded and performed at 50–70% of the maximum unloaded speed. Load will be increased when exercises are performed faster than 70% of the unloaded speed. Primary outcomes will be sprinting capacity (15-m Muscle Power Sprint Test) and goal attainment scaling score of walking-related treatment goals. Secondary outcomes will be walking speed (1-min walk test), endurance (10-m shuttle-run test), gross motor function, lower-limb strength and parent-reported mobility.

Results and conclusions Not yet available.

Trial registration NTR5189.

Introduction and purpose

Cerebral palsy (CP) comprises of a group of disorders in the development of movement and posture, attributed to non-progressive disturbances that have occurred in the developing fetal or infant brain (1). Prevalence rates of CP are about 2 per 1000 births in Europe of which 82% has a spastic CP (2). Motor impairment in CP is multi-factorial and includes problems such as spasticity, coordination problems, loss of selective motor control and muscle weakness (2). In the group of children with a spastic CP, 60–70% has achieved walking ability with or without assistive mobility devices between 6–12 years, which is level I–III according to the Gross Motor Function Classification System (GMFCS) (1;3). Ambulant children with CP are often integrated in community schools and recreational facilities, and thus are required to perform the same activities alongside their peers with typically development (TD), like playing in the schoolyard and walking from school class to the gym or at school outings (4). As a result of their motor impairments children with CP have problems in daily life with keeping up with peers in their walking ability (4).

Muscle strength is an important impairment that is closely related to walking ability in children with CP (5). Plantar flexor strength seems to be particularly important for walking because of its contribution to generate power at push-off, which is necessary for step length and walking speed. Muscle power production of plantar flexors is more reduced than more proximal muscle groups in children with CP in comparison with TD children (6).

Several studies have examined methods to improve the walking ability of children with CP by muscle strengthening (7). Progressive Resistance Exercise training (PRE) has therefore been applied to improve gross motor function and walking ability through increases in muscle strength. However, despite increases in strength in most lower-limb muscles there were only limited or no improvements of walking ability after PRE training (7). The improvement in muscle strength after PRE is apparently not transferred to the walking activity. Moreau et al. (8) suggested that training at higher, more functional movement velocities than generally used in PRE training might lead to functional improvements. This suggestion was based on their findings that children with CP have a reduced ability to rapidly generate forces (8). This is likely to affect daily life and playing activities, such as games in playground or sports such as football, as in almost all these activities the child is more involved in high-intensity physical activities of short duration. These high-intensity physical activities may be limited in children with CP due to the aforementioned reduced ability to rapidly generate forces (8). It has indeed been shown that children with CP have lower sprint capacity than their TD peers (9). Training of high-velocity movement may be especially important during the growing years when neural plasticity and motor coordination are most sensitive to change (10).

Another possible reason for limited effects of PRE training on walking ability is that most previous studies did not train the plantar flexor muscles while, as said before, plantar flexor strength is strongly related to the generated power at push-off in walking (11). Plantar flexor muscles in ambulant children with CP produce as little as 48% of the force of those in matched TD children (6). This is already seen in young children with CP from the age of 6 years old (6). Yet several common treatments are likely to weaken the plantar flexors, like for example botulin toxin injections and casting periods to increase muscle length. However, training of the plantar flexor muscles seems particularly important for improving walking ability in this population.

Therefore, we developed a functional power training program, called MegaPower training, consisting of loaded functional exercises, such as walking, running and climbing stairs, performed at high velocities. Training duration, frequency and intensity are based on strength training guidelines for youth from the National Strength and Conditioning Association (NSCA) (10;12). In this functional power training program the children will train on high movement velocities to promote neuromuscular adaptations and maximize the training effects on walking ability (10;13). To our knowledge, combining high-movement velocity with external loads and a controlled progression in load during the training period, embedded in functional exercises has never been investigated in children with CP. The children will be guided carefully during the exercises by the trainer and a story about super heroes will keep the children motivated to give their best effort during the training.

The aim of this study is to evaluate the effect of resistance training at high movement velocities in functional exercises (functional power training) on walking ability in young children with CP.

This paper describes the design and training protocol of a double-baseline trial to assess the effectiveness of functional power training on walking ability (sprinting ability, walking speed and endurance) and plantar flexor strength in young children with cerebral palsy compared to a period of usual care.

Methods

Participants

We will include 22 ambulant children (GMFCS I and II) with a predominantly spastic CP aged 4–10 years. Parents and/or the children have a treatment question related to walking ability (such as being able to walk longer or faster). The children have to be able to

understand and follow instructions. Exclusion criteria will be: 1) treatment with botulinum toxin A in lower limb and/or serial casting of lower limb less than 6 months before the start of the functional power training, 2) selective dorsal rhizotomy treatment less than a year before the functional power training, 3) walking is not (yet) the preferred way of mobility.

Design and procedure

This research protocol has a 'double-baseline' design (see Figure 3.1). The children who will participate act as their own controls by comparing the changes in outcome measures in a 14-weeks usual care period with the changes in a 14-weeks training intervention (functional power training) that follows immediately after the usual care period. Measurements will be done before the usual care period (pretest 1), after the 14wk usual care period, which is also the start of the training period (pretest 2), after the 14wk training period (posttest), and a follow-up test will be scheduled 14-weeks after the posttest to assess if the potential improvement will remain (follow-up test).

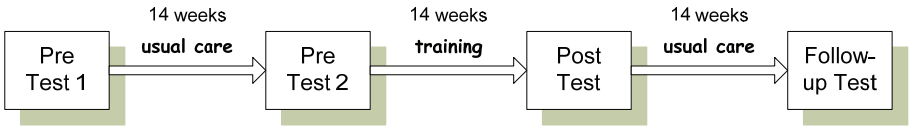

Figure 3.1 Double-baseline research protocol with follow-up measurement.

Primary outcomes in this study are sprinting capacity measured with the 15 meter Muscle Power Sprint Test and evaluation of treatment goals with a focus on walking reported by parents and children measured with goal attainment scaling (see Figure 3.2). Secondary outcomes are walking capacity measured with 1-minute walk test (walking speed), 10 meter shuttle run test (endurance), gross motor function and lower-limb strength. Parent-reported mobility performance will be measured with the Mobility Questionnaire (MobQues) and the Functional Mobility Scale (FMS). For each child body mass and body height will be measured. GMFCS level and type of CP will be determined by a pediatric physiotherapist together with the physician.

The Medical Ethics Committee of the Slotervaart medical center and Reade rehabilitation research center in Amsterdam, the Netherlands approved this study and written informed consent forms will be obtained from the parents of each participant.

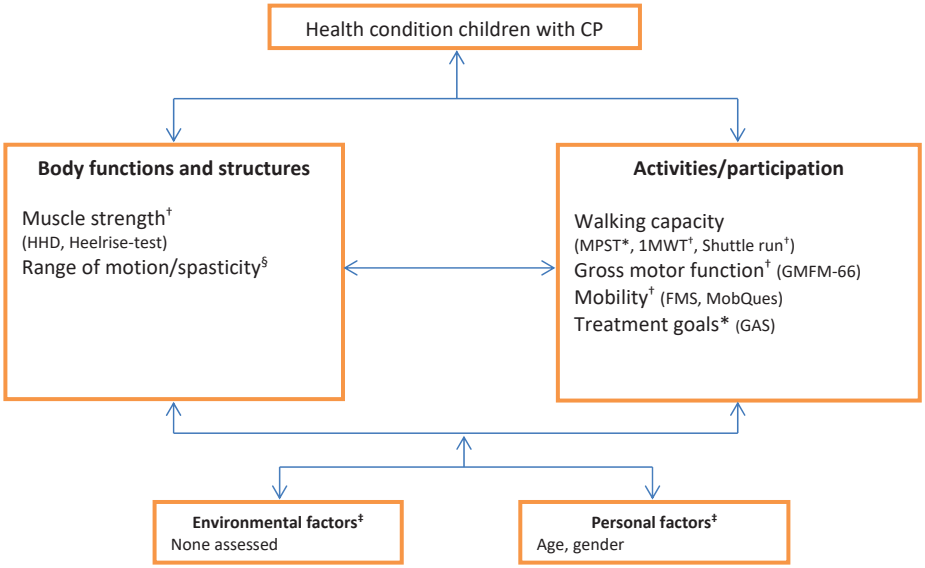

Figure 3.2 Study outcomes used on the different ICF levels: * primary outcomes; † secondary outcomes; § adverse outcomes; ‡ control outcomes.
Abbreviations: CP, cerebral palsy; HHD, hand-held dynamometer; GMFM, Gross Motor Function Measure; MPST, Muscle Power Sprint Test; 1MWT, 1-minute walk test; FMS, Functional Mobility Scale; MobQues, Mobility Questionnaire; GAS, Goal Attainment Scaling.

Setting

Participants will be recruited from a rehabilitation centre, two special needs schools for children with physical disabilities, and an outpatient clinic of a university medical center. The training and assessments will take place in two special schools for physically disabled children and in a rehabilitation centre in the Netherlands.

Intervention

In the intervention period children will follow the functional power training for a period of 14 weeks, 3 times a week. Each training session lasts 60 minutes. The training consists of the following phases: 1. warm-up (10 min), 2. 3–4 different power exercises (35 min), 3. end game (15 min). The training sessions will be in small groups (3–6 children) and will be supervised by the same number of therapists. During each training session children will wear regular sport shoes without their splints if they have any, and sport outfit. Each training session starts with a warm up with walking and running exercises and dynamic calf muscle stretching exercises. Power exercises and the end game will be

chosen in line with the treatment goals of the parents and children. A story about super heroes and secret missions is made to keep the children motivated and to give their best effort during the training sessions. They all will receive their own T-shirt with their super hero to stimulate the group feeling and training motivation.

Power exercises

For all participants 4 to 6 different power exercises will be selected that are relevant for the treatment goals set by the parents and children for improving daily life activities. In each training session the children will perform 3 or 4 of the power exercises (see Table 3.1). The power exercises are specifically designed to strengthen the plantar flexor muscles while performing functional exercises. Characteristics of the power exercises are described in Table 3.1. Key elements of the power exercises are: a) functional loaded multi-joint exercises like running and walking with a focus on the ankle push-off, b) high movement velocity (similar to the velocity used in daily/playing activities), c) progressive load. The following exercises will be used: 1) running, 2) walking, 3) pushing chair, 4) stair climbing, 5) propelling scooter, and 6) sideways walking. Training volume is determined by load, movement velocity and number of repetitions (Table 3.1). The exercise load will be adjusted to a level that allows the children to perform at 70% of their maximum unloaded speed. Each exercise will be performed at maximal effort for 25s, with a resting period of 30–50s, and with 6 to 8 repetitions each exercise. For each power exercise, velocity and distance will be calculated at baseline (Table 3.2). When children become faster (i.e. performs the exercise in less than 25s at maximal effort) load will be increased (by steps of approximately 10% of the current load) to maintain the target velocity. To motivate the children and to control the movements during the exercises, every child will be supervised individually during the power exercises (one on one).

Primary outcome measures

Sprint performance

Sprint performance will be measured with the Muscle Power Sprint Test (MPST). The MPST is an intermittent sprint test, in which the child stops and starts at standardized intervals. The MPST measures the sprint capacity of the child expressed in mean power and peak power (14). Children sprint at maximum speed over 15 meters with 6 repetitions. Between the 6 sprints is a 10s break in which the child can turn and be ready for the next sprint. The time used for each sprint is measured with a stopwatch. Power output for each sprint will be estimated from the collected data using the following equations:

Table 3.1 Characteristics and training volume of the six different functional power exercises

Exercise	Load	Method of loading	Sets	Duration	Rest	Intensity
Running	Load level that allows child to perform at 50–70% of maximal unloaded running speed	Dragging a loaded box over-ground with belt around the hips (Figure 3.3A)	6 to 8 repetitions	25s	30–50s	Maximal effort
Walking	Load level that allows child to perform at 50–70% of maximal unloaded walking speed	Dragging a loaded box over-ground with belt around the hips (Figure 3.3A)	6 to 8 repetitions	25s	30–50s	Maximal effort
Pushing chair	Load level that allows child to perform at 50–70% of maximal unloaded running speed	Chair, with a loaded box underneath (Figure 3.3B)	6 to 8 repetitions	25s	30–50s	Maximal effort
Stair climbing	Load level that allows child to perform at 50–70% of maximal speed for unloaded stair climbing	Loaded vest (Figure 3.3C)	6 to 8 repetitions	25s	30–50s	Maximal effort
Propelling a three-wheel scooter	Load level that allows child to perform at 50–70% of maximal speed for unloaded scooter propelling, for each leg separately	Loaded box attached to scooter (Figure 3.3D)	6 to 8 repetitions	25s	30–50s	Maximal effort
Sideways walking	Load level that allows child to perform at 50–70% of maximal unloaded sideways walking speed	Dragging a loaded box over-ground with belt around the hips	6 to 8 repetitions	25s	30–50s	Maximal effort

1) *velocity (m/sec) = 15 meter/time*, 2) *acceleration (m/sec²) = velocity/time*, 3) *force (kg·m/sec²) = body mass * acceleration*, 4) *power (W) = force * velocity* (14). For each of the six 15-meter runs the power will be calculated. Peak power will be defined as the highest power output of those 6 runs. Mean power will be defined as average power output of the 6 runs. The reliability of the MPST, with an Intraclass Correlation Coefficient (ICC) of 0.97, as well as the feasibility and construct validity are good (14;15).

Treatment goals reported by parents and/or children

Treatment goals will be measured by the Goal Attainment Scaling (GAS), an individualized measurement to evaluate parents' and children's progress towards activity and participation

Figure 3.3 Examples of MegaPower exercises: A) running while dragging a loaded box, B) pushing a loaded chair, C) running up the stairs with loaded vest, D) propelling a three-wheel scooter while dragging a loaded box.

Table 3.2 Calculation of training velocity, distance and load for each functional power exercise

Exercise	Velocity	Target distance	Starting load	Progression
Running	Maximal unloaded running speed is calculated as the average speed over the six trials of the MPST[1].	Target distance (m) is calculated as maximal running speed (m/s) * 25s * 0.7 (i.e. 70% of maximal unloaded speed)	Starting load is determined by asking maximal effort of the child when running the target distance while dragging a loaded box. If the child completes the target distance in less than 25s, load is added to the box. If the child cannot complete the target distance within 25s, load is removed from the box. Indication for starting load is 25% of the child's body mass.	Load is increased with approximately 10% of the current load (0.5 kg is the lowest amount of load to increase) when the child becomes faster during the training period, i.e. reaches the target distance of the exercise in less than 25s (i.e. >70% max speed).
Pushing chair	Maximal unloaded running speed is calculated as the average speed over the six trials of the MPST[1].	Target distance (m) is calculated as maximal running speed (m/s) * 25s * 0.7 (i.e. 70% of maximal unloaded speed)	Starting load is determined by asking maximal effort of the child when running the target distance while pushing a loaded chair. If the child completes the target distance in less than 25s, load is added to the chair. If the child cannot complete the target distance within 25s, load is removed from the chair. Indication for starting load is 15% of the child's body mass.	Load is increased with approximately 10% of the current load (0.5 kg is the lowest amount of load to increase) when the child becomes faster during the training period, i.e. reaches the target distance of the exercise in less than 25s (i.e. >70% max speed).
Walking	Maximal unloaded walking speed is calculated as the average walking speed of the 1-minute walk test.	Target distance (m) is calculated as maximal walking speed (m/s) * 25s * 0.7	Starting load is determined by asking maximal effort of the child when walking the target distance while dragging a loaded box. If the child completes the target distance in less than 25s, load is added to the box. If the child cannot complete the distance within 25s, load is removed from the box. Indication for starting load is around 45% of the child's body mass.	Load is increased with approximately 10% when the child becomes faster during the training period, i.e. reaches the target distance in less than 25s (i.e. >70% max speed).

Exercise	Velocity	Target distance	Starting load	Progression
Stair climbing	Maximal number of stair steps is determined as the number of steps when climbing the stairs as fast as possible in 25s with unloaded vest.	Maximal number of steps in 25s * 0.7	Starting load is determined by asking maximal effort of the child when climbing stairs with a loaded vest. If the child completes the target number of steps in less than 25s, load is added to the vest. If the child cannot complete the target number of steps within 25s, load is removed from the vest. Indication for starting load is around 15% of the child's body mass.	Load is increased with approximately 10% when the child becomes faster during the training period, i.e. reaches the target distance in less than 25s (i.e. >70% max speed).
Propelling a three-wheel scooter	Maximal speed is determined (for each leg separately) by asking the child to propel the scooter with an unloaded box as fast as possible for 25s.	Target distance (m) is calculated as distance propelling the scooter unloaded in 25s * 0.7	Starting load is determined by asking maximal effort of the child when propelling the scooter over the target distance while dragging a loaded box attached to the scooter. If the child completes the target distance in less than 25s, load is added to the box. If the child cannot complete the distance within 25s, load is removed from the box. Indication for starting load is around 15% of the child's body mass.	Load is increased with approximately 10% when the child becomes faster during the training period, i.e. reaches the target distance in less than 25s (i.e. >70% max speed).
Sideways walking	Maximal unloaded sideways walking speed is determined by asking the child to walk sideways as fast as possible while dragging an unloaded box over 25s.	Target distance (m) is calculated as distance sideways walking unloaded in 25s * 0.7	Starting load is determined by asking maximal effort of the child when sideways walking the calculated target distance while dragging a loaded box. If the child completes the target distance in less than 25s, load is added to the box. If the child cannot complete the distance within 25s, load is removed from the box. Indication for starting load is around 35% of the child's body mass.	Load is increased with approximately 10% when the child becomes faster during the training period, i.e. reaches the target distance in less than 25s (i.e. >70% max speed).

[1] MPST, Muscle Power Sprint Test six times 15 meter sprint.

goals (16). GAS is a sensitive, evaluative measurement which describes the change of individuals or groups after treatment (ICC=0.86) (17). It is a 6-point scale measurement, with the score -2 representing the level equal to start, score -1 less progress than expected, score 0 for the expected level of functioning, score +1 and score +2 for achievement of more and much more than was expected, respectively, and -3 for deterioration. We will adhere to the following criteria for scale development: a) goals will be set by experienced pediatric physical therapists in consultation with parents and children, based on their main aim of therapy in terms of activity and participation domains of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY), b) the six levels of the GAS scales will be specific, measurable, achievable, realistic/relevant and time-related (SMART), c) scales will be constructed ordinal with incremental steps of equal intervals (17).

Secondary outcome measures

Walking ability

The secondary outcome measures on the activity level of the ICF-CY on walking capacity will be the 1-Minute Walk Test (1MWT) and the 10-meter Shuttle Run Test (SRT) as shown in Figure 3.2.

The 1MWT measures walking speed as the distance walked in 1 minute. The children will be asked to walk as fast as possible without running around an oval track. The reliability (ICC=0.94) and validity of the 1MWT are good (18;19).

The SRT is developed for children with CP GMFCS level I and II and measures endurance (20). Children walk or run between 2 markers delineating the respective course of 10-m, at a set incremental speed determined by a signal. Experienced pediatric physical therapists will accompany the children to help the children pace themselves with the audio signal. The test is finished when, on 2 consecutive paced signals, the children do not reach the marker. Children with GMFCS level I will perform the SRT-I with starting speed 5 km/h. Children with GMFCS level II will perform the SRT-II with starting speed 2 km/h. Both SRT tests increase 0.25km/h in speed every minute. Reliability and validity of the SRT-I and SRT-II are good (SRT-I, ICC=0.87–0.97 and SRT-II, ICC=0.94–0.99) (20).

Gross motor function

Gross motor function will be measured with the 66-item version of the Gross Motor Function Measure (GMFM-66). The GMFM-66 is a standardized tool designed to evaluate change in gross motor function in children with CP (21;22). Items that will be tested are

for instance activities in walking, running and jumping skills. There is a 4-point scoring system for each item. Good reliability, validity and responsiveness of the GMFM-66 has been demonstrated (21;22).

Mobility performance reported by parents

Mobility performance will be measured with the Functional Mobility Scale (FMS) and the Mobility Questionnaire (MobQues). The FMS is a questionnaire to classify functional mobility in children with CP in the age of 4 till 18 years (23). It scores functional mobility over three distinct distances, chosen to represent mobility in the home (5 meter), at school (50 meter) and in the wider community (500 meter). Parents will be asked to rate the child's usual walking performance of the three distances according to the need for assistive devices, such as walking frame, crutches or wheelchair. Test - retest reliability of the FMS is good (Kappa=0.86–0.92) (23) as well as the construct, content and concurrent validity (23).

The Mobility Questionnaire (MobQues) is a Dutch questionnaire for parents with children in the age of 2–13 years to determine the extent of difficulty the child has with his mobility. The questionnaire addresses 28 mobility activities in everyday life and includes indoor activities, such as 'how difficult is it for your child to go up stairs?' as well as outdoor activities, such as 'how difficult is it for your child to walk on sand?'. The response options, given on a 5-point scale, are: not difficult at all, slightly difficult, somewhat difficult, very difficult, impossible without help. The test has a good intrarater reliability (ICC=0.96) (24) and content and construct validity (25).

Muscle strength

Secondary outcome measures in terms of body function and structures will be isometric muscle strength of the plantar flexors, quadriceps, hip abductors and dynamic muscle strength of the plantar flexors. The make-method will be used, where the child gradually builds up force against a hand-held dynamometer (microFET Hand-held Dynamometer, Biometrics BV, Almere, the Netherlands) for about 5s. The children will be allowed one or two practice trials for each test until the investigator is confident that the child understands the task. Each child will perform subsequently three repetitions for each muscle group and the maximum force (peak force) for each repetition will be registered. When the concentration and/or motivation of the child is not optimal a fourth or fifth repetition will be performed. Strong verbal encouragement during the measurement will be given to produce maximal effort. A standardized protocol will be used for positioning of the child, joint fixation, joint positioning and dynamometer resistance (26). Lever arm will

be measured between standardized landmarks with a hard tape measure, according to test procedures of Van Vulpen et al. (26). Torque (Nm) will be calculated by multiplying force (Newton) by the length (meter) of the lever arm. To improve reliability, isometric muscle strength will be measured at two different test occasions (different days, with a maximum of 7 days within measurements) on each measurement moment (Figure 3.1). The mean of the six measurements (two test occasions with three repetitions each) will be used in the analysis. Isometric strength measurements have good reliability in children with CP when measured with three repetitions in two different test occasions (ICC=0.88–0.98)(26).

Dynamic muscle strength of the plantar flexors will be measured with the standing heel-rise test on one limb. Both limbs will be tested. The number of repetitions for standing heel-rise on one limb, will be measured based on a standardized protocol. Children are allowed to touch the examiner only with a single finger for balance. The test is terminated when the child leans or pushes down on the examiner, the child's knees flexes, or when the child gives up or asks to stop despite encouragement. Moderate to good reliability is found for the heel-rise test in young children with CP (ICC=0.86–0.98)(26).

Adverse outcomes

Range of motion

Range of motion of the ankle (dorsiflexion) will be measured as joint angle with a goniometer with knees flexed and with the knees extended while the child is lying in supine position. (11).

Spasticity

Spasticity in the hamstrings, soleus and gastrocnemius muscles will be assessed by the joint angle at which a 'catch' (defined as a sudden increase in muscle tone, blocking further movement) will occur in a fast passive stretch (<1 second) while the child is lying in supine position (27).

Statistical analysis

Sample size

A power calculation was performed for the primary outcome measure, i.e. the mean power (W) of the Muscle Power Sprint Test. A pilot study (n=10) showed an increase of 85% (mean±SD: 13.1±12.2 W) in mean power after functional power training. Calculations were based on a within-subject design with a dependent t-test, a power of 0.8, alpha of

0.05 and an effect size 0.7. According to the power calculation, a sample size of at least 19 children is needed. Twenty-two children will be recruited to allow a dropout of 10%.

Changes in usual care period (Δt0-t1), training period (Δt1-t2) and follow-up period (Δt2-t3) will be calculated. Paired sample t-tests (if normally distributed) and Wilcoxon signed rank tests (if not normally distributed) will be used to determine whether the changes within these periods differ significantly between periods. All statistical analyses will be performed using SPSS Statistics 22.0 software (IBM Corporation, New York, USA).

Discussion

The 'double-baseline' design will be used instead of a randomized-control trial (RCT) to increase the feasibility of the study in a heterogeneous group of children with CP. Using this design, participants serve as their own controls by comparing the changes during a usual care period to the changes during an intervention period in which the participants follow the functional power training. The advantage of this design is that statistical power can be reached with smaller subject groups. Previous randomized-controlled intervention studies in children with CP suffered from low statistical power due to small sample sizes and heterogeneous groups, emphasizing the need for alternative statistical approaches (28). Apart from this higher feasibility and increased statistical power, another advantage of this design is that all children will receive the intervention.

Acknowledgements

This study was supported by grants from Stichting Mitialto, Wetenschappelijk College Fysiotherapie, Duyvensz-Nagel Stichting, Dr. Izak Wessel Stichting and Revalidatiefonds.

References

(1) Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl 2007 Feb;109:8-14.

(2) Graham HK, Rosenbaum P, Paneth N, Dan B, Lin JP, Damiano DL, et al. Cerebral palsy. Nat Rev Dis Primers 2016;2:15082.

(3) Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. Pediatrics 2008 Jan;121(1):e187-e192.

(4) Bax MC, Flodmark O, Tydeman C. Definition and classification of cerebral palsy. From syndrome toward disease. Dev Med Child Neurol Suppl 2007 Feb;109:39-41.

(5) Ferland C, Lepage C, Moffet H, Maltais DB. Relationships between lower limb muscle strength and locomotor capacity in children and adolescents with cerebral palsy who walk independently. Phys Occup Ther Pediatr 2012 Aug;32(3):320-32.

(6) Wiley ME, Damiano DL. Lower-extremity strength profiles in spastic cerebral palsy. Dev Med Child Neurol 1998 Feb;40(2):100-7.

(7) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014 Feb;35(2):239-49.

(8) Moreau NG, Falvo MJ, Damiano DL. Rapid force generation is impaired in cerebral palsy and is related to decreased muscle size and functional mobility. Gait Posture 2012 Jan;35(1):154-8.

(9) Verschuren O, Maltais DB, Douma-van Riet D, Kruitwagen C, Ketelaar M. Anaerobic performance in children with cerebral palsy compared to children with typical development. Pediatr Phys Ther 2013;25(4):409-13.

(10) Lloyd RS, Faigenbaum AD, Stone MH, Oliver JL, Jeffreys I, Moody JA, et al. Position statement on youth resistance training: the 2014 International Consensus. Br J Sports Med 2014 Apr;48(7):498-505.

(11) Gage J, Schwartz M, Koop S, Novacheck T. The identification and treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009.

(12) Faigenbaum AD, Lloyd RS, Myer GD. Youth resistance training: past practices, new perspectives, and future directions. Pediatr Exerc Sci 2013 Nov;25(4):591-604.

(13) Moreau NG, Holthaus K, Marlow N. Differential adaptations of muscle architecture to high-velocity versus traditional strength training in cerebral palsy. Neurorehabil Neural Repair 2013 May;27(4):325-34.

(14) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability for running tests for measuring agility and anaerobic muscle power in children and adolescents with cerebral palsy. Pediatr Phys Ther 2007;19(2):108-15.

(15) Verschuren O, Bongers BC, Obeid J, Ruyten T, Takken T. Validity of the muscle power sprint test in ambulatory youth with cerebral palsy. Pediatr Phys Ther 2013;25(1):25-8.

(16) Steenbeek D, Ketelaar M, Lindeman E, Galama K, Gorter JW. Interrater reliability of goal attainment scaling in rehabilitation of children with cerebral palsy. Arch Phys Med Rehabil 2010 Mar;91(3):429-35.

(17) Steenbeek D, Gorter JW, Ketelaar M, Galama K, Lindeman E. Responsiveness of Goal Attainment Scaling in comparison to two standardized measures in outcome evaluation of children with cerebral palsy. Clin Rehabil 2011 Dec;25(12):1128-39.

(18) McDowell BC, Kerr C, Parkes J, Cosgrove A. Validity of a 1 minute walk test for children with cerebral palsy. Dev Med Child Neurol 2005 Nov;47(11):744-8.

(19) McDowell BC, Humphreys L, Kerr C, Stevenson M. Test-retest reliability of a 1-min walk test in children with bilateral spastic cerebral palsy (BSCP). Gait Posture 2009 Feb;29(2):267-9.

(20) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. Phys Ther 2006 Aug;86(8):1107-17.

(21) Avery LM, Russell DJ, Raina PS, Walter SD, Rosenbaum PL. Rasch analysis of the Gross Motor Function Measure: validating the assumptions of the Rasch model to create an interval-level measure. Arch Phys Med Rehabil 2003 May;84(5):697-705.

(22) Russell DJ, Avery LM, Rosenbaum PL, Raina PS, Walter SD, Palisano RJ. Improved scaling of the gross motor function measure for children with cerebral palsy: evidence of reliability and validity. Phys Ther 2000 Sep;80(9):873-85.

(23) Harvey AR, Morris ME, Graham HK, Wolfe R, Baker R. Reliability of the functional mobility scale for children with cerebral palsy. Phys Occup Ther Pediatr 2010 May;30(2):139-49.

(24) van Ravesteyn NT, Dallmeijer AJ, Scholtes VA, Roorda LD, Becher JG. Measuring mobility limitations in children with cerebral palsy: interrater and intrarater reliability of a mobility questionnaire (MobQues). Dev Med Child Neurol 2010 Feb;52(2):194-9.

(25) van Ravesteyn NT, Scholtes VA, Becher JG, Roorda LD, Verschuren O, Dallmeijer AJ. Measuring mobility limitations in children with cerebral palsy: content and construct validity of a mobility questionnaire (MobQues). Dev Med Child Neurol 2010 Oct;52(10):e229-e235.

(26) van Vulpen LF, De GS, Becher JG, de Wolf GS, Dallmeijer AJ. Feasibility and test-retest reliability of measuring lowerlimb strength in young children with cerebral palsy. Eur J Phys Rehabil Med 2013 Dec;49(6):803-13.

(27) van den Noort JC, Scholtes VA, Harlaar J. Evaluation of clinical spasticity assessment in cerebral palsy using inertial sensors. Gait Posture 2009 Aug;30(2):138-43.

(28) Graham JE, Karmarkar AM, Ottenbacher KJ. Small sample research designs for evidence-based rehabilitation: issues and methods. Arch Phys Med Rehabil 2012 Aug;93(8 Suppl):S111-S116.

Improved walking capacity and muscle strength after functional power-training in young children with cerebral palsy

Liesbeth F. van Vulpen
Sonja de Groot
Eugene A.A. Rameckers
Jules G. Becher
Annet J. Dallmeijer

4

Neurorehabil Neural Repair 2017 Sep;31(9):827-41

Abstract

Background Strength training programs for children with cerebral palsy (CP) showed inconclusive evidence for improving walking, despite improvements in strength. Recent studies have suggested that strength training with high movement velocity is more effective for improving walking than traditional resistance training.

Objective The purpose of this study was to evaluate the effect of functional high velocity resistance training (power-training) to improve muscle strength and walking capacity of children with CP.

Method 22 children with spastic CP participated (13 bilateral, GMFCS level I (N=10) and II (N=12), 7.5 years (SD 1.8, range 4–10 y)). Within-subjects, changes in a 14-weeks usual care period were compared with changes in a 14-week functional power-training period (in groups, 3x/wk). Outcome measures were the muscle power sprint test (MPST), 1-minute walk test (1MWT), 10-m shuttle run test (SRT), gross motor function (GMFM-66), isometric strength of lower-limb muscles and dynamic ankle plantar flexor strength.

Results Changes during the training period were significantly larger than changes in the usual care period for all outcome measures ($p<.05$). Large improvements were found during the training period for walking capacity (ΔMPST [mean]: 27.6W [95% CI 15.84–39.46, 83% increase], Δ1MWT: 9.4m [95% CI 4.17–14.68, 13%], ΔSRT: 4.2 [95% CI 2.57–5.83, 56%], ΔGMFM-66: 5.5 [95% CI 3.33–7.74, 7%]) and muscle strength (18–128%), while outcomes remained stable in the usual care period.

Conclusions The results indicate that functional power-training is an effective training for improving walking capacity in young children with cerebral palsy.

Trial registration NTR5189.

Introduction

Children with cerebral palsy (CP) have significantly impaired walking capacity because of their motor impairments (1). These motor impairments are multi-factorial (spasticity, loss of selectivity and muscle weakness) (1). Ambulant children with spastic CP (60–70%) are integrated in community schools and recreational activities and need to perform the same activities as their typical developing (TD) peers, such as playing in the schoolyard, participating in physical education class and taking part in school outings (2;3). However, children with CP often experience difficulties keeping up with their peers in these daily activities because of their insufficient walking capacity (4).

A key aspect of the decreased walking capacity in children with CP is lower-limb muscle weakness: children with CP only have about 36% to 82% of the muscle strength of TD children (5). Compared to TD children, they produce even less muscle power in the distal muscle groups (the plantar flexors) than in the more proximal muscle groups (6;7). This power production of plantar flexor muscles is important for step length and walking speed by generating ankle power at push-off (8). Dallmeijer et al. (9) showed that, in comparison with TD children, ankle power generated at push-off is indeed reduced in children with CP by more than 40% during gait (9).

Several methods have been used to increase muscle strength to enhance the walking capacity of children with CP; the most commonly used method is progressive resistance exercise training (PRE) (10). However, despite an increase in strength in most lower-limb muscles, this training method did not improve the walking capacity of children with CP (10;11). Apparently the newly gained muscle strength does not transfer to functional improvements in walking. Moreau et al. (12) suggested that to get functional improvements, the strength training velocity has to be at a higher, more functional movement velocity than generally used in PRE training. This suggestion is based on their findings that children with CP have a reduced capacity to rapidly generate forces (12). This capacity is especially needed in playing activities such as running and sprinting games on the playground and in sports (13). Previous studies have in fact shown reduced sprinting capacity (as part of walking capacity) in children with CP (13;14).

Another possible reason for the limited effects of PRE on walking capacity is lack of training that is targeted to the plantar flexor muscles. This muscle group is more difficult to train in functional exercises, especially when children wear orthoses. Muscle weakness is already present at an early age in children with CP (5). At the same time, the plantar flexor muscles are hampered in gaining strength by several commonly used treatments

for children with CP, such as botulinum toxin injections and casting periods (1). While these treatments address the problem of plantar flexor muscle shortening, they are also likely to weaken these muscles or at least hamper muscle growth (15;16). Strengthening the plantar flexor muscles is therefore particularly important for children with CP.

To strengthen the plantar muscles and to improve walking capacity in children with CP, we developed a functional power-training program that consists of resistance training with exercises at high movement velocities. This functional power-training consists of loaded functional exercises, such as walking, running and climbing stairs, performed at high movement velocities (17).

The purpose of this study was to evaluate the effect of this functional power-training on walking capacity (sprinting capacity, walking speed, endurance) and muscle strength with a double-baseline design. We compared changes in walking capacity and muscle strength during a 14-week usual care period with changes in walking capacity and muscle strength during a 14-week functional power-training period.

Methods

Participants

Inclusion criteria were ambulant children aged 4–10 years, with a spastic CP, and Gross Motor Function Classification System (GMFCS) I and II. Parents and/or the children had a treatment question related to walking capacity (such as being able to walk longer or faster). The children had to be able to understand and follow instructions. The exclusion criteria were treatment with botulinum toxin A in lower limb and/or serial casting of lower limb less than 6 months before the start of the functional power-training and selective dorsal rhizotomy treatment less than a year before the functional power-training. Children who did not (yet) choose walking as their preferred way of mobility were also excluded.

Design and procedure

This study had a double-baseline design, in which the participating children acted as their own controls: a 14-week usual care period was compared with a 14-week functional power-training intervention that followed immediately after the usual care period. Measurements were performed before the usual care period (*Pre1*); after the 14wk usual care period (which was also the start of the training period) (*Pre2*); after the 14wk training period (*Post*); and 14 weeks after the post-test (*Follow-up*) to assess if the potential improvements

were maintained. In the usual care period and the follow-up period children followed their regular physical therapy sessions (one to two times a week, 30 min.) which differed from child to child. Detailed descriptions of the design and procedure of the study can be found in Van Vulpen et al. (2016) (17).

The Medical Ethics Committee, Amsterdam, the Netherlands, approved this study, and written informed consent forms were obtained from the parents of each participant.

Setting

Participants were recruited from a rehabilitation center, two special needs schools for children with physical disabilities and an outpatient clinic of a university medical center. The training and assessments took place in the two special needs schools for physically disabled children and the rehabilitation center.

Intervention: functional power-training

During the intervention period, children followed the functional power-training program. The training period was 14 weeks, with sessions 3 times a week and each training session lasting 60 minutes. This was based on strength training recommendations for improving muscle strength in children with or without CP (18-21). The functional power-training replaced the conventional physiotherapy program if there was one. The power-training was specifically designed to strengthen the plantar flexor muscles during functional exercises and consisted of the following phases: a warm-up (10 min), three to four different power exercises (35 min) and an end game (15 min).

The training sessions were in small groups (3–6 children), with one supervising therapist for each child (3 to 6 therapists). Children wore regular sport shoes without orthoses. Each training session started with a warm up with walking and running exercises and dynamic calf muscle stretching exercises. A story about super heroes and secret missions kept the children motivated to give their best effort. They all received their own T-shirt with their super hero to stimulate the group feeling and training motivation.

Power exercises

All participants received three to six different power exercises for improving daily life activities (Table 4.1), chosen in line with the treatment goals set by their parents. The children performed three to four of the six different power exercises in each session (Table 4.1). Key elements of the power exercises were functional loaded multi-joint exercises like running and walking with a focus on the ankle push-off, high movement velocity (similar

Table 4.1 Characteristics and training volume of the six different power exercises and progression of each exercise during the training period

						Progression during training period			
Exercise	Velocity	Sets	Resistance	Duration	Rest	N[5]	Start[3] Mean, kg (SD)	End[4] Mean, kg (SD)	%
Running	Running movement is retained to 50–70% of maximum running speed[1]	6 to 8 reps	Dragging a loaded box over-ground with belt around the hips	25s	30–50s	22	7.7 (5.4)	14.5 (6.3)	87
Walking	Walking movement is retained to 50–70% of walking speed[2]	6 to 8 reps	Dragging a loaded box over-ground with belt around the hips	25s	30–50s	11	15.9 (11.6)	27.3 (11.6)	72
Pushing chair	Running movement is retained to 50–70% of maximum running speed[1]	6 to 8 reps	Chair, with a loaded box underneath	25s	30–50s	22	4.4 (5.5)	10.4 (6.6)	139
Stair climbing	Climbing stairs movement is retained to 50–70% of maximal amount of steps possible in 25s	6 to 8 reps	Loaded vest	25s	30–50s	22	3.9 (2.6)	6.9 (3.2)	79
Propelling a stable scooter	Movement is retained to 50–70% of maximal speed, determined for each leg separately	6 to 8 reps	Loaded box attached to scooter	25s	30–50s	17 15	M[6]: 3.6 (2.8) L[7]: 4.0 (2.8)	9.7 (2.8) 9.3 (3.7)	169 139
Sideways walking	Movement is retained to 50–70% of maximum sideways walking speed	6 to 8 reps	Dragging a loaded box over-ground with belt around the hips	25s	30–50s	3 2	M[6]: 8.9 (0.6) L[7]: 10.1 (1.1)	17.3 (6.7) 17.3 (8.8)	94 71

[1] Maximum running speed is obtained with the 6 times 15-meter Muscle Power Sprint Test.
[2] Maximum walking speed is obtained with the 1-minute walk test.
[3] Start load determined at 3 weeks of training period.
[4] End load determined at the end of the 14 weeks training period.
[5] N is number of children that performed the exercise during the training period.
[6] Most affected leg.
[7] Least affected leg.

to the velocity used in daily/playing activities) and progressive load. The exercises were performed at 50–70% of their maximum unloaded speed by retaining the movement with load. Each exercise was performed during 25s on maximal effort, with a resting period of 30–50s, and six to eight repetitions each exercise.

The training volume was determined by load, velocity and number of repetitions (Table 4.1). When children became faster and performed the distance of the exercise under 25s, the load was increased with 10%. A more detailed description of the training protocol is described elsewhere (17).

Measurements

Walking capacity

The primary outcome measure of walking capacity was the Muscle Power Sprint Test (MPST). The MPST measures the sprint capacity of the child expressed in mean power and peak power (22). This test is reliable (ICC=0.97) with good feasibility and construct validity (22). Children sprint at maximum speed over 15 meters with six repetitions, with a 10s break in between to turn around for the next 15 meters sprint as described in the protocol of the test. The power output for each of the six sprints was estimated from the collected data using the following equations: 1) *velocity (m/sec)* = 15 *meter/time*, 2) *acceleration (m/sec²) = velocity/time*, 3) *force (kg·m/sec2) = body mass * acceleration*, 4) *power (W) = force * velocity* (22). Peak power was defined as the highest power output of the six runs. Mean power was defined as the average power output of the six runs.

The secondary outcome measures on walking capacity were walking speed, measured with the 1-minute walk test (1MWT), and walking endurance, measured with the 10 meter Shuttle Run Test (SRT). The 1MWT measures the distance walked in one minute. The children were asked to walk as fast as possible without running around an oval track. The reliability (ICC=0.94) and validity of the 1MWT are good (23;24). The 10 meter SRT, made for children with CP GMFCS level I and II, measures endurance. Children walked or ran between two markers that were 10 m apart, at a set incremental speed determined by a signal. Children with GMFCS level I performed the SRT-I with starting speed at 5km/h; children with GMFCS level II performed the SRT-II with starting speed at 2km/h. Both SRT tests increase 0.25km/h in speed every minute. Reliability of the SRT-I and SRT-II is good (SRT-I, ICC=0.97 and SRT-II, ICC=0.99) (25).

Gross motor function was measured with the 66-item version of the Gross Motor Function Measure (GMFM-66). Items that were tested included activities in walking, running and

jumping skills. There is a 4-point scoring system for each item converted to an interval scale (range 0–100 points). The GMFM-66 is a standardized tool designed to evaluate change in gross motor function in children with CP; it has been internationally validated (26;27).

Muscle strength

The isometric muscle strength of the plantar flexors (with knees extended and with knees flexed), knee extensors and hip abductors were measured. The make-method was used, (microFET, Biometrics BV, Almere, The Netherlands) for about 5s. Each child subsequently performed three repetitions for each muscle group, and the maximum force (peak force) for each repetition was registered. A fourth or fifth repetition was performed if the concentration and/or motivation of the child were not optimal.

A standardized protocol was used for positioning of the child, joint fixation and joint positioning (28). Torque (Nm) was calculated by multiplying force (Newton) by the length (meter) of the lever arm. To improve reliability, isometric muscle strength was measured at two different test occasions (different days, with a maximum of 7 days within measurements). The mean of these six measurements (two test occasions with three repetitions each) was used in the analysis. Isometric strength measurements have good reliability when measured with three repetitions in two different test occasions (28).

The dynamic muscle strength of the plantar flexors was measured with the standing heel-rise test on one limb. The most and least affected limb were both tested with a standardized protocol (28). Moderate to good reliability was found for the heel-rise test in young children with CP (ICC 0.86–0.98) (28).

Control outcomes

Each child was weighed on a mechanical scale (Seca 761 mechanical scale, Seca BV, Hamburg, Germany) and body height was measured with a height scale (Seca 217 Stadiometer, Seca BV, Hamburg, Germany) with the child standing against a wall. GMFCS and type of CP were determined by a pediatric physiotherapist together with the physician. To determine the most and least affected leg of each child, selective motor control of knee extension and ankle dorsiflexion were tested by the modified Trost Selective Motor Control test (29). The modified Trost Selective Motor Control test has three levels of control: 0, no ability to perform isolated movement; 1, partial ability to perform isolated movement; and 2, complete ability to perform isolated movement (29). Passive ankle dorsiflexion range of motion and popliteal angle measurements were performed with a goniometer with the child in supine position (8).

Statistical analysis

Sample size

A power calculation was performed for the primary outcome measure, the Muscle Power Sprint Test (W). A pilot study (N=10) showed an increase of 85% (mean±SD: 13.1±12.2 W) after functional power-training. Calculations were based on a within-subject design with a dependent t-test, a power of 0.8, alpha of 0.05 and an effect size 0.7. According to the power calculation, a sample size of at least 19 children was needed. Twenty-two children were recruited to allow a dropout of 10%.

Mean and standard deviations were calculated of walking capacity and muscle strength at each measurement occasion (*Pre1*, *Pre2*, *Post* and *Follow-up*). Changes in walking capacity and muscle strength during the usual care period (Δ *Pre2–Pre1*), functional power-training period (Δ *Post–Pre2*) and follow-up period (Δ *Follow-up–Post*) were calculated. Paired sampled *t* tests (if normally distributed) and Wilcoxon signed rank tests (if not normally distributed) were used to determine the possible intervention effect by comparing the changes during the functional power-training period with changes in the usual care. All statistical analyses were performed using SPSS Statistics 22.0 software (IBM Corporation, New York, USA).

Results

Characteristics of the study population

Twenty-two children participated in this study. Personal characteristics of the participants are presented in Table 4.2. Of 22 participants, three had interfering treatment with the protocol (serial casting for 1 to 3 weeks) in their usual care period, and one participant had serial casting (3 weeks) in the follow-up period. As an intention-to-treat analyses, these four participants were included in the analyses. One participant was hospitalized during the follow-up measurements because of a medical problem that was unrelated to the training and therefore missed the follow-up test. Three participants had a surgical intervention (selective dorsal rhizotomy without muscle lengthening) more than a year before start of the usual care period.

Training compliance

The children participated in 88% (range: 57–100%) of the scheduled training sessions. One child missed 33% of all possible training sessions because of logistic problems of

Table 4.2 Characteristics of the participants

Boys (N)	11
Girls (N)	11
Age (years), mean (SD)	7.5 (1.8)
Range (years)	4.0–10.2
Body mass (kg), mean (SD)	26.0 (8.2)
Range (kg)	14.0–45.5
Height (m), mean (SD)	1.24 (0.11)
Range (m)	0.99–1.45
GMFCS[1] I (N)	10
GMFCS II (N)	12
Unilateral spastic CP[2] (N)	9
Bilateral spastic CP (N)	13
Regular school (N)	13
Special needs education (N)	9
Intervention period in springtime (N)	11
Intervention period in autumn (N)	11
AFO[3] in daily live (N)	13
No AFO in daily live (N)	9

[1] GMFCS, Gross Motor Function Classification System (32).
[2] SCPE, Surveillance of Cerebral Palsy in Europe, classification of subtypes CP (2).
[3] AFO, ankle foot orthosis on one limb or both. Supramalleolar orthosis and orthopedics shoes or registered as no AFO in daily life.

the parents. Another child missed 43% of the 14-wk functional power-training because the family moved at the end of the training period. Other reasons for not attending the training were illness, vacation, medical appointment, family visits or unknown.

Training progression

Load increased significantly in the power exercises over the intervention period. It took one to three weeks for the children to get used to the exercises, and set the appropriate training load. Children showed increases in the load they could pull, bear or push in the power exercises in the 14th week compared with the 3th week in the training period, ranging from 72% in walking to 170% in propelling the scooter (Table 4.1).

Walking capacity

There were two missing values for the 1-minute walk test: one participant could not be tempted to walk instead of running during the post-test, and one participant did not bring the same footwear at post test.

Table 4.3 shows the intervention effect by comparing the changes during the functional power-training period with the changes in the usual care period within participants: all walking capacity variables showed significant improvements during the functional power-training.

Table 4.4 shows walking capacity variables of all measurement moments. We found no significant changes in the walking capacity variables in the usual care period (from *Pre 1* to *Pre 2* measurement) except for a small decline in peak power on the MPST (41.0 W at *Pre 1* to 37.3 W at *Pre 2*, p=.017). All walking capacity variables showed a significant increase in the *functional power-training* period (from *Pre 2* to *Post*). In the follow-up period the MPST showed a decline in mean power (from *Post*: 57.1 W to Follow-up: 52.0 W, p=.041) and in peak power (from *Post*: 67.4 W to Follow-up: 60.8 W, p=.037); however, compared with the measurement at the start of the training (*Pre 2*), they still showed a higher mean power (from *Pre 2*: 31.2 W to Follow-up: 52.0 W, p<.001) and peak power (from *Pre 2*: 37.3 W to Follow-up: 60.8 W, p<.001). Similar results were seen in the SRT, while walking speed (1MWT) maintained the training effect with no decline after Follow-up.

The individual changes of the participants in the MPST and the 1-minute walk test in the usual care period and the *functional power-training* period are illustrated in Figure 4.1A and 4.1B.

Muscle strength

One participant could not perform the isometric plantar flexor muscle strength test (knees flexed and knees extended) of the most affected limb because of motor control problems.

All muscle strength variables showed significant increases during the *functional power-training* compared with the usual care period (Table 4.3).

There were no changes in the muscle strength variables in the usual care period; isometric strength of all muscle groups and dynamic muscle strength increased significantly in the *functional power-training* period (Table 4.4). There were no changes in muscle strength in the follow-up period except for the plantar flexors tested with the knees flexed which showed a small further increase (from 14.9 Nm at *Post* to 15.8 Nm at Follow-up, p=.015).

The individual changes of the isometric and dynamic muscle strength of the participants in the plantar flexor muscles are illustrated in Figure 4.2.

Table 4.3 Mean (±SD) changes during the usual care period, intervention period, and intervention effect (differences between changes in usual care period and changes in intervention period)

	Change in usual care period (Pre2-Pre1)		Change in functional power-training period (Post-Pre2)		Change intervention - change usual care (effect functional power-training)	
	n	Mean (SD)	n	Mean (SD)	Mean (95% CI)	p-value
Walking capacity						
Muscle Power Sprint Test Average time (s)	22	0.14 (0.68)	22	-1.47 (1.37)	-1.61 (-2.37–-0.84)	<.001
Muscle Power Sprint Test Mean power (W)	22	-1.75 (4.37)	22	25.85 (23.50)	27.60 (15.84–39.46)	<.001
Muscle Power Sprint Test Peak power (W)	22	-3.71 (6.67)	22	30.18 (30.14)	33.88 (18.42–49.35)	<.001
1-Minute Walk (m)	20	-0.17 (8.56)	20	9.26 (8.92)	9.42 (4.17–14.68)	.001
Shuttle Run Test I & II (minutes)	22	-0.17 (8.56)	22	3.52 (2.47)	4.21 (2.57–5.83)	<.001
GMFM-66	22	-0.06 (2.39)	22	5.48 (3.24)	5.54 (3.33–7.74)	<.001
Isometric muscle strength, Nm Most affected leg[1]						
Plantar flexors with knees extended (Nm)	21	0.73 (2.35)	21	5.33 (2.35)	4.62 (2.92–6.32)	<.001
Plantar flexors with knees flexed (Nm)	21	0.48 (2.19)	21	3.35 (1.47)	2.99 (1.57–4.41)	<.001
Knee extensors (Nm)	22	1.32 (4.81)	22	4.45 (3.34)	3.19 (0.43–5.95)	.026
Hip abductors (Nm)	22	0.83 (3.46)	22	6.54 (5.30)	5.52 (2.11–8.93)	.003

	Change in usual care period (Pre2-Pre1)		Change in functional power-training period (Post-Pre2)		Change intervention - change usual care (effect functional power-training)	
	n	Mean (SD)	n	Mean (SD)	Mean (95% CI)	p-value
Isometric muscle strength, Nm Least affected leg[1]						
Plantar flexors with knees extended (Nm)	22	0.74 (3.18)	22	5.07 (3.62)	4.32 (1.77–6.87)	.002
Plantar flexors with knees flexed (Nm)	22	0.96 (2.61)	22	3.45 (2.70)	2.37 (0.25–4.49)	.030
Knee extensors (Nm)	22	1.51 (4.57)	22	4.61 (3.72)	3.10 (0.41–5.79)	.026
Hip abductors (Nm)	22	0.82 (3.81)	22	6.62 (5.45)	5.99 (2.38–9.59)	.002
Dynamic muscle strength plantar flexors Standing Heel Rise Test						
Most affected leg, number (repetitions)	22	0.16 (4.62)	22	7.52 (6.03)	7.36 (4.07–10.66)	<.001[2]
Least affected leg, number (repetitions)	22	-0.57 (4.27)	22	8.30 (8.59)	8.86 (4.04–13.68)	.002[2]

[1] Most and least affected leg was determined by using the modified Trost's SMC test for selective motor control for the ankle dorsal flexion possibility (29).
[2] Tested with Wilcoxon signed rank test.

Table 4.4 Pre, Post and Follow-up values (Means and standard deviation (SD)) for walking capacity -and muscle strength variables

	Pre1[1]	Pre2[1]	Post training[1]	Follow-up[1]	Pre2-Pre1		Post-Pre2		Follow-up-Post		Follow-up-Pre2	
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	n	p-value	n	p-value	n	p-value	n	p-value
Walking capacity												
Muscle Power Sprint Average time (s)	6.9 (2.9)	7.1 (3.0)	5.6 (2.1)	5.8 (2.0)	22	.347	22	<.001	21	.074	21	<.001
Muscle Power Sprint Mean power (W)	33.0 (23.7)	31.2 (22.8)	57.1 (39.2)	52.0 (36.7)	22	.074	22	<.001	21	.041	21	<.001
Muscle Power Sprint Peak power (W)	41.0 (29.4)	37.3 (26.5)	67.4 (48.0)	60.8 (43.8)	22	.017	22	<.001	21	.037	21	<.001
1-Minute Walk (m)	74.1 (16.6)	73.4 (15.2)	82.3 (12.8)	82.2 (12.5)	21	.708	20	<.001	19	.824	20	<.001
Shuttle Run Test I & II (minutes)	7.0 (3.3)	6.3 (3.0)	9.8 (4.1)	8.4 (3.5)	22	.075	22	<.001	21	<.001	21	<.001
GMFM-66	78.2 (7.9)	78.1 (7.9)	83.6 (7.1)	83.5 (8.0)	22	.909	22	<.001	21	.912	21	<.001
Isometric muscle strength, Nm Most affected leg[2]												
Plantar flexors with knees extended (Nm)	14.5 (7.6)	15.2 (7.4)	20.5 (9.1)	20.3 (8.5)	21	.185	21	<.001	20	.943	20	<.001
Plantar flexors with knees flexed (Nm)	7.5 (4.2)	8.1 (4.2)	11.7 (5.0)	12.1 (5.2)	21	.190	21	<.001	20	.239	20	<.001
Knee extensors (Nm)	22.2 (8.8)	23.4 (10.2)	27.8 (10.9)	26.7 (10.3)	22	.243	22	<.001	21	.241	21	<.001
Hip abductors (Nm)	14.1 (7.0)	14.9 (8.0)	21.3 (10.9)	19.3 (7.8)	22	.264	22	<.001	21	.542	21	<.001

	Pre1[1]	Pre2[1]	Post training[1]	Follow-up[1]	Pre2-Pre1		Post-Pre2		Follow-up-Post		Follow-up-Pre2	
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	n	p-value	n	p-value	n	p-value	n	p-value
Isometric muscle strength, Nm Least affected leg[2]												
Plantar flexors with knees extended (Nm)	18.3 (7.7)	19.0 (6.6)	24.1 (9.4)	23.7 (7.3)	22	.276	22	<.001	21	.738	21	<.001
Plantar flexors with knees flexed (Nm)	10.8 (6.0)	11.7 (5.2)	14.9 (5.6)	15.8 (5.6)	22	.164	22	<.001	21	.015	21	<.001
Knee extensors (Nm)	24.8 (10.3)	26.4 (10.8)	31.0 (12.2)	29.8 (12.0)	22	.136	22	<.001	21	.531	21	.006
Hip abductors (Nm)	15.3 (6.9)	16.1 (7.8)	22.8 (10.5)	20.8 (7.6)	22	.335	22	<.001	21	.391	21	<.001
Dynamic muscle strength plantar flexors Standing Heel Rise Test												
Most affected leg, number (repetitions)	4.4 (6.7)	4.6 (5.5)	12.1 (9.1)	10.7 (8.7)	22	.451[3]	22	<.001[3]	21	.445[3]	21	.001[3]
Least affected leg, number (repetitions)	12.6 (9.4)	12.0 (9.6)	20.3 (9.0)	17.6 (9.3)	22	.796[3]	22	.001[3]	21	.127[3]	21	.011[3]

[1] Pre1 is at the start of the 14 weeks *usual care* period. Pre2 is at the end of the of the *usual care* period which is also the start of the 14 weeks f*unctional power-training* period. Post training is at the end of the *functional power-training* period which is also the start of the 14 weeks *follow-up period*. Follow-up is at the end of the 14 weeks follow-up period.
[2] Most and least affected leg was determined by using the modified Trost's SMC test for selective motor control for the ankle dorsal flexion possibility (19).
[3] Tested with Wilcoxon signed rank test.

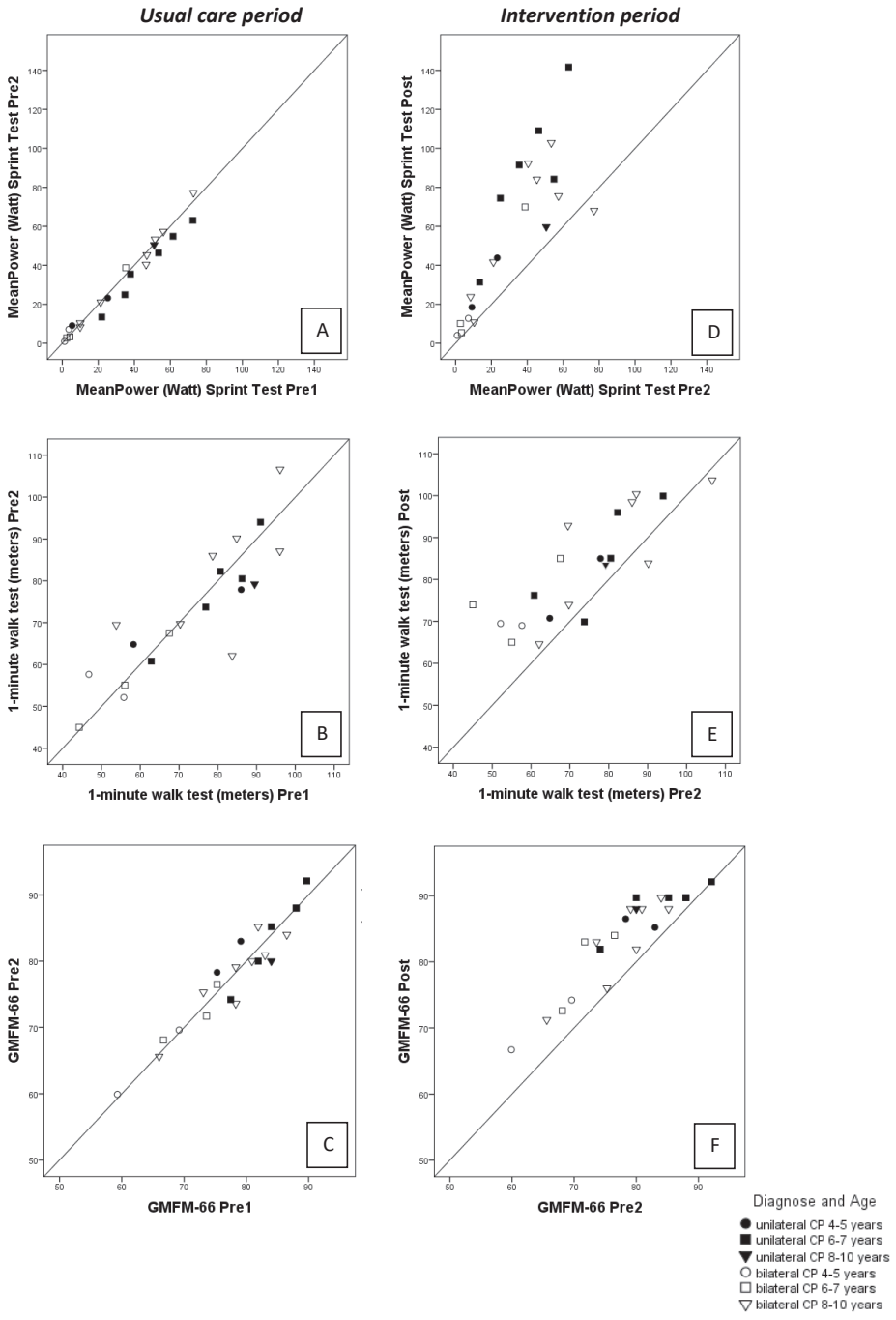

Figure 4.1 Individual changes in walking capacity variables and Gross Motor Function in *usual care* period (A sprint capacity, B walking speed, C gross motor function) and in *functional power-training* period (D, E, F).

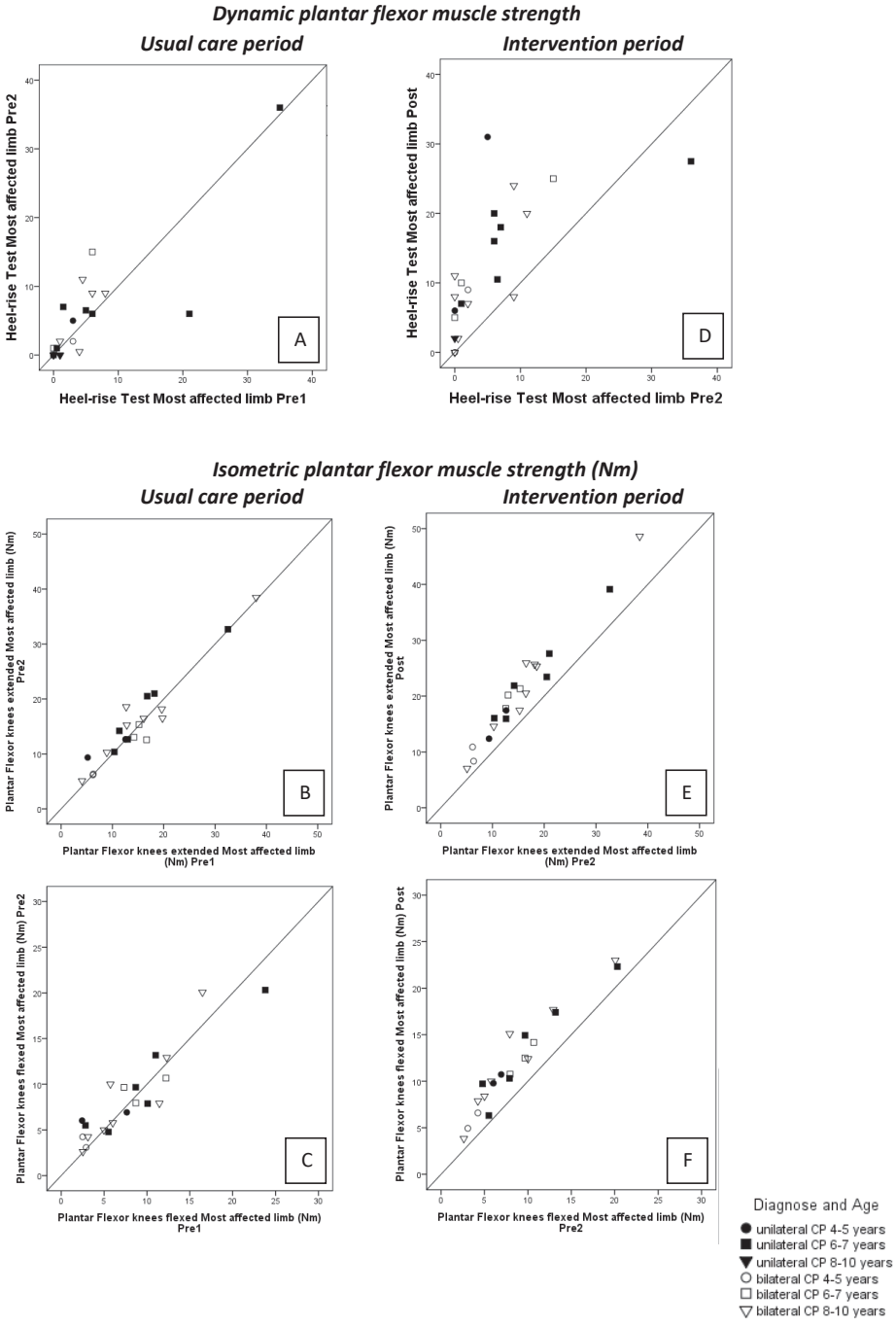

Figure 4.2 Individual changes in muscle strength variables in *usual care* period (dynamic strength (A) and isometric Nm (B: plantar flexor knees extended and C: plantar flexor knees flexed)) and in *functional power-training* period (D, E, F).

Adverse events

There were no adverse events associated with the *functional power-training*. Two participants complained of foot pain in the first weeks of the training period. One participant had ankle pain during one week of the training period because of a fall at school, and one participant had knee pain because of activities in the weekend unrelated to the training. These four children were examined by their physicians, who found no serious injuries. The children did not miss any training session due to their foot or ankle pain.

No muscle shortening was found after the *functional power-training*. Passive range of motion at the ankle joint and popliteal angle measurements are described in Appendix 4.1a and 4.1b.

Discussion

The results indicate that power-training increases walking capacity and muscle strength. All outcome measures on the walking capacity showed substantial increases ranging from 13 to 83% after the *functional power-training*. Previous studies into conventional strength training programs have not found significant effects on the walking capacity of children with CP (10;11). The main differences between these conventional strength training programs and our functional power-training program is the specificity of the strength training: we used a higher movement velocity in the progressive loaded strength training exercises and incorporated strength exercises in functional movements like walking and sprinting.

The children in our study showed an increase of 83% in sprint capacity after the *functional power-training*, measured with the Muscle Power Sprint Test (MPST), which was our primary outcome. In the follow-up period a small but significant reduction (8.9%) in sprint capacity was found. Verschuren et al. (2007) found an increase of 25% in sprint capacity, also measured with the MPST, after a functional training program of 8 months, 2 times a week for 45 minutes, in children with CP (7–18y) (30). Their intervention had some factors that could be compared with our training intervention: they incorporated many running exercises with repeated bouts of 20–30 s, also used group training and they had the same training frequency (48 versus 42 training hours). The difference between the training programs was that our *functional power-training* program incorporated external load in the running exercises by letting the children push a weighted-chair and drag a weighted-box, thus ensuring enough resistance in the exercises for muscle strengthening. The added progressive load in the exercises may have partly induced the larger increase

in sprint capacity in our study. Another difference is that we trained younger children (4–10 years old in our study versus 7–18 years), who may show more neural and muscular plasticity (31) and may therefore be more sensitive to changes in motor coordination necessary for increasing their sprint capacity (20).

The children in our study showed an increase of 13% in their walking velocity, measured with the 1-minute walk test. This increase in walking velocity is larger than earlier increases of 4%, considered clinically relevant (32), in a group of children with CP (9–17 y) who received a high-velocity strength training (33). Our findings support the suggestion that the velocity in strength training exercises has to be higher than generally used in conventional strength training exercises in order to get functional improvements. Our results also suggest that adding specificity of strength exercises, i.e. incorporation of the strength exercise in functional movements, further increases these improvements (34).

The specificity of the strength training exercises in our study—which ensured that the power generated by the muscles was directly related to walking capacity—has the potential of training motor performance such as coordination, strength and endurance (19). Improved coordination can be expected as a result of the repetitiveness of the functional exercises, which help refine the efficient motor patterns that are required for optimal functional performance like walking, sprinting and climbing stairs (35). In the present study we combined this principle with progressive exercise, which may explain the large increases of 83% in sprint capacity. We also found an increase of 56% for the shuttle run test during the trainings period and a decrease of 14% in the follow-up period. A possible explanations for this increase is that the specificity of the strength exercises may have led to improved coordination of the muscles, which in turn may have led to an improved walking economy of the children, with a lower aerobic demand at equal velocity (36). Another explanation may lie in the similarity of our training to that of high intensity interval training (HIIT). Previous studies have shown that HIIT increases both the sprint and endurance in unimpaired subjects (37) and children with CP (30). These improvements may be the result of peripheral adaptations within the muscles (i.e. increased strength) rather than central adaptations (within the cardiopulmonary system) (38).

We found larger improvements in plantar flexor muscles strength in the more affected—weaker—limb than in the less affected limb (isometric strength 34% increase in more affected limb versus 27% increase in less affected limb and dynamic strength 128% versus 80%). This contrasts previous information that suggests that resistance training programs with multi-joint exercises strengthening of the target (weaker) muscles is prevented by compensation of other (less affected) muscle groups (19;21). Such compensations would

yield larger improvements in muscle strength of the less affected limb, but we found the opposite. This could be explained by the specific and progressive character of the strength exercises; the children were forced to produce power with both legs. It is unknown to what extent strength improvements in other non-measured muscle groups, e.g. hip flexors and extensors and the knee flexors, have contributed to the observed functional improvements. To reduce the burden for the children we limited strength assessments to the plantar flexors, knee extensors and hip abductors.

Some, but not all, of the functional effects persisted in the follow-up period. Although we found a significant decrease in sprint capacity and in the shuttle run test, participants still had an increase in sprint capacity (67%) and in the shuttle run test (33%) at the end of the follow up period compared with the start of the *functional power-training* period. The increases in muscle strength were maintained in the follow-up period.

Some authors suggested that muscle tightness after muscle strengthening exercises might occur and consequently might have an adverse effect on gait (39). In our study, however, a significant increase in the ankle range of motion (ROM) with knees extended (m. gastrocnemius length) was found after the training period which decreased in the follow-up period. The changes are too small to be of clinical relevance but could be relevant for understanding the changes in muscle morphology after resistance exercise. Some of the exercises provoked the children to make larger steps than usual and to lean forward, which might have provided repeated dynamic stretches in the gastrocnemius muscles. A previous study with inclined treadmill gait training in adults with CP found similar ROM improvements (40). Increased muscle length can also be explained by muscle fiber hypertrophy, which would lead to increased muscle length in highly pennate muscle like the m. gastrocnemius (41). These findings indicate that the power training did not negatively affect muscle length, but may even have a positive effect on ROM of the ankle.

Clinical significance

Walking capacity is important for children with CP, and it is often the aim of muscle strength training programs. There is, however, inconclusive evidence that strength training programs improve walking capacity. Nevertheless, our study suggests that strengthening interventions are effective in improving muscle strength *and* walking capacity in young children with CP if they involve high-velocity strengthening exercises incorporated in functional movements of sufficient intensity and volume (maximal effort, 3 sets of 6 to 8 repetitions), frequency (3 times a week) and duration (14 weeks). Our results also suggest that targeting the plantar flexor muscles might improve walking and sprinting performance.

Limitations

We used a double-baseline design instead of a randomized-control trial (RCT) to increase the feasibility of our study in a heterogeneous group of children with CP. In this design the children served as their own controls by comparing the changes in usual care period to the changes in during the *functional power training* period. As an RCT is considered the highest level of evidence, double-baseline could be considered as a limitation of the study. However, the double-baseline design has been described as a good alternative in evidence-based rehabilitation for heterogeneous patient populations (42), In addition, the number of children that are needed is less than in a RCT because similar statistical power can be reached with smaller subjects groups, which makes the double-baseline design more feasible and less expensive (42).

Conclusions

The results indicate that functional power-training is an effective training to improve walking capacity and muscle strength in young children with cerebral palsy. As hypothesized, functional strength training at higher movement velocities resulted in improvements in both walking capacity and muscle strength.

Acknowledgments

The authors thank all the children, parents and trainers who participated. This study was funded by Mitialto foundation, Duyvensz-Nagel foundation, Dutch Rehabilitation fund and Royal Dutch Society for Physical Therapy.

References

(1) Graham HK, Rosenbaum P, Paneth N, Dan B, Lin JP, Damiano DL, et al. Cerebral palsy. Nat Rev Dis Primers 2016;2:15082.

(2) Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. Pediatrics 2008 Jan;121(1):e187-e192.

(3) Gorter JW, Rosenbaum PL, Hanna SE, Palisano RJ, Bartlett DJ, Russell DJ, et al. Limb distribution, motor impairment, and functional classification of cerebral palsy. Dev Med Child Neurol 2004 Jul;46(7):461-7.

(4) Bax MC, Flodmark O, Tydeman C. Definition and classification of cerebral palsy. From syndrome toward disease. Dev Med Child Neurol Suppl 2007 Feb;109:39-41.

(5) Dallmeijer AJ, Rameckers EA, Houdijk H, De GS, Scholtes VA, Becher JG. Isometric muscle strength and mobility capacity in children with cerebral palsy. Disabil Rehabil 2015 Nov 25;1-8.

(6) Elder GC, Kirk J, Stewart G, Cook K, Weir D, Marshall A, et al. Contributing factors to muscle weakness in children with cerebral palsy. Dev Med Child Neurol 2003 Aug;45(8):542-50.

(7) Stackhouse SK, Binder-Macleod SA, Lee SC. Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. Muscle Nerve 2005 May;31(5):594-601.

(8) Gage J, Schwartz M, Koop S, Novacheck T. The identification and treatment of gait problems in cerebral palsy. London: Mac Keith Press; 2009.

(9) Dallmeijer AJ, Baker R, Dodd KJ, Taylor NF. Association between isometric muscle strength and gait joint kinetics in adolescents and young adults with cerebral palsy. Gait Posture 2011 Mar;33(3):326-32.

(10) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014 Feb;35(2):239-49.

(11) Franki I, Desloovere K, De CJ, Feys H, Molenaers G, Calders P, et al. The evidence-base for basic physical therapy techniques targeting lower limb function in children with cerebral palsy: a systematic review using the International Classification of Functioning, Disability and Health as a conceptual framework. J Rehabil Med 2012 May;44(5):385-95.

(12) Moreau NG, Falvo MJ, Damiano DL. Rapid force generation is impaired in cerebral palsy and is related to decreased muscle size and functional mobility. Gait Posture 2012 Jan;35(1):154-8.

(13) Verschuren O, Maltais DB, Douma-van RD, Kruitwagen C, Ketelaar M. Anaerobic performance in children with cerebral palsy compared to children with typical development. Pediatr Phys Ther 2013;25(4):409-13.

(14) Balemans AC, van WL, De Heer SJ, Van den Brink J, De Koning JJ, Becher JG, et al. Maximal aerobic and anaerobic exercise responses in children with cerebral palsy. Med Sci Sports Exerc 2013 Mar;45(3):561-8.

(15) Fortuna R, Vaz MA, Sawatsky A, Hart DA, Herzog W. A clinically relevant BTX-A injection protocol leads to persistent weakness, contractile material loss, and an altered mRNA expression phenotype in rabbit quadriceps muscles. J Biomech 2015 Jul 16;48(10):1700-6.

(16) Minamoto VB, Suzuki KP, Bremner SN, Lieber RL, Ward SR. Dramatic changes in muscle contractile and structural properties after 2 botulinum toxin injections. Muscle Nerve 2015 Oct;52(4):649-57.

(17) van Vulpen LF, de Groot S, Rameckers EA, Becher J, Dallmeijer AJ. Effectiveness of functional power training on walking ability in young children with cerebral palsy: study protocol of double-baseline trial. Pediatr Phys Ther 2017 Jul;29:275-82.

(18) Baechle TR, Earle RW. Essentials of Strength training and conditioning. third ed. Champaign, Il 61825-5076: Human Kinetics; 2008.

(19) Faigenbaum AD, Lloyd RS, Myer GD. Youth resistance training: past practices, new perspectives, and future directions. Pediatr Exerc Sci 2013 Nov;25(4):591-604.

(20) Lloyd RS, Faigenbaum AD, Stone MH, Oliver JL, Jeffreys I, Moody JA, et al. Position statement on youth resistance training: the 2014 International Consensus. Br J Sports Med 2014 Apr;48(7):498-505.

(21) Verschuren O, Ada L, Maltais DB, Gorter JW, Scianni A, Ketelaar M. Muscle strengthening in children and adolescents with spastic cerebral palsy: considerations for future resistance training protocols. Phys Ther 2011 Jul;91(7):1130-9.

(22) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability for running tests for measuring agility and anaerobic muscle power in children and adolescents with cerebral palsy. Pediatr Phys Ther 2007;19(2):108-15.

(23) McDowell BC, Kerr C, Parkes J, Cosgrove A. Validity of a 1 minute walk test for children with cerebral palsy. Dev Med Child Neurol 2005 Nov;47(11):744-8.

(24) McDowell BC, Humphreys L, Kerr C, Stevenson M. Test-retest reliability of a 1-min walk test in children with bilateral spastic cerebral palsy (BSCP). Gait Posture 2009 Feb;29(2):267-9.

(25) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. Phys Ther 2006 Aug; 86(8):1107-17.

(26) Avery LM, Russell DJ, Raina PS, Walter SD, Rosenbaum PL. Rasch analysis of the Gross Motor Function Measure: validating the assumptions of the Rasch model to create an interval-level measure. Arch Phys Med Rehabil 2003 May;84(5):697-705.

(27) Russell DJ, Leung KM, Rosenbaum PL. Accessibility and perceived clinical utility of the GMFM-66: evaluating therapists' judgements of a computer-based scoring program. Phys Occup Ther Pediatr 2003;23(2):45-58.

(28) van Vulpen LF, De GS, Becher JG, de Wolf GS, Dallmeijer AJ. Feasibility and test-retest reliability of measuring lowerlimb strength in young children with cerebral palsy. Eur J Phys Rehabil Med 2013 Dec;49(6):803-13.

(29) Smits DW, van Groenestijn AC, Ketelaar M, Scholtes VA, Becher JG, Gorter JW. Selective motor control of the lower extremities in children with cerebral palsy: inter-rater reliability of two tests. Dev Neurorehabil 2010;13(4):258-65.

(30) Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Uiterwaal CS, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. Arch Pediatr Adolesc Med 2007 Nov;161(11):1075-81.

(31) Gillett JG, Boyd RN, Carty CP, Barber LA. The impact of strength training on skeletal muscle morphology and architecture in children and adolescents with spastic cerebral palsy: A systematic review. Res Dev Disabil 2016 Sep;56:183-96.

(32) Oeffinger D, Bagley A, Rogers S, Gorton G, Kryscio R, Abel M, et al. Outcome tools used for ambulatory children with cerebral palsy: responsiveness and minimum clinically important differences. Dev Med Child Neurol 2008 Dec;50(12):918-25.

(33) Moreau NG, Holthaus K, Marlow N. Differential adaptations of muscle architecture to high-velocity versus traditional strength training in cerebral palsy. Neurorehabil Neural Repair 2013 May;27(4):325-34.

(34) Peungsuwan P, Parasin P, Siritaratiwat W, Prasertnu J, Yamauchi J. Effects of Combined Exercise Training on Functional Performance in Children With Cerebral Palsy: A Randomized-Controlled Study. Pediatr Phys Ther 2017 Jan;29(1):39-46.

(35) Blundell SW, Shepherd RB, Dean CM, Adams RD, Cahill BM. Functional strength training in cerebral palsy: a pilot study of a group circuit training class for children aged 4-8 years. Clin Rehabil 2003 Feb;17(1):48-57.

(36) Carson RG. Changes in muscle coordination with training. J Appl Physiol 2006 Nov;101(5): 1506-13.

(37) Laursen PB, Jenkins DG. The scientific basis for high-intensity interval training: optimising training programmes and maximising performance in highly trained endurance athletes. Sports Med 2002;32(1):53-73.

(38) MacDougall D, Sale D. Continuous vs. interval training: a review for the athlete and the coach. Can J Appl Sport Sci 1981 Jun;6(2):93-7.

(39) Damiano DL, Arnold AS, Steele KM, Delp SL. Can strength training predictably improve gait kinematics? A pilot study on the effects of hip and knee extensor strengthening on lower-extremity alignment in cerebral palsy. Phys Ther 2010 Feb;90(2):269-79.

(40) Lorentzen J, Kirk H, Fernandez-Lago H, Frisk R, Scharff NN, Jorsal M, et al. Treadmill training with an incline reduces ankle joint stiffness and improves active range of movement during gait in adults with cerebral palsy. Disabil Rehabil 2017 May;39(10):987-93.

(41) Gough M, Shortland AP. Could muscle deformity in children with spastic cerebral palsy be related to an impairment of muscle growth and altered adaptation? Dev Med Child Neurol 2012 Jun;54(6):495-9.

(42) Graham JE, Karmarkar AM, Ottenbacher KJ. Small sample research designs for evidence-based rehabilitation: issues and methods. Arch Phys Med Rehabil 2012 Aug;93(8 Suppl): S111-S116.

Appendix 4.1a Mean degrees (±SD) of the changes during the usual care period, intervention period, and intervention effect in Passive Range of Motion and differences between changes in usual care period, changes in intervention period and changes in follow-up period

	Change in usual care period (Pre2-Pre1)		Change in functional power-training period (Post-Pre2)		Change in follow-up period (Follow-up-Post)		Change intervention-change usual care		Change follow-up-change intervention	
	n	Mean (SD)	n	Mean (SD)	n	Mean (SD)	Mean (95% CI)	p-value	Mean (95% CI)	p-value
Ankle joint with knees extended										
Most affected leg	22	1.2 (3.4)	22	2.3 (4.9)	21	-2.4 (5.3)	1.04 (-2.0–4.1)	.484	-5.0 (-9.3–-0.7)	.026
Least affected leg	22	0 (4.9)	22	3.5 (5.6)	21	-2.9 (7.4)	3.50 (-0.3–7.3)	.071	-6.9 (-12.2–-1.5)	.015
Popliteal angle										
Most affected leg	22	4.1 (9.1)	22	2.3 (11.5)	21	3.1 (13.6)	-1.8 (-10.4–6.8)	.664	0.5 (-3.1–4.1)	.785
Least affected leg	22	3.2 (14.0)	22	-0.7 (12.5)	21	0.7 (7.1)	-3.9 (-14.5–6.8)	.460	1.9 (-6.3–10.1)	.634

All values are in degrees. Pre1 is at the start of the 14 weeks *usual care* period. Pre2 is at the end of the of the *usual care* period which is also the start of the 14 weeks *functional power-training* period. Post training is at the end of the *functional power-training* period which is also the start of the 14 weeks *follow-up* period. Follow-up is at the end of the 14 weeks *follow-up* period. Most and least affected leg was determined by using the modified Trost's SMC test for selective motor control for the ankle dorsal flexion possibility (8).

Appendix 4.1b Pre, Post and Follow-up values (Mean degrees and standard deviation (SD)) for Passive Range of Motion of the ankle and Popliteal angle

	Pre1	Pre2	Post training	Follow-up	Pre2-Pre1		Post-Pre2		Follow-up-Post		Follow-up-Pre2	
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	n	p-value	n	p-value	n	p-value	n	p-value
Ankle joint with knees extended												
Most affected leg	7.05 (6.1)	8.27 (5.4)	10.55 (7.1)	8.05 (4.3)	22	.107	22	.040	21	.050	21	.910
Least affected leg	13.42 (4.7)	13.41 (5.2)	16.91 (7.1)	14.29 (6.0)	22	1.0	22	.008	21	.082	21	.419
Popliteal angle												
Most affected leg	41.14 (14.5)	45.23 (17.9)	47.50 (12.4)	48.33 (14.3)	22	.047	22	.365	21	.785	20	.308
Least affected leg	35.91 (17.9)	39.09 (19.3)	38.41 (16.9)	38.81 (16.0)	22	.296	21	.80	20	.651	20	.820

All values are in degrees. Pre1 is at the start of the 14 weeks *usual care* period. Pre2 is at the end of the of the *usual care* period which is also the start of the 14 weeks *functional power-training* period. Post training is at the end of the *functional power-training* period which is also the start of the 14 weeks *follow-up* period. Follow-up is at the end of the 14 weeks *follow-up* period.
Most and least affected leg was determined by using the modified Trost's SMC test for selective motor control for the ankle dorsal flexion possibility (8).

Improved parent-reported mobility and achievement of individual goals on activity and participation level after functional power-training in young children with cerebral palsy: a double-baseline controlled trial

Liesbeth F. van Vulpen
Sonja de Groot
Eugene A.A. Rameckers
Jules G. Becher
Annet J. Dallmeijer

5

Eur J Phys Rehabil Med 2018 Mar 7 (Epub ahead of print)

Abstract

Background In children with cerebral palsy (CP), strength training programs to improve walking capacity and participation in activities of daily living are commonly used in clinical practice, despite lacking evidence of its effectiveness. It has been suggested that strength training with high movement velocity could be more effective than traditional resistance training to improve functional abilities such as walking. In a recently published study, we have demonstrated the positive effects of functional high-velocity resistance (power) training on muscle strength and walking capacity in young children with CP. Whether this type of training is also effective in achieving individual predefined goals in daily activities and self-reported mobility limitations, has not yet been described however.

Aim To evaluate the effect of functional power-training on parent-reported mobility and achievement of individual goals on activity and participation level in young children with CP.

Design A double-baseline design was used to compare a 14-week period usual care with a 14-week period of functional power-training (3 times a week) and a follow-up period of 14-weeks.

Population Twenty-two children with spastic CP (13 bilateral, GMFCS level I (N=10) and level II (N=12), mean age 7.5 years (SD 1.8, range 4–10 y)) and their parents participated.

Methods Outcome measures were goal attainment scaling (GAS) of individual daily activity related treatment goals, mobility performance as measured using the Functional Mobility Scale (FMS-5m, 50m and 500m), and the parent-reported Mobility Questionnaire (MobQues).

Results After power-training, 86% of children achieved or exceeded their goal, compared with 14% in the usual care period ($p<.001$). The probability of improvement by one point or more on the FMS-500 meter after functional power-training was 10 times higher, compared with the usual care period (Relative Risk=10.0 with 95% CI 1.4–71.3). No changes were found in the FMS-5m and FMS-50m categories. Improvement on the MobQues was significantly greater after power-training compared with usual care (7.9% (95% CI 2.7–13.0, $p=.005$)). The improvement in performance in the activities defined in the treatment goals continued during the follow-up period.

Conclusion and clinical rehabilitation impact The results indicated that functional power-training is an effective training to achieve personalized treatment goals for activities in daily life and parent-reported mobility performance in young children with cerebral palsy.

Trial registration NTR5189.

Introduction

Children with cerebral palsy (CP) experience activity and participation limitations (1). These limitations are often related to impaired walking capacity such as difficulty achieving a certain pace, walking a certain distance or running. Most children with CP whom are able to walk are integrated in community schools and recreational activities. They want to participate in the same activities as their typical, developing peers, such as playing in the schoolyard, participating in gym class and taking part in school outings (2;3). Because of their reduced walking capacity they often experience difficulties keeping up with their peers in these daily activities (1;4;5).

One aspect of decreased walking capacity in children with CP is lower-limb muscle weakness: children with CP only have about 36% to 82% of the muscle strength of typical developing children (6). Several methods have been used to increase muscle strength to enhance the walking capacity of children with CP. The most commonly used method is progressive resistance exercise training (PRE) (7). However, despite an increase in strength in most lower-limb muscles, this training method did not improve walking capacity of children with CP (7-9). Apparently the muscle strength gained does not translate into functional improvement in walking. Moreau et al. (2011) suggested that to get functional improvement, the velocity of the strength training has to be higher, and include functional movements at a higher velocity than generally used in PRE training (10). This suggestion is based on their findings that children with CP have a reduced capacity for rapid force generation (10). This capacity, in particular, is needed in play activities such as playground running and sprinting games and in sports. To improve both muscle strength and the walking capacity, we developed a functional power-training program that consists of resistance training with exercises at high movement velocity (11). This functional power-training program improved lower-limb muscle strength (13–83%) as well as walking capacity (18–128%) in young children with CP (12).

One of the main goals in rehabilitation therapy is to change the individual activity level and the degree of participation in group activities. It is therefore important to know if improved walking capacity (18–128%) after the functional power-training, has led to achieving individual predefined goals in daily activities and to a reduction in mobility limitations. In addition to reaching treatment goals, a reduction in parent-reported mobility limitation is an important outcome, because the limitations experienced reduce participation in leisure activities (13-16).

The purpose of this study was to evaluate the effect of functional power-training on the achievement of individual goals in activities of daily living set by children and/or parents, and parent-reported mobility limitation of their children. With a double-baseline design, we compared changes in individual goals and parent-reported mobility limitation during a 14-week usual care period with changes during a 14-week functional power-training period. After a follow-up period of 14 weeks, we evaluated whether the potential changes in individual goals and parent-reported mobility limitation remained. Our hypothesis was that children and parents would achieve greater improvement in their individual goals with respect to the activities of daily living and mobility performance after a period of power training than after the usual care period.

Materials and methods

Participants

Inclusion criteria were: 1) ambulant children (GMFCS I and II) with predominantly spastic CP, aged 4– 10 years; 2) parents and/or children had a walking-related treatment question, such as being able to walk longer distances or walk at a higher walking velocity; 3) children had to be able to understand and follow simple instructions. Exclusion criteria were: 1) treatment with botulinum toxin A in the lower limb and/or serial casting of the lower limb fewer than 6 months before the start of functional power-training, 2) selective dorsal rhizotomy treatment less than a year before functional power-training, 3) walking is not (yet) the preferred way of mobility.

Design and procedure

This study had a double-baseline design (see Figure 5.1). The participants served as their own controls by comparing the changes in outcome measures after a 14-week period of usual care with the changes after a 14-week training intervention (functional power-training) that followed immediately on the usual care period. Measurements were performed before the usual care period (Pre-1), after the 14-week usual care period, which was also the start of the training period (Pre-2), after the 14-week training period (Post), and follow-up tests were performed 14-weeks after the "Post"-test to assess whether the improvement endured (Follow-up). During both the usual care period and the follow-up period, the frequency of children's physical therapy sessions remained the same, although it differed from child to child.

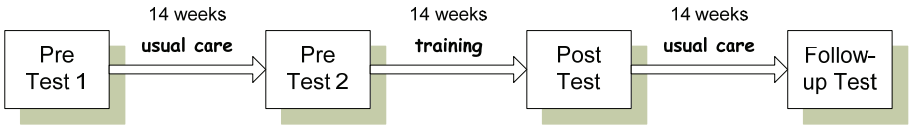

Figure 5.1 Double-baseline research protocol with follow-up measurement.

The primary outcome measure in this study was the evaluation of treatment goals with a focus on activities of daily living, defined by parents and children, and measured using the Goal Attainment Scaling (GAS). Secondary outcome measures were Parent-reported mobility performance as assessed using the Mobility Questionnaire (MobQues) and the Functional Mobility Scale (FMS). GMFCS level and CP type were determined by a pediatric physiotherapist together with a physician.

This study (NL46189.048.13) was approved by the Medical Ethics Committee of Slotervaart medical center and Reade rehabilitation research center in Amsterdam, the Netherlands, and written informed consent was obtained from the parents of each participant.

Setting

Participants were recruited from a rehabilitation centre, two special needs schools for children with physical disabilities, and a university medical centre outpatient clinic. Training and assessments took place in two special schools for physically disabled children and in a rehabilitation centre in the Netherlands.

Intervention

During the intervention period, children engaged in functional power-training for a period of 14 weeks, 3 times a week. Each training session lasted 60 minutes. The training consisted of the following phases: 1. warm-up (10 min); 2. 3–4 different power exercises (35 min); and 3. a final, concluding game (15 min). The training sessions were conducted in small groups (3–6 children) and were supervised by an equal number of therapists. Power exercises and final game activities were chosen in line with the treatment goals of parents and children. A story about super heroes and secret missions was created to keep children motivated and to encourage them to give their best effort during the training sessions. All participants received a T-shirt with their own super hero to foster a group feeling and to increase motivation for training.

Power exercises

We previously presented a detailed description of the power exercises, and how we determined the starting load for each exercise (11). For all participants, 4 to 6 different power exercises were selected; these exercises were relevant for the treatment goals set by the parents and children for improved performance in activities of daily living. In each training session the children performed 3 or 4 of the power exercises. The power exercises were specifically designed to strengthen the plantar flexor muscles while performing loaded functional exercises. Key elements of the power exercises are: a) functional loaded multi-joint exercises such as running and walking with a focus on the ankle push-off; b) high movement velocity (similar to the velocity used in everyday play activities); c) progressive load. The exercises consisted of the following: 1) running, 2) walking, 3) pushing a chair, 4) climbing stairs, 5) propelling a scooter, and 6) walking sideways. Training volume was determined by load, velocity of the movement and number of repetitions. The exercise load was adjusted to a level that allowed children to perform at 70% of their maximum non-loaded velocity. Each exercise was performed at peak effort for 25 seconds, with a resting period of 30–50 seconds, and repeated 6 to 8 times.

Primary outcome measure

Treatment goals reported by parents and/or participants

The improvement in individual goals was measured by Goal Attainment Scaling (GAS), an individualized measurement to evaluate children's progress towards goals on the level of activity and participation (17). GAS is a sensitive, evaluative measure which reliably describes the change in individuals after treatment (ICC=0.86) (18). It is a 6-point scale measurement, where a score of -2 represents a level that is unchanged from the baseline, a score of -1 represents less progress than expected, 0 stands for the level of improvement that is expected, +1 and +2 are scores for respectively more and much more improvement than expected, and a score of -3 represents a deterioration. We adhered to the following criteria for scale development: a) goals were set by experienced pediatric physical therapists in consultation with parents and children, based on their expressed main therapy objective regarding level of activity and participation domains of the ICF-CY; b) the six GAS scale levels were Specific, Measurable, Achievable, Realistic/Relevant and Time-related (SMART); c) the GAS scale is an ordinal scale with incremental, equal interval steps (18). If parents and/or participant had more than one treatment goal, they had to specify the importance of each goal. The treatment goal with the highest priority was included in the analysis. After every 14-week period, the treatment goals were evaluated with parents and children and new goals were set.

Secondary outcome measures

Parent-reported mobility performance

Mobility performance was measured using the Functional Mobility Scale (FMS) and the Mobility Questionnaire (MobQues), both completed by parents. The FMS measures functional mobility in children with CP aged 4 to 18 years (19). It measures functional mobility over three distinct distances, chosen to represent mobility in the home (5 meter), at school (50 meter) and in the wider community (500 meter). Parents were asked to rate the child's usual walking performance in these three distances with an eye towards a possible need for assistive devices. On this 6-point scale, a rating of 6 means that the child walks independently on all surfaces, and a 5 means the ability to walk unsupported on level surfaces. Children using walking sticks are rated 4, children using crutches are rated 3, those using a walker or frame for support are rated 2, and children using a wheelchair, stroller or buggy having a rating of 1. Test-retest reliability of the FMS is good (Kappa=0.86–0.92) (19) as are construct, content and concurrent validity (20).

The Mobility Questionnaire (MobQues) is a Dutch questionnaire for parents with children aged 2–13 years to determine the degree of difficulty the child experiences with mobility tasks. The questionnaire scores 28 mobility tasks in everyday life and includes indoor activities, such as "how difficult is it for your child to go upstairs?" as well as outdoor activities, such as "how difficult is it for your child to run on grass?". The response options, given on a 5-point scale, are: not difficult at all (score 4), slightly difficult (score 3), somewhat difficult (score 2), very difficult (score 1), impossible without help (score 0). A total score for these 28 items is computed by addition of the individual scores. This score is divided by the highest possible score and multiplied by 100 to obtain a score on a scale of 0 to 100; with a low score represents severe limitations in mobility. The test has good intra-rater reliability (ICC=0.96) (21) and content and construct validity (22).

Training compliance was tracked by means of a personal training log for each participant.

Statistical analysis

A power calculation was performed and described previously (11). The power calculation revealed that, based on the main outcome measure sprint power, a sample size of at least 19 children was needed. Twenty-two children were recruited to allow for a dropout rate of 10% (11). Frequencies of the goal attainment scores were calculated at the end of the usual care period, the functional power-training period and the follow-up period. Wilcoxon signed rank tests were used to determine the possible effects of intervention

by comparing the goal attainment scores achieved during the functional power-training period with goal attainment scores achieved during the usual care period.

Frequencies of FMS-score changes were calculated for every period. We dichotomized the FMS-score by grouping children whose FMS-score improved by one or more points, and grouping children whose FMS-score did not improve or deteriorated. The dichotomous FMS-scores were used to calculate participants' Relative Risk (RR) of improving by one or more points on the FMS, with a 2x2 table (RR= a/(a+b)) / c/(c+d)) (23).

For the MobQues scores mean and standard deviations were calculated at each point of measurement (*Pre-1*, *Pre-2*, *Post* and *Follow-up*). Changes in MobQues scores during the usual care period (Δ *Pre-2* - *Pre-1*), functional power-training period (Δ *Post* - *Pre-2*) and follow-up period (Δ *Follow-up* - *Post*) were calculated. Paired sampled t-tests (if normally distributed) and Wilcoxon signed rank tests (if not normally distributed) were used to determine the possible effects of intervention by comparing changes during the functional power-training period with changes during the usual care and changes during follow-up. All statistical analyses were performed using SPSS Statistics 22.0 software (IBM Corporation, New York, USA) with an intention-to-treat analysis.

Results

Characteristics of the study population

Personal characteristics of the participants are presented in Table 5.1. Twenty-two children participated in this study, three of which had interfering treatment (serial casting for 1 to 3 weeks) during their usual care period, and one participant had serial casting (3 weeks) during the follow-up period. These four participants were included in the intention-to-treat analysis. One participant was hospitalized during the follow-up period because of a medical problem that was unrelated to the training and this participant therefore missed all the follow-up tests.

Training compliance

The children participated in 88% (range: 57–100%) of the scheduled training sessions. One child missed 33% of all possible training sessions because of logistical problems of the parents. Another child missed 43% of the training sessions because the family moved away during the training period. Other reasons for not attending the training sessions were illness, holidays, medical appointments, family visits and unspecified reasons.

Table 5.1 Characteristics of the participants

Boys (N)	11
Girls (N)	11
Age (years), mean (SD)	7.5 (1.8)
Range (years)	4.0–10.2
Body mass (kg), mean (SD)	26.0 (8.2)
Range (kg)	14.0–45.5
Height (m), mean (SD)	1.24 (0.11)
Range (m)	0.99–1.45
GMFCS[1] I (N)	10
GMFCS II (N)	12
Unilateral spastic CP (N)	9
Bilateral spastic CP (N)	13
Regular school (N)	13
Special needs education (N)	9
Intervention period in springtime (N)	11
Intervention period in autumn (N)	11
AFO[2] in daily live (N)	13
No AFO in daily live (N)	9

[1] GMFCS, Gross Motor Function Classification System.
[2] AFO, ankle foot orthosis on one limb or both. Supramalleolar orthosis and orthopedics shoes or registered as no AFO in daily life.

Goal Attainment Scaling

Examples of treatment goals, formulated by parents and children were: "I would like my child to be able to join in more when class mates are playing soccer in the schoolyard" and "I would like my child to climb stairs faster so she can keep up with her class mates at school". We were able to distinguish three categories of treatment goals set by parents and children; 1) to improve running (31%): to run faster, more easily, without falling, with better agility to keep up with peers in the schoolyard and at play; 2) to improve walking (45%): to walk faster, longer distances, without support, without falling, at a higher pace, to keep up with peers in the schoolyard, at school and on family outings; 3) to improve gross motor activities (24%): to be able to stand up in an open space, to be able to jump rope, to climb stairs more easily and faster.

Figure 5.2 shows the outcome of the GAS by number of participants for each period. During the usual care period, 68% of the children showed no change (score -2), 18% showed an improvement but not enough to achieve their goal (score -1) and 14% of children achieved their goal (score 0). After power-training, 0% of children had no change, 14% improved but not to the level of achieving the goal (score -1) and 86% achieved or

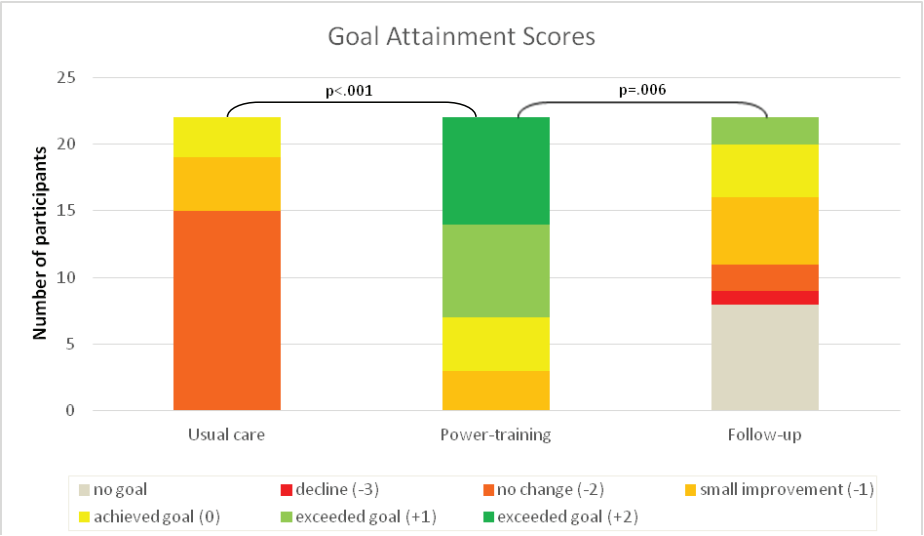

Figure 5.2 Number of participants on each category of Goal Attainment Score after the 14wk usual care period, 14wk power-training period and 14wk follow-up period.

exceeded their goal (score 0, +1, +2). During the follow-up period, eight parents and children were satisfied with how things were going and could not formulate a (new) goal for the follow-up period. Furthermore, two children (14%) showed no change in their goal (score -2), one child showed a deterioration (score -1), 36% showed improvement but not enough to achieve their goal (score -1) and 43% of children achieved their goal (score 0, +1, or +2) (see Figure 5.2). The degree to which goals were achieved during the usual care period (median score -2 (range -2 to 0)) and during the functional power-training period (median score +1 (range -1 to +2)) differed significantly (p<.001). The degree to which goals were achieved during the functional power-training period and during the follow-up (median score -1 (range -3 to 1)) also differed significantly (p=.006).

Functional Mobility Scale

Statistical analysis of the FMS was performed on the data obtained from 21 children. One Pre-2 value is missing because of unknown reasons and one follow-up value is missing because the participant had to be hospitalized at the time of the follow-up measurements. None of the children walked with crutches or sticks, so a score of 3 or 4 was never reported by the parents. Functional power-training had no effect on achievements over a 5-meter distance. After functional power-training, two participants changed from a score 1 (not possible without support from an adult or only possible in a wheelchair) to a score of 5

(walking independently on a level surface without walking aids) over the 50-meter distance, and they maintained their improvements in the follow-up period. The FMS 500m showed that, at the start of the functional power-training period, 52% (n=11) of children used a walking frame, wheelchair or were supported by their parents (score 1 and 2) whereas at the end of the functional power-training period, only 13.6% (n=3) of the children depended on a walking aid, wheelchair or support by their parents (Figure 5.3). The Relative Risk (RR) value was 10 (with a 95% CI of 1.4–71.3) indicating that the probability for improvement by one point or more on the FMS 500m after functional power-training was 10 times higher than following the usual care period. Children had a 4.8% chance of improving, by one point or more, on the FMS 500m after the usual care period, whereas they had a 47.6% chance of improving on the FMS 500m after the functional power-training period.

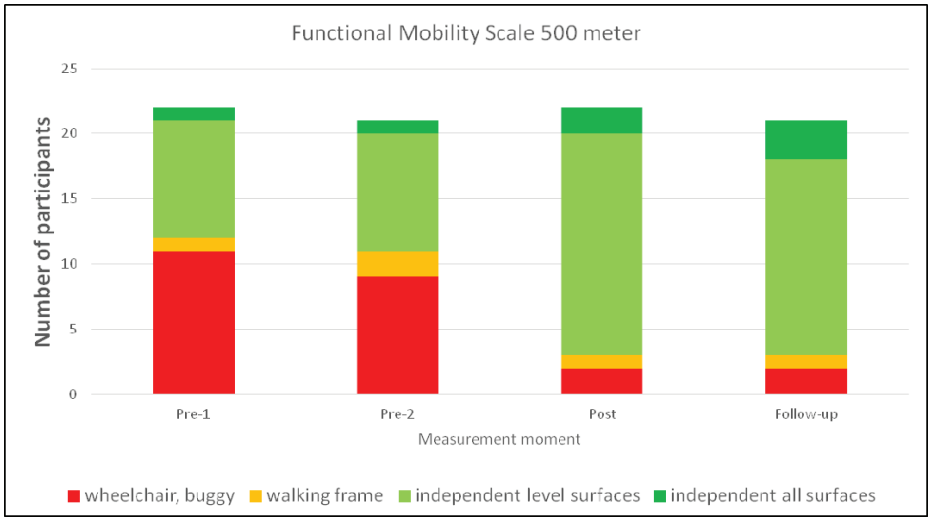

Figure 5.3 Number of participants in each category on Functional Mobility Scale at measurement moment Pre-1 (start of the 14wk usual care period), Pre-2 (end of usual care period, also start of the training period), Post (after the 14wk power-training period) and Follow-up (after 14wk follow-up period).

Mobility Questionnaire

Seventeen questionnaires were included in the statistical analyses of the MobQues. Four parents did not fill out the MobQues at any measurement moment because they lacked the necessary language skills (two cases) or they were insufficiently motivated to return the questionnaire (two cases). One parent forgot to fill out the questionnaire at Pre-1 and one parent did not return the questionnaire at Post measurement, both for unknown reasons.

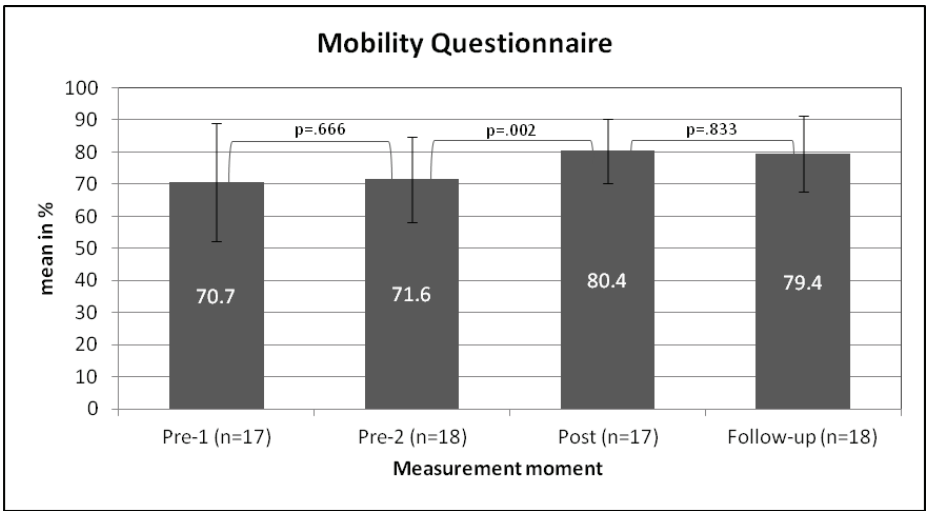

Figure 5.4 Mean scores with SD of the Mobility Questionnaire at each measurement moment; Pre-1 (start of the 14wk usual care period), Pre-2 (end of usual care period, also start of the training period), Post (after the 14wk power-training period) and Follow-up (after 14wk follow-up period).

Figure 5.4 shows the mean values at each time of measurement. Changes in the MobQues during the functional power-training period differed significantly from the changes during the usual care period (7.9% (95% CI 2.7–13.0, p=.005). Parents reported that mobility was less difficult after functional power-training. For instance, walking and running outdoors became easier and walking up and down stairs became less strenuous.

Discussion

The current study shows that the improvement in mobility and participation in activities of daily living that children and parents experienced – measured by the achievements at individualized goals and self-reported mobility – was significantly more pronounced following functional power-training compared to changes during the usual care period. These results are consistent with the increase in walking capacity and leg muscle strength after functional power-training, as reported in a previous study (12).

GAS offers the opportunity to measure achievements in individual goals set by parents and children, which is one of the most important outcomes of rehabilitation treatment (16;24). After functional power-training, 86% of children achieved or exceeded their goals, whereas after a usual care period only 14% of children achieved their goals. In our study, of the 86% of children who achieved their goal 18% improved to a level that

was expected (score 0) while 68% improved to a level that exceeded expectations (score +1, +2). This indicates that the interventions have led to an improvement greater than expected by parents, therapists and/or schoolteachers, all of whom were involved in setting goals and scoring.

FMS-500m scores indicate that mobility performance had improved in the wider community. No improvements were found in mobility performance in the home (FMS-5m) and at school (FMS-50m) setting, a finding that may be explained by the expected ceiling effect. Almost all children scored at level 5 and 6 on the FMS-5m and FMS-50m, which corresponds with our inclusion criteria (GMFCS level I and II). Compared with the changes during the usual care period (Relative Risk=10), the probability of one or more points improvement in FMS 500m score following functional power-training was 10 times higher, indicating that these children were able to walk larger distances more easily. The FMS is often used to evaluate gait improvement after surgery (25-29). Our study found much more improvement than what has been reported in these previous surgery trials. Harvey et al. showed a deterioration in the FMS 500m score during the first 3 to 6 months post-surgery (OR=0.24 at 3mo and OR=0.5 at 6mo) and improvements one and two years post-surgery (OR=1.47 at 12mo and OR=2.23 at 24mo) (26). The marked improvement in mobility performance (500m) in our study is in line with earlier findings that functional power-training is very effective in increasing walking capacity (such as walking and running speed) (12), so improvements in parent-reported mobility were expected. Previous strength training studies have not found any improvement in mobility capacity or performance, however (7-9). Important differences between these studies and those that looked at functional power-training are that the latter involve strength training with high velocity exercises that also are functional movements (12). Secondly, 45% of parents stated that increased walking distance and speed would help their child participate more in activities of daily living, which was associated with their treatment goals. The fact that parents were focused on the importance of walking greater distances at a higher speed may have been helpful in achieving the goal of improved walking performance. After functional power-training, parents also reported that children were better able to participate in trips into town, shopping, visiting friends or walking without needing a walker or wheelchair.

The decrease in mobility limitation in everyday situations experienced by children, as measured using the MobQues, was mirrored by, and consistent with, the improvement in mobility performance (FMS), walking capacity and gross motor function following power training reported earlier (12). Mobility is an important aspect of rehabilitation

therapy because it is a strong determinant of participation in activities of daily living (30). Children with physical disabilities are less involved in leisure activities than their peers; their activities are more passive, home-based, and they lack variety (5). Also when children and parents experience mobility limitations, their participation in leisure activities declines (13;14;30). A previous strength training study that looked at the effects of 14 weeks of lower-limb strength training, did not find that mobility limitations as measured by MobQues was reduced, despite reporting an increase in muscle strength (9). A likely explanation for this difference in effects is that we used high-velocity training and integrated strength training in functional exercises that were related to the treatment goals. This indicates that functional power-training is more effective in increasing muscle strength and reducing mobility limitations than progressive resistance exercise.

Limitations of the study

In the present study we did not evaluate the children's self-confidence. Parents did report several positive effects of functional power training on increased self-confidence in their children, however. This was manifested by prolonged playing in the playground after school hours, not being afraid to join in soccer with peers at school, and by accepting new physical challenges that parents thought children would not have attempted before engaging in power training. These effects need to be further investigated in future research.

To increase our study's feasibility in a heterogeneous group of children with CP, we used a double-baseline design rather than a randomized-controlled trial (RCT) (12). In this design the children served as their own controls by comparing changes that occurred during an usual care period with changes during a period of functional power training. A double-baseline design could be argued to be a limitation of the study since randomized controlled trials are generally considered to generate the highest level of evidence. However, a double-baseline design has been described as a robust alternative method for evaluating evidence-based rehabilitation for heterogeneous patient populations (31). In addition, the number of children in this approach is smaller than what is needed for an RCT because an adequate statistical power can be reached with smaller subject groups. This makes the double-baseline design more feasible and less expensive (31).

Conclusion

This study shows that children and parents experience a significant improvement in mobility and in the ability to participate in activities of daily living following functional

power-training, as demonstrated by children's performance in individualized goals and by self-reported mobility.

Clinical messages

- Functional power-training is a strength training method with high movement velocity in progressively loaded exercises, integrated in functional movements aimed at improved performance in the activities of daily living in children with CP.
- Parents noticed significant improvements in mobility and children improved on personalized treatment goals relating to activities of daily living.

Acknowledgement

The authors wish to thank all children, parents and trainers who participated in this study. This study has been funded by Mitialto foundation, the Duyvensz-Nagel foundation, the Dutch Rehabilitation fund and Royal Dutch Society for Physical Therapy.

References

(1) Bax M, Goldstein M, Rosenbaum P et al. Proposed definition and classification of cerebral palsy, April 2005. Dev Med Child Neurol 2005;47:571-6.

(2) Beckung E, Hagberg G, Uldall P, Cans C. Probability of walking in children with cerebral palsy in Europe. Pediatrics 2008;121:187-92.

(3) Gorter JW, Rosenbaum PL, Hanna SE et al. Limb distribution, motor impairment, and functional classification of cerebral palsy. Dev Med Child Neurol 2004;46:461-7.

(4) Fauconnier J, Dickinson HO, Beckung E et al. Participation in life situations of 8-12 year old children with cerebral palsy: cross sectional European study. BMJ 2009;338:1458.

(5) Shikako-Thomas K, Majnemer A, Law M, Lach L. Determinants of participation in leisure activities in children and youth with cerebral palsy: systematic review. Phys Occup Ther Pediatr 2008;28:155-69.

(6) Dallmeijer AJ, Rameckers EA, Houdijk H, De GS, Scholtes VA, Becher JG. Isometric muscle strength and mobility capacity in children with cerebral palsy. Disabil Rehabil 2015;1-8.

(7) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014;35:239-49.

(8) Franki I, Desloovere K, De CJ et al. The evidence-base for conceptual approaches and additional therapies targeting lower limb function in children with cerebral palsy: a systematic review using the ICF as a framework. J Rehabil Med 2012;44:396-405.

(9) Scholtes VA, Becher JG, Janssen-Potten YJ, Dekkers H, Smallenbroek L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. Res Dev Disabil 2012;33:181-8.

(10) Moreau NG, Falvo MJ, Damiano DL. Rapid force generation is impaired in cerebral palsy and is related to decreased muscle size and functional mobility. Gait Posture 2012;35:154-8.

(11) van Vulpen LF, de Groot S, Rameckers EA, Becher J, Dallmeijer AJ. Effectiveness of functional power training on walking ability in young children with cerebral palsy: study protocol of a double-baseline trial. Pediatr Phys Ther 2017;29:275-82.

(12) van Vulpen LF, de Groot S, Rameckers EA, Becher J, Dallmeijer AJ. Improved walking capacity and muscle strength after functional power-training in young children with cerebral palsy. Neurorehabil Neural Repair 2017;31:827-41.

(13) Bjornson KF, Zhou C, Stevenson R, Christakis DA. Capacity to Participation in Cerebral Palsy: Evidence of an Indirect Path Via Performance. Arch Phys Med Rehabil 2013;94:2365-72.

(14) Bult MK, Verschuren O, Jongmans MJ, Lindeman E, Ketelaar M. What influences participation in leisure activities of children and youth with physical disabilities? A systematic review. Res Dev Disabil 2011;32:1521-9.

(15) Chiarello LA, Palisano RJ, Maggs JM et al. Family priorities for activity and participation of children and youth with cerebral palsy. Phys Ther 2010;90:1254-64.

(16) Graham HK, Rosenbaum P, Paneth N et al. Cerebral palsy. Nat Rev Dis Primers 2016;2:15082.

(17) Steenbeek D, Ketelaar M, Lindeman E, Galama K, Gorter JW. Interrater reliability of goal attainment scaling in rehabilitation of children with cerebral palsy. Arch Phys Med Rehabil 2010;91:429-35.

(18) Steenbeek D, Gorter JW, Ketelaar M, Galama K, Lindeman E. Responsiveness of Goal Attainment Scaling in comparison to two standardized measures in outcome evaluation of children with cerebral palsy. Clin Rehabil 2011;25:1128-39.

(19) Harvey AR, Morris ME, Graham HK, Wolfe R, Baker R. Reliability of the functional mobility scale for children with cerebral palsy. Phys Occup Ther Pediatr 2010;30:139-49.

(20) Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The Functional Mobility Scale (FMS). J Pediatr Orthop 2004;24:514-20.

(21) van Ravesteyn NT, Dallmeijer AJ, Scholtes VA, Roorda LD, Becher JG. Measuring mobility limitations in children with cerebral palsy: interrater and intrarater reliability of a mobility questionnaire (MobQues). Dev Med Child Neurol 2010;52:194-9.

(22) van Ravesteyn NT, Scholtes VA, Becher JG, Roorda LD, Verschuren O, Dallmeijer AJ. Measuring mobility limitations in children with cerebral palsy: content and construct validity of a mobility questionnaire (MobQues). Dev Med Child Neurol 2010;52:229-35.

(23) Guyatt GH, Rennie D. User's guide to the medical literature. A manual for evidence-based clinical practice. Chicago: AMA Press, 2002.

(24) Chiarello LA, Bartlett DJ, Palisano RJ et al. Determinants of participation in family and recreational activities of young children with cerebral palsy. Disabil Rehabil 2016;38:2455-68.

(25) Blumetti FC, Wu JC, Bau KV et al. Orthopedic surgery and mobility goals for children with cerebral palsy GMFCS level IV: what are we setting out to achieve? J Child Orthop 2012;6:485-90.

(26) Harvey A, Graham HK, Morris ME, Baker R, Wolfe R. The Functional Mobility Scale: ability to detect change following single event multilevel surgery. Dev Med Child Neurol 2007;49:603-7.

(27) Terjesen T, Lofterod B, Skaaret I. Gait improvement surgery in ambulatory children with diplegic cerebral palsy. Acta Orthop 2015;86:511-7.

(28) Thomason P, Selber P, Graham HK. Single Event Multilevel Surgery in children with bilateral spastic cerebral palsy: a 5 year prospective cohort study. Gait Posture 2013;37:23-8.

(29) Yu S, Rethlefsen SA, Wren TA, Kay RM. Long-term ambulatory change after lower extremity orthopaedic surgery in children with cerebral palsy: a retrospective review. J Pediatr Orthop 2015;35:285-9.

(30) King G, McDougall J, Dewit D, Petrenchik T, Hurley P, Law M. Predictors of Change Over Time in the Activity Participation of Children and Youth with Physical Disabilities. Child Health Care 2009;38:321-51.

(31) Graham JE, Karmarkar AM, Ottenbacher KJ. Small sample research designs for evidence-based rehabilitation: issues and methods. Arch Phys Med Rehabil 2012;93:111-6.

Improvements in muscle strength are associated with improvements in walking capacity in young children with cerebral palsy

Liesbeth F. van Vulpen
Sonja de Groot
Eugene A.A. Rameckers
Jules G. Becher
Annet J. Dallmeijer

6

Submitted

Abstract

Aim To study the relationships between changes in lower-limb muscle strength and changes in walking capacity during 14-week periods of usual-care, power-training and follow-up in children with spastic cerebral palsy (CP).

Method 22 Children with spastic CP participated (13 bilateral, GMFCS level I (N=10) and II (N=12), 7.5±1.8 years). Generalized estimating equations were used to evaluate the multivariate relationships between within-subject changes in muscle strength (isometric strength of ankle plantar flexors gastrocnemius (GASTR) and soleus (SOL), Knee Extensors (KE) and Hip Abductors (HA)) and walking capacity (muscle power sprint test (MPST), 1-minute walk test (1MWT), 10-m shuttle run test (SRT)).

Results Changes in HA were associated with changes in MPST (R2=0.66), changes in GASTR en HA (less affected leg) were associated to changes in SRT (R2=0.43), and changes in GASTR (affected leg) were associated to changes in 1MWT (R2=0.21). All associations showed better walking capacity with increased strength.

Interpretation Improvements in HA and GASTR muscle strength were associated to better walking capacity in young children with CP. This suggests that walking capacity, especially sprint capacity, can be improved by increasing strength of these muscles by functional power-training in this population.

Trial registration NTR5189.

Introduction

Children with cerebral palsy (CP) have impaired walking capacity because of their motor impairments (1). These motor impairments include spasticity, loss of selective motor control and muscle weakness (1). As a consequence these children experience problems in keeping up with their typical developing peers during activities, such as playing in the schoolyard, participating in gym class and taking part in school outings (2). Optimizing walking capacity is, therefore, an important aim of treatment at a young age.

Apart from spasticity and reduced selective motor control, muscle weakness is often reported as an important factor affecting walking capacity (1). Walking children with CP only have about 36% to 82% of the muscle strength of typically developing children (3;4). Previous cross-sectional studies have reported a moderate to strong relationship between leg muscle strength and walking capacity in children and young adults with CP, showing better walking capacity in those with stronger muscles (3;5-7). Therefore, studies focused on muscle strengthening with the aim to increase walking capacity in these children. Unfortunately, systematic reviews showed that there is inconclusive evidence that the walking capacity can be improved by increasing muscle strength through progressive resistance exercises (8;9). Some studies showed increases in muscle strength of some of the targeted leg muscles after progressive resistance training programs but this was not accompanied by increases in walking capacity (10;11). Lack of specificity of the strength training programs is a possible explanation for the absence of functional effects (12;13). Most studies used training equipment, like a leg press or an isokinetic dynamometer, to improve strength instead of using functional movements like walking and running. Additionally, the lower movement velocity of the traditional resistance training did not comply with movement velocities that are usually seen in daily activities of young children. In addition, rapid force development seems to be compromised in children with CP (14). Therefore, a different training approach with higher movement velocity in progressive loaded strength training exercises was developed, and incorporated in functional movements like walking and sprinting (15).

Recently, we published the results of this specific training program (called functional power-training) on muscle strength and walking capacity in young children with CP (13). The results showed that power-training was highly effective for increasing muscle strength and walking capacity in young children with CP (13). In this functional power-training study the functional strength exercises, like running and stair climbing, were conducted by children at maximum effort on 70% of their maximum speed, while the exercise was progressively loaded to ensure the principles of strength training. Children

trained 14 weeks, 3 times a week for 60 minutes and showed significant increases of 18–128% in muscle strength of all targeted muscles (ankle plantar flexors, hip abductors, knee extensors) and in sprint capacity (83%), walking speed (13%) and running endurance (56%) during this training compared with a preceding 14-week usual-care period in the same patient group (13). It is, however, not yet clear to what extent improvements in walking capacity do relate to improvements in muscle strength, and which muscle groups account for these changes in walking capacity.

Therefore, the aim of this study was to evaluate whether changes in muscle strength could explain the changes in walking capacity, defined as sprint capacity, walking speed and running endurance. We performed a secondary analyses of the data of the before-mentioned power-training trial (13), assessing associations between changes in muscle strength and changes in walking capacity during 14-weeks usual-care period, 14-weeks functional power-training period and 14-weeks follow-up (usual-care).

Method

Participants

Participants were walking children aged 4–10 years, with a spastic CP, and Gross Motor Function Classification System (GMFCS) I and II. Children had to be able to understand and follow simple instructions. Exclusion criteria were treatment with botulinum toxin A in the lower limb and/or serial casting of the lower limb less than 6 months before the start of the functional power-training and selective dorsal rhizotomy treatment less than a year before the functional power-training.

Design and procedure

This study consists of secondary analyses of data that were collected in a study on the effects of functional power-training on walking capacity and leg strength in young children with CP (13). In short, the study had a double-baseline design, with three different time periods; 1) a 14-week usual-care period, 2) a 14-week functional power-training period and 3) a 14-week follow-up period with usual-care (15).

Measurements were performed before and after each period. During the power-training period children received loaded functional exercises like running and walking with a progressive load, 3 times a week for 60 minutes. Exercises were performed at 50–70% of their maximum unloaded speed by applying resistance to the movement with an

external load. Each exercise was performed for 25s on maximal effort, with a resting period of 30–50s, and six to eight repetitions for each exercise (15). Children followed their regular physical therapy sessions during usual-care period and follow-up period. Detailed descriptions of the power-training study can be found in Van Vulpen et al. (2017) (15).

For descriptive purposes, GMFCS and type of CP were determined by a pediatric physiotherapist together with a physiatrist. To determine more and less affected leg of each child, selective motor control of knee extension and ankle dorsiflexion were tested by the modified Trost Selective Motor Control test (16).

Medical Ethics Committee of the Slotervaart medical center and Reade rehabilitation research center in Amsterdam, the Netherlands, approved this study, and written informed consent was obtained from the parents of each participant.

Setting

Participants were recruited from a rehabilitation center, two special needs schools for children with physical disabilities and an outpatient clinic of a university medical center.

Measurements

Walking capacity

Walking capacity was defined as sprint capacity, endurance and walking speed. Sprint capacity of the child was expressed in mean power and peak power, measured with the 6x15m Muscle Power Sprint Test (MPST) (15;17). The power output for each of the six sprint runs was estimated using sprinting velocity and body mass (17). Mean power was defined as the average power output of the six runs. Running endurance, expressed in exercise time by half a minute accuracy (stage), was measured with the 10m Shuttle Run Test (SRT), modified for children with CP GMFCS level I and II (18). Walking speed was measured with the distance covered in the 1-minute walk test (1MWT), with instructions to walk in one minute as fast as possible (19).

Muscle strength

Isometric muscle strength in both legs of plantar flexors with knees extended (m. gastrocnemius (GASTR)) and with knees flexed (m. soleus (SOL)), knee extensors (KE) and hip abductors (HA) was measured with a hand-held dynamometer (microFET, Biometrics BV, Almere, The Netherlands) using the make-method (20). A standardized

protocol was used for positioning of the child, joint fixation and joint positioning (20). Torque (Nm) was calculated by multiplying force (Newton) by the length (meter) of the lever arm. Each strength test was performed 3 times and maximum force for each repetition was registered. To improve reliability, strength tests were measured at two different test days. The mean of these six measurements (two test days with three repetitions on each day) per muscle group was used in the analysis (20).

Dynamic muscle strength of plantar flexors of the more and less affected leg was measured with a standing heel-rise test on one leg following a standardized protocol (20). Maximum number of repetitions for each leg was noted.

Statistical analysis

Changes in muscle strength and walking capacity during the usual-care period, functional power-training period and follow-up period were calculated and expressed as a percentage of the tests outcome at the start of each period, except for the dynamic muscle strength (heel-rise test) which was expressed as change in number of repetitions possible. Generalized Estimating Equation (GEE) analysis was used to evaluate the relationship between the changes in muscle strength (independent variable) and changes in walking capacity (dependent variable). Muscle strength for each leg, more affected and less affected, was included in the models. GEE takes the dependency of related samples (repeated measurements due to the three time periods, and more and less affected leg) into account. First, univariate modeling was applied. Secondly, interaction effects between muscle strength and age (4–6 years old or 7–10 years old) or for diagnosis (unilateral CP or bilateral CP) were assessed. Thirdly, multivariate modeling was used with backward selection procedure with the isometric muscle strength variables. Variables were removed from the model one by one, starting with the least significant independent variable, until only variables remained that met the criteria $p<.05$. All statistical analyses were performed using SPSS Statistics 22.0 software (IBM Corporation, New York, USA).

Results

Characteristics of the study population

Personal characteristics of the 22 participants in this study are presented in Table 6.1. One participant was hospitalized during the follow-up measurement because of a medical problem that was unrelated to the training. There were two missing values for the 1MWT: one participant could not be tempted to walk instead of run during the post-test, and one

Table 6.1 Characteristics of the participants

Boys (N)	11
Girls (N)	11
Age (years), mean (SD)	7.5 (1.8)
Range (years)	4.0–10.2
Body mass (kg), mean (SD)	26.0 (8.2)
Range (kg)	14.0–45.5
Height (m), mean (SD)	1.24 (0.11)
Range (m)	0.99–1.45
GMFCS[1] I (N)	10
GMFCS II (N)	12
Unilateral spastic CP[2] (N)	9
Bilateral spastic CP (N)	13
Regular school (N)	13
Special needs education (N)	9
AFO[3] in daily live (N)	13
No AFO in daily live (N)	9

[1] GMFCS, Gross Motor Function Classification System.
[2] SCPE, Surveillance of Cerebral Palsy in Europe, classification of subtypes CP (2).
[3] AFO, ankle foot orthosis on one limb or both. Supramalleolar orthosis and orthopedics shoes or registered as no AFO in daily life.

participant changed footwear. One participant could not perform the isometric GASTR and SOL tests of the more affected limb because of motor control problems.

Table 6.2 shows the results for univariate regression analyses with relationships between changes in walking capacity and changes in muscle strength. All muscle groups showed significant associations with MPST (R^2 ranging from 9% to 62%), with SRT (R^2 ranging from 11% to 44%) and with 1MWT (R^2 ranging from 5% to 21%). The relation between changes in walking capacity and changes in GASTR of the more affected leg and the less affected leg are visualized in Figure 6.1.

Interaction effect for age was found for the association between GASTR in the less affected leg and MPST, showing a stronger association in older than younger children (Table 6.2). No other interaction effects for age or for type of diagnosis (unilateral or bilateral CP) were found for the association between muscle strength and MPST. Four interaction effects were found for age regarding the association between muscle strength and SRT and 1MWT. Interaction effects for type of diagnose were found in eight associations with SRT and 1MWT with different muscle strength groups (Table 6.2), showing that associations were stronger for the bilateral CP children than the unilateral CP children (Table 6.2).

Table 6.2 Results for univariate regression analysis with changes in walking capacity (Muscle Power Sprint Test (MPST), Shuttle Run Test (SRT), 1-Minute Walk Test (1MWT)) as dependent variable and changes in muscle strength (isometric and dynamic strength) as independent variables

	More affected leg			Less affected leg		
	β (SE)	p-value	R^2	β (SE)	p-value	R^2
Dependent variable: Sprint Power MPST (Δ%)						
Constant Gastrocnemius (Δ%)	6.36 (6.85) 1.85 (0.44)	<.001	0.35	18.60 (7.16) 1.38 (0.42)[a]	.001	0.13
Constant Soleus (Δ%)	9.09 (5.06) 1.08 (0.27)	<.001	0.25	18.96 (8.44) 0.85 (0.27)	.002	0.09
Constant Hip abductors (Δ%)	9.28 (4.61) 1.16 (0.21)	<.001	0.43	13.14 (2.95) 1.48 (0.19)	<.001	0.62
Constant Knee extensor (Δ%)	15.78 (4.67) 1.98 (0.28)	<.001	0.41	15.36 (5.48) 2.20 (0.44)	<.001	0.32
Constant Dynamic plantar flexor (Δ)	25.54 (5.82) 5.01 (0.72)	<.001	0.22	28.93 (5.54) 4.35 (1.15)	<.001	0.21
Dependent variable: Endurance SRT (Δ%)						
Constant Gastrocnemius (Δ%)	0.37 (2.75) 0.95 (0.23)	<.0001	0.25	0.93 (3.96) 1.15 (0.37)[c]	.002	0.26
Constant Soleus (Δ%)	1.36 (4.25) 0.57 (0.20)[a,c]	.001	0.19	2.23 (4.45) 0.66 (0.21)[a]	.001	0.17
Constant Hip abductors (Δ%)	6.82 (4.00) 0.39 (0.21)	.059	0.11	5.25 (2.57) 0.68 (0.14)	<.001	0.33

	More affected leg			Less affected leg		
	β (SE)	p-value	R^2	β (SE)	p-value	R^2
Constant	7.66 (3.28)			7.08 (3.36)		
Knee extensor (Δ%)	0.79 (0.16)	<.001	0.16	0.92 (0.23)	<.001	0.14
Constant	8.36 (3.03)			9.62 (3.13)		
Dynamic plantar flexor (Δ)	3.42 (0.60)	<.001	0.27	3.57 (0.56)[b]	<.001	0.44
Dependent variable: Walking Speed 1MWT (Δ%)						
Constant	0.91 (1.56)			2.91 (1.50)		
Gastrocnemius (Δ%)	0.28 (0.08)	<.001	0.21	0.19 (0.07)	.004	0.06
Constant	2.65 (1.76)			1.82 (1.66)		
Soleus (Δ%)	0.11 (0.04)	.011	0.06	0.18 (0.08)[b]	.032	0.12
Constant	3.21 (1.73)			3.01 (1.29)		
Hip abductors (Δ%)	0.09 (0.03)	.001	0.07	0.15 (0.04)	<.001	0.16
Constant	3.74 (1.53)			3.01 (1.32)		
Knee extensor (Δ%)	0.15 (0.05)	.001	0.05	0.23 (0.06)[c]	<.001	0.09
Constant	4.30 (1.44)			4.29 (1.48)		
Dynamic plantar flexor (Δ)	0.58 (0.20)	.004	0.07	0.70 (0.19)[c]	<.001	0.15

[a] Interaction effect for age showing a stronger association in older children (7–10 years old) than the younger children (4–6 years old).
[b] Interaction effect for age showing a stronger association in the younger children (4–6 years old) than the older children (7–10 years old).
[c] Interaction effect for diagnose showing a stronger association in the children with bilateral CP than in the children with unilateral CP.

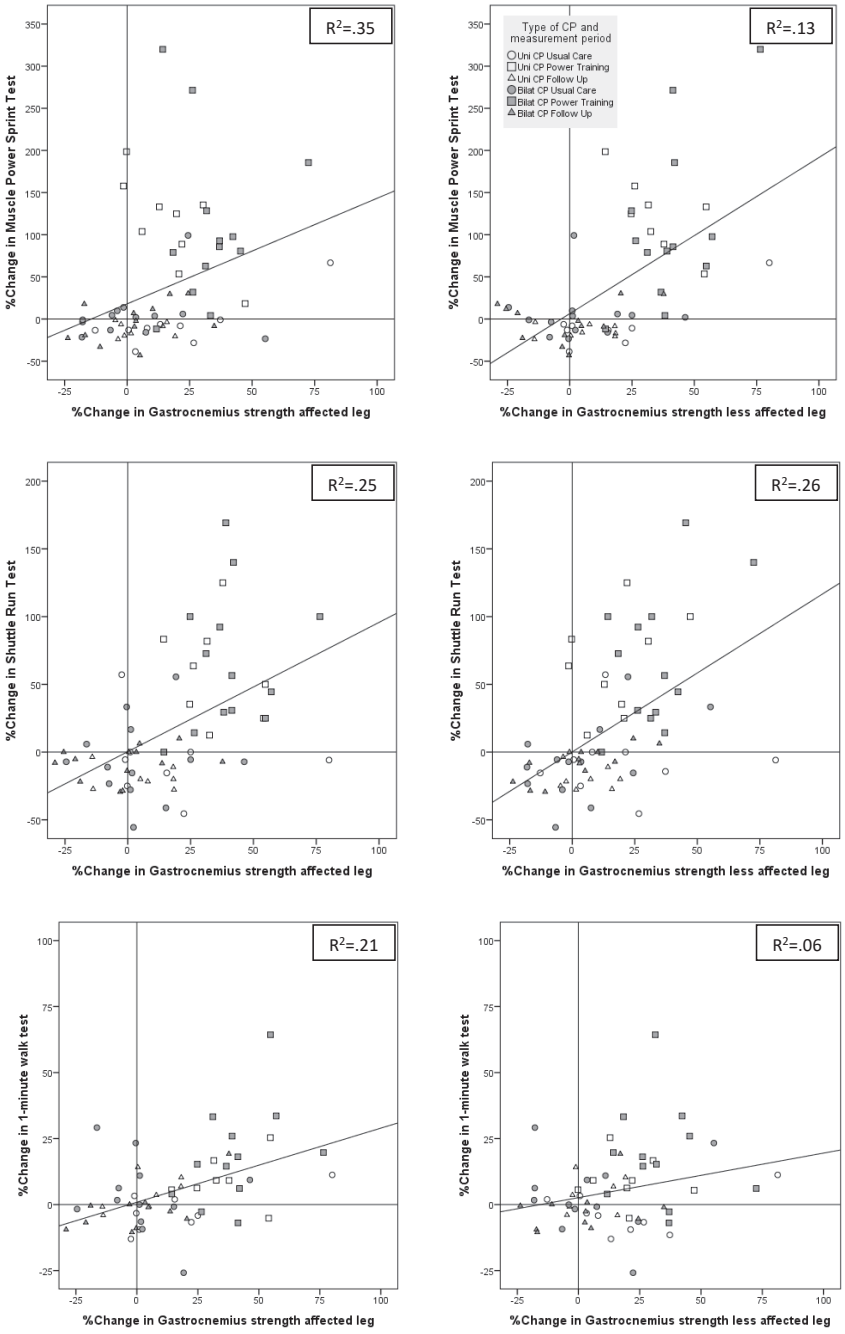

Figure 6.1 Changes in walking capacity (muscle power sprint test, shuttle run test and 1-minute walk test) with changes in isometric muscle strength of m. gastrocnemius (plantar flexor knees extended more and less affected leg) over three time periods (14wks usual care, 14wks power training and 14wks follow-up with usual care).

Table 6.3 shows the results of multivariate regression. Changes in HA strength of both legs had a significant association (R^2=0.66) with MPST changes. Changes in GASTR and HA strength, both in the less affected leg, had a significant association (R^2=0.43) with SRT changes. Changes in GASTR strength in the more affected leg had a significant association (R^2=0.21) with 1MWT changes. KE and SOL strength changes had no significant associations with walking capacity changes in the multivariate models.

Table 6.3 Results of multiple regression analysis for relationship between changes in walking capacity as dependent variable and changes in isometric muscle strength as independent variables

Dependent variables Walking capacity	Independent variable in final model Isometric muscle strength	β (SE)	p-value	R^2 final model
Muscle power sprint test (Δ%)	Constant	7.13 (3.41)		
	Hip abductors affected leg (Δ%)	0.46 (0.14)	.001	
	Hip abductors less affected leg (Δ%)	1.17 (0.21)	<.001	0.66
Shuttle run test (Δ%)	Constant	-3.01 (3.98)		
	Gastrocnemius less affected leg (Δ%)	0.80 (0.36)	.027	
	Hip abductors less affected leg (Δ%)	0.54 (0.14)	<.001	0.43
1-minute walk test (Δ%)	Constant	0.91 (1.56)		
	Gastrocnemius affected leg (Δ%)	0.28 (0.08)	<.001	0.21

Discussion

The aim of the current study was to evaluate whether changes in muscle strength could explain changes in walking capacity in children with CP. We therefore compared the within-subject changes in lower-limb muscle strength with the within-subject changes in walking capacity in three different time periods (14-week usual-care period, 14-week power-training period and a 14-week follow-up period with usual-care). The results of the present study indicated that larger changes in muscle strength are associated to larger changes in walking capacity. Especially the improvements in hip abductor (HA) and gastrocnemius (GASTR) muscle strength were associated to better sprint capacity (R^2=0.66), walking endurance (R^2=0.43), and to a lesser extend to walking speed (R^2=0.21). This suggests that the increased walking capacity after functional power-training is partly due to the increased strength of HA and GASTR.

In a recently published study, we showed the positive effects of functional power-training on muscle strength and walking capacity in young children with CP, which were the same participants as in the current study (13). The participating children acted as their own

controls in this double-baseline design: a 14-week usual-care period was compared with a 14-week functional power-training intervention that followed immediately after the usual-care period (13). No significant changes in muscle strength and walking capacity were seen in the usual-care period. Large increases in strength and walking capacity were found in the functional power period and subsequently in the follow-up period muscle strength remained stable while only small decreases in walking capacity were found (13). In the current study we showed that these within-subject muscle strength changes (no changes, large increases and small decreases) over these three time periods explained the changes in walking capacity.

Previous intervention studies suggested that improvements in walking capacity are not expected after strength training (8;9). In contrast, the strong associations between changes in muscle strength and changes in walking capacity in the present study indicate that improving strength may improve walking capacity. This discrepancy might be attributed to the character of the power-training and secondly to the magnitude of the increases found after the power-training. Firstly, the character of the power-training could be the explanation for these strong associations due to; A) the higher movement velocity used in the progressive loaded strength training exercises in the power-training, and B) the incorporation of the strength exercises in functional movements like walking and sprinting. Secondly, the larger increases in muscle strength (18–128%) compared with previous strength training studies (10;11), is a likely explanation for its positive effect on walking capacity. It is suggested that muscle strength increases in children are primarily neuromuscular changes instead of muscle volume changes (21). Also, Rose et al. (2004) suggested that treatments aimed at building strength, such as high-intensity strength training, may be effective for reducing the movement deficit and improving gait in that they increase voluntary excitatory drive and muscle activation (22). This would indicate that neuromuscular changes could be more effective to increase walking capacity than muscle volume changes alone. This is, however, not evaluated in our study and might be interesting to assess in future research.

The importance of the HA and GASTR muscle strength for improvements in walking capacity is in agreement with previous studies assessing multivariable cross-sectional associations between strength of similar leg muscles and walking in children with CP (3;5-7;23). The difference between these cross-sectional studies and our longitudinal study was that we evaluated the association in changes between strength and walking capacity over three time periods, which could give more information about the effect of increases and decreases in these domains. Lack of association between changes in KE and

changes in walking capacity in the multivariate models in our study might indicate that muscle strengthening should concentrate more on improving HA and GASTR strength when aiming to improve walking capacity.

The functional power-training focused on increasing plantar flexor strength by power exercises that aimed to elicit a powerful push-off during walking (15). Plantar flexors are also often targeted in treatments with botulinum toxin injections, orthoses and serial casting to reduce spasticity, increase ROM in the ankle joint and to increase walking quality. However, as a side effect, these treatments may exacerbate muscle weakness (24) or at least hamper muscle growth. The present results showed that power-training increases plantar flexor strength and subsequently increases walking capacity as a counterweight for these treatments.

In our study we found that changes in muscle strength of the more and less affected leg both influenced walking capacity. Most previously cross-sectional studies, which evaluated associations between walking capacity and muscle strength in CP, only included muscle strength of the non-dominant — presumably the more affected — leg because of the assumption that increasing muscle strength of the non-dominant side would have more effect on walking capacity than the dominant (i.e. stronger) leg (3;5-7). However, a previous study by De Groot et al. (2012) showed that strength of the less affected leg of adults with CP showed stronger correlations with cycling performance than the more affected leg (25). Results of our study suggest that changes in muscle strength of the more affected leg as well as the less affected leg are determining factors for changes in walking capacity.

We found significant interaction effects between muscle strength changes and type of diagnose (bilateral CP versus unilateral CP) in a few associations (13%) between muscle strength and walking capacity, where the children with bilateral CP showed stronger associations than the children with unilateral CP. A study by Balemans et al. (2015) showed that associations between changes in HA strength and changes in walking speed (1MWT) were stronger for children with bilateral CP compared with unilateral CP (26). This is partly in agreement with our findings, indicating that children with bilateral CP could benefit more from strength training to increase walking capacity than children with unilateral CP. Balemans et al. also stated that the different associations in children with unilateral and bilateral CP might explain the fact that previous strength training studies (10;11) did not show an improved walking capacity with increased muscle strength, since no distinction was made between children with bilateral or unilateral CP in these studies (26). However, we found large changes in muscle strength and walking capacity after the

functional power-training in children with bilateral CP as well as in those with unilateral CP (13). The larger changes in isometric muscle strength found in our study (27–44%) (13) than in previous strength training studies (11–26%) (10;11) might be the explanation for finding no interaction effects on type of diagnose in 87% of the associations.

Conclusion

Improvements in HA and GASTR muscle strength of both less and more affected leg were associated to better walking capacity in young children with CP, suggesting that walking capacity, especially sprint capacity, can be improved by increasing strength of these muscles in this population. Based on our results, we can advise to add functional power-training to the treatment program of young children with CP to increase muscle strength and subsequently walking capacity.

What this paper adds

- Improvements in hip abductor and gastrocnemius muscle strength are associated to better walking capacity in young children with CP.
- 66% of the change in sprint power is explained by the change in hip abductor strength.
- Add functional power-training to the treatment program of young children with CP to improve muscle strength and subsequently walking capacity.

Acknowledgements

The authors wish to thank all children, parents and trainers who participated in this study. The authors declare that none of them have any conflicts of interest with respect to the research, authorship, and/or publication of this article. This study has been funded by Mitialto Foundation, Duyvensz-Nagel Foundation, Dutch Rehabilitation Fund and Royal Dutch Society for Physical Therapy.

References

(1) Graham HK, Rosenbaum P, Paneth N, Dan B, Lin JP, Damiano DL, et al. Cerebral palsy. Nat Rev Dis Primers 2016;2:15082.

(2) Bax MC, Flodmark O, Tydeman C. Definition and classification of cerebral palsy. From syndrome toward disease. Dev Med Child Neurol Suppl 2007 Feb;109:39-41.

(3) Dallmeijer AJ, Rameckers EA, Houdijk H, De GS, Scholtes VA, Becher JG. Isometric muscle strength and mobility capacity in children with cerebral palsy. Disabil Rehabil 2015 Nov 25;1-8.

(4) Wiley ME, Damiano DL. Lower-extremity strength profiles in spastic cerebral palsy. Dev Med Child Neurol 1998 Feb;40(2):100-7.

(5) Eek MN, Beckung E. Walking ability is related to muscle strength in children with cerebral palsy. Gait Posture 2008 Oct;28(3):366-71.

(6) Ferland C, Lepage C, Moffet H, Maltais DB. Relationships between lower limb muscle strength and locomotor capacity in children and adolescents with cerebral palsy who walk independently. Phys Occup Ther Pediatr 2012 Aug;32(3):320-32.

(7) Ross SA, Engsberg JR. Relationships between spasticity, strength, gait, and the GMFM-66 in persons with spastic diplegia cerebral palsy. Arch Phys Med Rehabil 2007 Sep;88(9):1114-20.

(8) Franki I, Desloovere K, De CJ, Feys H, Molenaers G, Calders P, et al. The evidence-base for conceptual approaches and additional therapies targeting lower limb function in children with cerebral palsy: a systematic review using the ICF as a framework. J Rehabil Med 2012 May;44(5):396-405.

(9) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014 Feb;35(2):239-49.

(10) Scholtes VA, Becher JG, Janssen-Potten YJ, Dekkers H, Smallenbroek L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. Res Dev Disabil 2012 Jan;33(1):181-8.

(11) Taylor NF, Dodd KJ, Baker RJ, Willoughby K, Thomason P, Graham HK. Progressive resistance training and mobility-related function in young people with cerebral palsy: a randomized controlled trial. Dev Med Child Neurol 2013 Sep;55(9):806-12.

(12) Moreau NG, Holthaus K, Marlow N. Differential adaptations of muscle architecture to high-velocity versus traditional strength training in cerebral palsy. Neurorehabil Neural Repair 2013 May;27(4):325-34.

(13) van Vulpen LF, De GS, Rameckers E, Becher JG, Dallmeijer AJ. Improved Walking Capacity and Muscle Strength After Functional Power-Training in Young Children With Cerebral Palsy. Neurorehabil Neural Repair 2017 Sep;31(9):827-41.

(14) Moreau NG, Falvo MJ, Damiano DL. Rapid force generation is impaired in cerebral palsy and is related to decreased muscle size and functional mobility. Gait Posture 2012 Jan;35(1):154-8.

(15) van Vulpen LF, De GS, Rameckers EAA, Becher JG, Dallmeijer AJ. Effectiveness of Functional Power Training on Walking Ability in Young Children With Cerebral Palsy: Study Protocol of a Double-Baseline Trial. Pediatr Phys Ther 2017 Jul;29(3):275-82.

(16) Smits DW, van Groenestijn AC, Ketelaar M, Scholtes VA, Becher JG, Gorter JW. Selective motor control of the lower extremities in children with cerebral palsy: inter-rater reliability of two tests. Dev Neurorehabil 2010;13(4):258-65.

(17) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability for running tests for measuring agility and anaerobic muscle power in children and adolescents with cerebral palsy. Pediatr Phys Ther 2007;19(2):108-15.

(18) Verschuren O, Takken T, Ketelaar M, Gorter JW, Helders PJ. Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. Phys Ther 2006 Aug;86(8):1107-17.

(19) McDowell BC, Humphreys L, Kerr C, Stevenson M. Test-retest reliability of a 1-min walk test in children with bilateral spastic cerebral palsy (BSCP). Gait Posture 2009 Feb;29(2):267-9.

(20) van Vulpen LF, De GS, Becher JG, de Wolf GS, Dallmeijer AJ. Feasibility and test-retest reliability of measuring lowerlimb strength in young children with cerebral palsy. Eur J Phys Rehabil Med 2013 Dec;49(6):803-13.

(21) Faigenbaum AD, Kraemer WJ, Blimkie CJ, Jeffreys I, Micheli LJ, Nitka M, et al. Youth resistance training: updated position statement paper from the national strength and conditioning association. J Strength Cond Res 2009 Aug;23(5 Suppl):S60-S79.

(22) Rose J, McGill KC. Neuromuscular activation and motor-unit firing characteristics in cerebral palsy. Dev Med Child Neurol 2005 May;47(5):329-36.

(23) Davids JR, Oeffinger DJ, Bagley AM, Sison-Williamson M, Gorton G. Relationship of Strength, Weight, Age, and Function in Ambulatory Children With Cerebral Palsy. J Pediatr Orthop 2015 Jul;35(5):523-9.

(24) Barber LA, Barrett RS, Gillett JG, Cresswell AG, Lichtwark GA. Neuromechanical properties of the triceps surae in young and older adults. Exp Gerontol 2013 Nov;48(11):1147-55.

(25) de Groot S, Dallmeijer AJ, Bessems PJ, Lamberts ML, van der Woude LH, Janssen TW. Comparison of muscle strength, sprint power and aerobic capacity in adults with and without cerebral palsy. J Rehabil Med 2012 Nov;44(11):932-8.

(26) Balemans AC, van WL, Becher JG, Dallmeijer AJ. Associations between fitness and mobility capacity in school-aged children with cerebral palsy: a longitudinal analysis. Dev Med Child Neurol 2015 Jan 12.

Discussion

The aim of this thesis was to evaluate the effectiveness of a functional power-training program to improve muscle strength, walking capacity and mobility performance of young children with spastic cerebral palsy (CP). We also aimed to investigate the feasibility and test-retest reliability of measuring lower-limb strength in young children with CP. In addition, we aimed to assess whether improvements in lower-limb muscle strength could explain improvements in walking capacity in young children with CP.

Chapter 2 showed that it was feasible and reliable to measure lower-limb muscle strength in young children with CP when taking mean values of 2 to 3 test occasions to reduce measurement error. The intervention study (chapters 3–5) evaluated a functional power-training consisting of loaded exercises with a high movement velocity in activities like running, walking and climbing stairs. Results showed that functional power-training improved lower-limb muscle strength, walking capacity and parent-reported mobility performance in young children (4–10 years) with CP. Chapter 6 showed that improvements in muscle strength of the hip abductor and gastrocnemius were associated with improvements in sprint capacity, walking endurance, and walking speed. These results imply that walking capacity can be improved by increasing strength particular of these muscles groups, with functional power-training in children with CP.

Specificity and intensity of strength training

Previous studies on conventional strength training programs have not found significant improvements in walking capacity of children with CP (1;2). Main difference between these conventional strength-training programs and the functional power-training program, which was studied in this thesis, is the specificity of the strength training.

Specificity of training is one of the most basic concepts to incorporate in strength training programs according the guidelines of the National Strength and Conditioning Association, as discussed in the general introduction in this thesis (3). The acronym *SAID*, which stands for *Specific Adaptation to Imposed Demands*, is used to explain specificity in training. The underlying principle is that the type of demand placed on the body dictates the type of adaptation that will occur. Previously conducted strength training studies in children with CP used low movement velocity in their loaded exercises. Everyday activities, however, often involve fast limb movements with contraction times of 50–200 ms (4), especially in daily childhood activities with sprinting and jumping (5;6), and should therefore be trained. In the functional power-training the high movement velocity in the power-exercises was used to adapt the muscles to the high rise in contractile force at the onset of contraction and to the total contractile impulse that can be produced within a

given contraction time. Secondly, the incorporation of the progressively loaded power-exercises in functional movements like running, walking and climbing stairs, was used to increase specificity of training so there was no need to transfer newly gained strength into a new movement.

Training intensity is, besides specificity, an important factor in the design of a resistance training program because it is the major stimulus related to changes in muscular fitness (7-10). Training intensity usually refers to the amount of resistance used for a specific exercise (e.g., percentage of 1-repetition maximum [1-RM]) (7;8;10). Fry (2004) defined intensity as a function of power in a study of the role of resistance exercise intensity on muscle fiber adaptations (9). According to this definition exercises that uses larger resistance and/or faster movement velocity will have a greater exercise intensity (9).

Training intensity of power-exercises in the functional power-training were progressively loaded with external weights and an imposed movement velocity (see Chapter 3, Table 3.3). The exercise intensity was determined by the velocity of the exercise (70% of their maximum unloaded speed during 25s) and maximum load that was added to the movement to reach the 70% of their maximum unloaded speed. This can be seen as a new kind of training and is hard to fit in a conventional strength training intensity where a 1-RM for a specific exercise is determined for one movement such as a squat, a deadlift or a snatch. A variety of terms are used for training programs with the aim to increase muscle strength, running speed and muscle power (see Table 7.1). Similarities regarding training principles in different intervention programs shown in Table 7.1, are the use of maximal-effort or "all-out" in the exercises, duration <30 seconds and progressiveness. The contrasts in these different types of interventions (Table 7.1) lay in different approaches of progressiveness (i.e., distance, external loads, movement speed) and different focus and aim of the intervention (i.e., improvement of muscle strength, muscle power, running economy, agility, anaerobic capacity, cardio-respiratory fitness) which determines the exercise time.

The repetitions of 25–30 seconds at high intensity in the power-exercises, used in our functional power-training program, are similar to the duration of exercise used in High-intensity Interval Training methods (HIT) (11). HIT can be defined as repeated bouts of short to moderate duration exercise (i.e. 10 seconds to 5 minutes) completed at high intensity (11). HIT programs are known to increase anaerobic and aerobic capacity (11-14). We found large improvements in sprint power and in the walking/running endurance after the functional power-training and it is expected that the children in our study increased their anaerobic and aerobic capacity as well. More research is needed

to identify the different components (e.g. VO_2peak, muscle power, energy cost during walking/sprint) of the improvements in sprint power and endurance after functional power-training.

The power-training program especially addressed the muscle weakness, which is seen as an important and influential impairment that affects walking ability and participation in daily life activities in children with CP. We, therefore, designed a training program that we could monitor carefully and control the progressiveness of the load in the exercises to be sure to get sufficient exercise intensity. Insufficient exercise intensity is often seen as the important factor of the failure in increasing muscle strength and walking capacity of previously studied strength training protocols (7-10).

How to reach and maintain the intensity in the power exercises?

In the functional power-training children had to perform each exercise on maximal effort (in 25s) and they had to repeat the same exercise 6 to 8 times with a rest period of 30 to 50s. This is, of course, quite intense for children. Some children had never exercised at this intensity before, e.g. they felt their increased heartbeat for the first time and never experienced sweating before. To be able to reach a sufficient exercise intensity, with maximal effort of the child in the exercises, it was important to make the training as fun as possible and to motivate the children as much as we could. Children cannot be forced in participating in a training they do not like. To reach the intensity in the power-exercises the training was performed in small groups and the children were supervised individually by a trainer. In this way we could effectively and safely guide the children through the exercises. All trainers received a training to improve knowledge about the training principles and the importance of the training intensity. It was sometimes difficult for trainers/therapists to encourage and motivate the children to give maximal effort in every power-exercise, especially when the trainers noticed that children got really tired during the exercises. Not all physiotherapist and occupational therapists were used to train children at this high intensity.

Braaksma et al. pointed out, in a systematic review on characteristics of physical activity interventions in children aged 6–12 years, that it is challenging to attain and maintain high training intensities in this group (15). In this review, studies with a higher effect size were intervention programs that tailored their training intensity more to the individual child and programs that trained smaller groups, allowing for closer supervision and thereby higher training intensity (15).

To enhance the children's' motivation and enjoyment in the power-training program and therewith to obtain the set training intensity, we used different motivational strategies (16;17):

- *Group training and storyline:* We trained in small groups (4–6 children) and included a storyline in which the children were asked for their help by a secret agent to solve a crime that was committed by villains (see Figure 7.1).
- *Involvement of friends and siblings:* Friends and siblings joined a training session. During this training session children with CP showed proudly how hard they could train and how strong and fast they had become in the exercises. Their friends and siblings could feel for themselves how intense the power-exercises were.
- *Progress charts to record exercise achievements:* Progress in the exercises was recorded in the children's training charts to monitor the progression in load in the exercises and to motivate children whom could see their one improvements.
- *Personal guidance of the child:* Personal guidance of the children made it possible to recognize and praise every individual accomplishment, to motivate the children to give maximal effort in the exercises, to give the child the right amount of rest between the exercises and it helped the children to focus on their individual goals. The personal guidance also ensured that the exercises were performed correctly and safe.
- *Establishing short-term goals with parents and children:* Goals of the training set by parents, and when possible the children, made it clear for parents and children why they were following this training program and what they were training for.

All these strategies created a bond between the children (and parents) who participated in the training group. The group setting of the training, i.e. to see their friends train hard, also made it easier for the children to still give their best when they were tired and thought they could not perform an exercise any more.

We highly recommend to take all these motivational strategies into account when implementing functional power-training.

Feasibility

The functional power program showed that it is feasible for children and their families to train 3 months, 3 times a week for 60 minutes. We actually experienced that parents and children want to repeat the training period yearly. This might be explained by the fact that they experienced the effectiveness in walking ability and reaching individual goals, and because the children enjoyed the training so much. Contributing factor is a

Figure 7.1 Motivational strategies used in the power-training with individual super hero on t-shirt, impression of the storyline with secret missions and assignments for the children.

spacious waiting area with coffee and tea facilities, were siblings can play or do their homework, parents can have conversations or finish their work. The power-training is now implemented in the regular care at the rehabilitation center in Amsterdam and, therefore, it is possible that children follow the power-training program multiple times during their youth depending on the goals set in their rehabilitation treatment.

Strength training in young children

Children with CP establish their gross motor patterns as walking, running and jumping around the age of 7 years old, depending on their GMFCS level (18). The quality of motor control used to accomplish the activities emerge later in childhood. Guidelines for strength training in children advice to start training children from the age of 6–7 years old, because for children younger than 6–7 years of age it is more difficult to correctly perform unknown strength exercises, to follow instructions and it is more difficult to get them to perform at their maximal effort to get the right intensity (overload principles)

(3;8;10). Despite these guidelines we included children from the age of 4 years old in our power-training, because we wanted to improve strength and walking capacity at this early age due to the known decreases in muscle strength already at this age, problems with gross motor activities and the start at primary school at the age of 4 years. The incorporation of the power-exercise in functional movements like walking, running and climbing stairs made it possible for young children to participate in the power-training program because they were already familiar with these movements.

Fun experiences with training at an early age

Furthermore, it is likely that improvements in muscle strength and motor skill performance with training interventions during the growing years will facilitate the establishment of desired behaviors and provide an optimal mechanism for promoting physical activity as an ongoing lifestyle choice (8). This view is supported by a longitudinal study showing that 6-years-old typically developing (TD) children with low and average levels of motor coordination, had lower levels of physical activity at age 10 compared with children with higher levels of motor coordination (19). Others reported that low levels of motor skill competency among school-age youth were associated with reduced levels of physical activity, low levels of cardio-respiratory fitness and increased risk of being overweight or obese (20). It is therefore important to start training and having fun experiences with exercise and sport at an early age.

Muscle strength changes

Increases in isometric and dynamic muscle strength from 18–128% were found after the power-training. However, it is unknown which underlying adaptations in neural regulation of muscular activity have occurred as a result of the functional power-training. In TD children, increases in muscle strength following resistance training are more attributed to neural changes (improvements in motor skills, increases in motor unit recruitment and firing rate, and changes in coordination) than to muscle hypertrophy (8;21;22). Furthermore, when muscle hypertrophy has been found after strength training in youth the change is small compared with the change in strength and with the hypertrophy found after training in adults (7). However, Gillett et al. concluded in a systematic review, that there was preliminary evidence that strength training leads to muscle hypertrophy in children and adolescents with CP (23). The muscle morphology and architecture of individuals with CP differ from TD individuals. Individuals with CP have a reduced muscle volume (24), reduced or similar fascicle lengths (25;26), increased intramuscular

fat (27), and increased tendon length (28). These differences in muscle morphology and architecture are thought to be contributing to the reduced muscle strength in children with CP compared with TD children. However, the morphological and architectural adaptation of muscle in response to the power-training remains unknown. To get a better understanding of the muscular responses to the power-training in this population we need to assess muscle volume. Full weight bearing 3D imaging (PedCAT) or 3-dimensional ultrasound measuring systems can be used to measure muscle volume (24;29). These measurements have to be easy to perform in a short time span, and has to be accurate and reliable so it can be used for young children with CP in a clinical environment (24;30).

Reversibility of training

The reversibility of training is sometimes referred to as "use it or lose it". As mentioned before, the acronym SAID explains that the human body makes adaptations to cope with the stresses placed on it during periods of exercise (3). This principle is used in training by overloading the body's systems beyond their normal levels over a period of time so that the system will adapt and it will become its new norm. During periods of inactivity the human body reverses these adaptations in an attempt to return itself to a norm as this is the current level of stress placed upon it. Therefore gains that have been made after a training period will be lost when there is a period of inactivity (3).

In the follow-up period after the power-training we saw that some, but not all, of the positive effects persisted in the follow-up period. Although we found a significant decrease in sprint capacity and in the shuttle run test, participants still had a better sprint capacity (67%) and higher shuttle run test result (33%) at the end of the follow-up period compared with the start of the functional power-training period. The increases in muscle strength were even maintained in the follow-up period.

It can be speculated that these unexpected good results are consequences of increased physical activity: we found gains in the parent-reported mobility and participation level after the power-training period. Children reaching their goals like "playing more soccer with my classmates at the schoolyard", "being able to walk the stairs more easily at school" and "being able to walk to school" are activities that are likely to be executed on daily bases during the follow-up period. It is therefore possible that these activities prevented a decrease of the power-training effect in the follow-up. However, it would be interesting to know how the body adaptations are after a longer follow-up period than the 4 months we used in this study.

Directions for future research

Future research is needed to get more understanding whether the increases in muscle strength and walking capacity led to a better walking efficiency, i.e. whether walking can be performed with a lower energy expenditure (31;32). This could have important clinical implications in the treatment of children with CP especially for those who have fatigue-related walking problems.

It is also unknown if the shown gains in mobility after the power-training in the children with GMFCS level I and II, do also apply to the children with GMFCS III and IV. Power-exercise should be adjusted for this population but training intensity and specificity, such as the original functional power training program, should remain the same.

We do not know if the same improvements in muscle strength and walking capacity after functional power-training will be found in adolescents and adults with CP, although, decreased lower-limb muscle strength has strong associations with decreased mobility in this population (33;34). Increases in muscle strength in TD adolescents and adults are more attributed to muscle hypertrophy than neural changes (8;21;22) and the power-exercises could be addressing more the neural changes. However, specificity of the power-exercises could enhance improvement of both muscle strength and mobility also in this older population.

Finally, further research is needed to investigate if we can transfer the principles of the functional power-training to the upper extremities as there is increasing evidence that muscle weakness of the upper limb also decreases the ability of children with CP to perform daily activities (35-38).

Implications for future research and clinical practice

- Evaluate muscle physiology and muscle architectural changes after power-training
- Evaluate walking efficiency and gait quality (kinematics, kinetics and timing of muscle activation) changes after power-training
- Evaluate muscle strength and activities in adolescents and adults with CP and in children with GMFCS III and IV after power-training
- Evaluate upper-extremity muscle strength and tasks in children and adolescents with CP after power-training

Table 7.1 Interventions with strength and/or speed components

Intervention	Study examples	Subjects	Training duration	Frequency of the training	Intensity	Type of exercises	Aim
Progressive Resistance Exercise (PRE) / Functional Strength Training	Scholtes et al. (39;40)	CP[1] 10.4y±1.10y Range 6–13y	12 wks	3 times a week, 45–60 minutes	3 progressively loaded exercises with 3 sets of 8 rep at 25–100% 8-RM 1 unloaded exercise 7–10 min per exercise 90 sec rest	Leg-press and functional exercise; e.g. sit-to-stand, climbing stairs	Muscle strength, strength/ endurance, gross motor function
	Taylor et al. (41)	CP 18.2y±1.11y	12 wks	2 times a week	Progressively loaded exercises 3 sets of 10–12 rep at 10–12RM 2 min rest between sets	Weights machines Lower-limb exercises	Muscle strength and mobility-related function
High-velocity strength training	Moreau et al. (42)	CP 13.9y±2.6y	8–10 wks 24 sessions	3 times a week	6 sets, 5 rep maximum-effort contractions, 30 deg/s to 120deg/s 1 min rest between sets	Isokinetic knee extension on Biodex dynamometer	Muscle power, muscle size/ muscle strength, fascicle length, walking performance
Explosive strength training (EST) / Progressive heavy resistance training	Kirk et al. (43)	CP 36.5y Range 18–59y	12 wks	3 times a week	3 sets of 12RM to 6-RM 90 sec rest in between sets Movement velocity: 1 sec concentric, 3 sec excentric	Weights machines 4 lower-body exercises conducted unilateral both sides 1 lower back exercise 1 abdominal exercise	Muscle strength, rate of force development, gait function, passive ankle joint stiffness
	Paavolainen et al. (44)	Endurance Athletes 23y±3y	9 wks	32% of endurance training replaced by EST of 9 training times a week	20–100m sprints 5–10 rep 30–200 rep jumping exercises and leg press, 0–40% of 1-RM max movement velocity 5-20 rep per set 15-90 min in total per EST	Various unloaded sprints Unloaded and loaded jumping exercises Leg-press	5-km running performance, muscle power, running economy
Plyometric exercises	Johnson et al. (45)	CP 9.3y Range 8–10y	8–14 wks	2 times a week 40 min	4 jumping and 4 throwing exercise; 3 sets of 5 rep of 8 plyometric (50-150 jumps and 55–120 throws per session) Progressiveness in number of rep, weight (ball), distance or height. 30–90 sec rest between sets	Jumping exercises and throwing exercise focused on horizontal power and vertical power.	Gross motor ability, agility, running speed, power

Intervention	Study examples	Subjects	Training duration	Frequency of the training	Intensity	Type of exercises	Aim
High-intensity Interval Training (HIT)	Astorino et al. (46)	TD[2] 25.3y±4.5y	2–3 wks	2–3 times a week Total of 6 sessions	4–6 repeated Wingate tests 30 sec 'all-out' with 5 min active recovery	Wingate ergometer	Cardiovascular function, Cardio respiratory fitness, Muscle power
Speed training	Kawamori et al. (47)	TD HG: 22.8y±3.3y LG: 22.3y±5.2y	8 wks	2 times a week	HG[3]: weigthed sled towing with heavy loads that reduced sprint velocity with 30% LG[4]: weigthed sled towing with light loads that reduced sprint velocity with 10% Max-effort sprint 3–4 rep of 5m, 3–6 rep of 10m and 3–4 rep of 15m with 1–2 min rest between rep. Progressiveness in external load and number of repetitions.	Short sprints with towing a Sprint runs with weighted sled towing	Sprint acceleration 0–5 m, muscle power
Speed endurance training / HIT	Iaia et al. (13)	Young endurance athletes	4 wks	3–4 times a week	8–12 rep of 30s max-effort sprint runs with 3 min rest 90–95% speed	Sprint runs unloaded	Adaptations of skeletal muscle ion transport proteins, work capacity, work performance
Aerobic and anaerobic exercises	Verschuren et al. (48)	CP 11.6y±2.5y	8 months	2 times a week 45 min	• First 3 months aerobic exercises: 3 rep 3 to 6 min 60–80% with 3–6 min rest in between rep. • Second 3 months combined aerobic exercise with anaerobic exercise: 2–3 sets of 5 rep of 20–25sec max effort • Last 2 months anaerobic exercise: 3 sets of 5 rep of 25–30sec max effort	Aerobic and anaerobic exercises	Aerobic capacity, anaerobic capacity, muscle strength, health related Quality of Life

1. CP = cerebral palsy; 2. TD = typically developing individuals; 3. HG = high-loaded group; 4. LG = light-loaded group

References

(1) Franki I, Desloovere K, De CJ, Feys H, Molenaers G, Calders P, et al. The evidence-base for conceptual approaches and additional therapies targeting lower limb function in children with cerebral palsy: a systematic review using the ICF as a framework. J Rehabil Med 2012 May;44(5):396-405.

(2) Park EY, Kim WH. Meta-analysis of the effect of strengthening interventions in individuals with cerebral palsy. Res Dev Disabil 2014 Feb;35(2):239-49.

(3) Baechle TR, Earle RW. Essentials of Strength training and conditioning. third ed. Champaign, Il 61825-5076: Human Kinetics; 2008.

(4) Aagaard P, Simonsen EB, Andersen JL, Magnusson P, Dyhre-Poulsen P. Increased rate of force development and neural drive of human skeletal muscle following resistance training. J Appl Physiol (1985) 2002 Oct;93(4):1318-26.

(5) Bailey RC, Olson J, Pepper SL, Porszasz J, Barstow TJ, Cooper DM. The level and tempo of children's physical activities: an observational study. Med Sci Sports Exerc 1995 Jul;27(7):1033-41.

(6) Van Praagh E, Dore E. Short-term muscle power during growth and maturation. Sports Med 2002;32(11):701-28.

(7) Faigenbaum AD, Kraemer WJ, Blimkie CJ, Jeffreys I, Micheli LJ, Nitka M, et al. Youth resistance training: updated position statement paper from the national strength and conditioning association. J Strength Cond Res 2009 Aug;23(5 Suppl):S60-S79.

(8) Faigenbaum AD, Lloyd RS, Myer GD. Youth resistance training: past practices, new perspectives, and future directions. Pediatr Exerc Sci 2013 Nov;25(4):591-604.

(9) Fry AC. The role of resistance exercise intensity on muscle fibre adaptations. Sports Med 2004;34(10):663-79.

(10) Verschuren O, Ada L, Maltais DB, Gorter JW, Scianni A, Ketelaar M. Muscle strengthening in children and adolescents with spastic cerebral palsy: considerations for future resistance training protocols. Phys Ther 2011 Jul;91(7):1130-9.

(11) Laursen PB, Jenkins DG. The scientific basis for high-intensity interval training: optimising training programmes and maximising performance in highly trained endurance athletes. Sports Med 2002;32(1):53-73.

(12) Gibala MJ. High-intensity interval training: a time-efficient strategy for health promotion? Curr Sports Med Rep 2007 Jul;6(4):211-3.

(13) Iaia FM, Thomassen M, Kolding H, Gunnarsson T, Wendell J, Rostgaard T, et al. Reduced volume but increased training intensity elevates muscle Na+-K+ pump alpha1-subunit and NHE1 expression as well as short-term work capacity in humans. Am J Physiol Regul Integr Comp Physiol 2008 Mar;294(3):R966-R974.

(14) Iaia FM, Bangsbo J. Speed endurance training is a powerful stimulus for physiological adaptations and performance improvements of athletes. Scand J Med Sci Sports 2010 Oct;20 Suppl 2:11-23.

(15) Braaksma P, Stuive I, Garst RME, Wesselink CF, van der Sluis CK, Dekker R, et al. Characteristics of physical activity interventions and effects on cardiorespiratory fitness in children aged 6-12 years-A systematic review. J Sci Med Sport 2017 Jul 20.

(16) Franklin BA. Motivating patients to exercise: strategies to increase compliance. Sports Med Digest ed. 1994.

(17) Medina J. Brain Rules. Second edition ed. Pear Press; 2014.

(18) Rosenbaum PL, Walter SD, Hanna SE, Palisano RJ, Russell DJ, Raina P, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. JAMA 2002 Sep 18;288(11):1357-63.

(19) Lopes VP, Rodrigues LP, Maia JA, Malina RM. Motor coordination as predictor of physical activity in childhood. Scand J Med Sci Sports 2011 Oct;21(5):663-9.

(20) Hardy LL, Reinten-Reynolds T, Espinel P, Zask A, Okely AD. Prevalence and correlates of low fundamental movement skill competency in children. Pediatrics 2012 Aug;130(2):e390-e398.

(21) McNee AE, Gough M, Morrissey MC, Shortland AP. Increases in muscle volume after plantarflexor strength training in children with spastic cerebral palsy. Dev Med Child Neurol 2009 Jun;51(6):429-35.

(22) Stackhouse SK, Binder-Macleod SA, Lee SC. Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. Muscle Nerve 2005 May;31(5):594-601.

(23) Gillett JG, Boyd RN, Carty CP, Barber LA. The impact of strength training on skeletal muscle morphology and architecture in children and adolescents with spastic cerebral palsy: A systematic review. Res Dev Disabil 2016 Sep;56:183-96.

(24) Barber L, Hastings-Ison T, Baker R, Barrett R, Lichtwark G. Medial gastrocnemius muscle volume and fascicle length in children aged 2 to 5 years with cerebral palsy. Dev Med Child Neurol 2011 Jun;53(6):543-8.

(25) Moreau NG, Teefey SA, Damiano DL. In vivo muscle architecture and size of the rectus femoris and vastus lateralis in children and adolescents with cerebral palsy. Dev Med Child Neurol 2009 Oct;51(10):800-6.

(26) Shortland AP, Harris CA, Gough M, Robinson RO. Architecture of the medial gastrocnemius in children with spastic diplegia. Dev Med Child Neurol 2002 Mar;44(3):158-63.

(27) Noble JJ, Fry NR, Lewis AP, Keevil SF, Gough M, Shortland AP. Lower limb muscle volumes in bilateral spastic cerebral palsy. Brain Dev 2014 Apr;36(4):294-300.

(28) Barber L, Barrett R, Lichtwark G. Medial gastrocnemius muscle fascicle active torque-length and Achilles tendon properties in young adults with spastic cerebral palsy. J Biomech 2012 Oct 11;45(15):2526-30.

(29) Wachowsky M, D'Souza S, Wirth T. Comparison of the static tibia-hindfoot angle and arch height between the Oford Foot Model (OFM) and a full weight bearing 3D-imaging (PedCAT). Gait Posture 2016;49S.

(30) Fry NR, Gough M, Shortland AP. Three-dimensional realisation of muscle morphology and architecture using ultrasound. Gait Posture 2004 Oct;20(2):177-82.

(31) Balemans AC, Bolster EA, Brehm MA, Dallmeijer AJ. Physical Strain: A New Perspective on Walking in Cerebral Palsy. Arch Phys Med Rehabil 2017 Dec;98(12):2507-13.

(32) Bolster EAM, Balemans ACJ, Brehm MA, Buizer AI, Dallmeijer AJ. Energy cost during walking in association with age and body height in children and young adults with cerebral palsy. Gait Posture 2017 May;54:119-26.

(33) Balemans AC, Van Wely L, Becher JG, Dallmeijer AJ. Associations between fitness and mobility capacity in school-aged children with cerebral palsy: a longitudinal analysis. Dev Med Child Neurol 2015 Jan 12.

(34) Dallmeijer AJ, Rameckers EA, Houdijk H, De Groot S, Scholtes VA, Becher JG. Isometric muscle strength and mobility capacity in children with cerebral palsy. Disabil Rehabil 2015 Nov 25;1-8.

(35) Braendvik SM, Elvrum AK, Vereijken B, Roeleveld K. Relationship between neuromuscular body functions and upper extremity activity in children with cerebral palsy. Dev Med Child Neurol 2010 Feb;52(2):e29-e34.

(36) Braendvik SM, Elvrum AK, Vereijken B, Roeleveld K. Involuntary and voluntary muscle activation in children with unilateral cerebral palsy--relationship to upper limb activity. Eur J Paediatr Neurol 2013 May;17(3):274-9.

(37) Smits-Engelsman BC, Rameckers EA, Duysens J. Late developmental deficits in force control in children with hemiplegia. Neuroreport 2004 Aug 26;15(12):1931-5.

(38) Smits-Engelsman BC, Rameckers EA, Duysens J. Muscle force generation and force control of finger movements in children with spastic hemiplegia during isometric tasks. Dev Med Child Neurol 2005 May;47(5):337-42.

(39) Scholtes VA, Becher JG, Comuth A, Dekkers H, Van DL, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise strength training on muscle strength and mobility in children with cerebral palsy: a randomized controlled trial. Dev Med Child Neurol 2010 Jun;52(6):e107-e113.

(40) Scholtes VA, Becher JG, Janssen-Potten YJ, Dekkers H, Smallenbroek L, Dallmeijer AJ. Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial. Res Dev Disabil 2012 Jan;33(1):181-8.

(41) Taylor NF, Dodd KJ, Baker RJ, Willoughby K, Thomason P, Graham HK. Progressive resistance training and mobility-related function in young people with cerebral palsy: a randomized controlled trial. Dev Med Child Neurol 2013 Sep;55(9):806-12.

(42) Moreau NG, Holthaus K, Marlow N. Differential adaptations of muscle architecture to high-velocity versus traditional strength training in cerebral palsy. Neurorehabil Neural Repair 2013 May;27(4):325-34.

(43) Kirk H, Geertsen SS, Lorentzen J, Krarup KB, Bandholm T, Nielsen JB. Explosive Resistance Training Increases Rate of Force Development in Ankle Dorsiflexors and Gait Function in Adults With Cerebral Palsy. J Strength Cond Res 2016 Oct;30(10):2749-60.

(44) Paavolainen L, Hakkinen K, Hamalainen I, Nummela A, Rusko H. Explosive-strength training improves 5-km running time by improving running economy and muscle power. J Appl Physiol 1999 May;86(5):1527-33.

(45) Johnson BA, Salzberg C, MacWilliams BA, Shuckra AL, D'Astous JL. Plyometric training: effectiveness and optimal duration for children with unilateral cerebral palsy. Pediatr Phys Ther 2014;26(2):169-79.

(46) Astorino TA, Allen RP, Roberson DW, Jurancich M. Effect of high-intensity interval training on cardiovascular function, VO2max, and muscular force. J Strength Cond Res 2012 Jan;26(1):138-45.

(47) Kawamori N, Newton RU, Hori N, Nosaka K. Effects of weighted sled towing with heavy versus light load on sprint acceleration ability. J Strength Cond Res 2014 Oct;28(10):2738-45.

(48) Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Uiterwaal CS, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. Arch Pediatr Adolesc Med 2007 Nov;161(11):1075-81.

Summary

Children with cerebral palsy (CP) often experience problems with keeping up with their peers in daily-life walking and running activities due to muscle weakness. Strength training programs are commonly used in clinical practice to improve muscle strength and therewith walking capacity. These strength training programs showed, however, inconclusive evidence for improving walking capacity despite some improvements in muscle strength. Recent insights suggested that strength training with high-movement velocity incorporated in functional movements might be more effective for improving walking capacity than traditional strength training programs. Therefore, the primary aim of this thesis was to evaluate the effectiveness of a functional high-velocity resistance training (power-training) on muscle strength, walking capacity and parent-reported mobility performance of young children with CP. First, we aimed to investigate the feasibility and test-retest reliability of measuring lower-limb strength in young children with CP. In addition, we aimed to investigate whether changes in lower-limb muscle strength could explain changes in walking capacity in young children with CP. The aims are elucidated in **chapter 1**.

Chapter 2 described the results of a study that investigated the feasibility, test-retest reliability and the optimal test design of lower-limb muscle strength measurements in young children with CP.

Isometric muscle strength of the hip abductor, knee extensor, and the plantar flexors was assessed with handheld dynamometry (HHD) and dynamic muscle strength of the ankle plantar flexor was assessed with the standing heel-rise test (SH), on two test occasions. Twenty ambulatory children with spastic CP (3–5 years (n=10) and 6–10 years (n=10)) were included in this study. Intraclass correlation coefficients (ICC) and Smallest Detectable Differences (SDD) were calculated to determine the optimal test design for detecting changes in strength. The results showed that the strength test instructions were easily understood by the children, also in the younger age group. The ICC values were all above 0.77 for a single measurement, which indicated good reliability. Although the SEM and SDD's were high in a single test occasion—especially in the 3–5 years old children—the isometric strength tests had acceptable SDDs (9–30%), when taking the mean values of 2–3 test occasions (separate days) and 2–3 repetitions. The SH test had large SDDs (40–128% for the younger age group, 23–48% for the older age group). The results of this study can be used to determine an individual test design for HHD measurement in young children with CP, depending on expected changes in muscle strength, muscle group and age of the child. Choosing the most optimal test design is a balance between the smallest SDD and the feasibility of performing repeated measures on separate days. The SH test

in young individuals with CP can only be used to determine large changes in dynamic plantar flexor strength when the average is taken over 3 test occasions.

Chapter 3 described the protocol of a double-baseline designed trial investigating the effectiveness of functional power-training on muscle strength and walking ability in young children with CP. It was aimed to include 22 children with bi- or unilateral spastic CP, aged 4–10 years, and to compare a 14-weeks functional power-training (3 times a week) with a 14-weeks usual care period prior to the functional power-training and a 14 weeks follow-up period. This study protocol provided a detailed description of the design and methodology of the newly developed functional power-training and the measurements to investigate its effectiveness. The power-training exercises were loaded and performed at 50–70% of the maximum unloaded walking and running speed of the children. Load was increased when exercises were performed faster than 70% of the unloaded speed. Primary outcomes were sprinting capacity (15-m Muscle Power Sprint Test) and goal attainment scaling score of walking-related treatment goals. Secondary outcomes were walking speed (1-min walk test), endurance (10-m shuttle-run test), gross motor function, lower-limb strength and parent-reported mobility.

Chapters 4 and 5 described the results of the double-baseline study comparing the within-subjects changes in muscle strength and walking ability of a 14-weeks usual care period with the changes in a 14-weeks functional power-training period and the changes in a 14-weeks follow-up period, in young children with CP. Twenty-two children with a mean age of 7.5 years old and a spastic CP (13 bilateral and 9 unilateral) participated in this study. Changes during the training period were significantly larger than changes in the usual care period for all outcome measures. **Chapter 4** showed that large improvements were found during the training period for walking capacity (13–83% increases) and for muscle strength (18–128%), while outcomes remained stable in the usual care period. The increases in muscle strength and in walking speed were maintained in the follow-up period. A significant decrease in sprint capacity and in the shuttle run test was found in the follow-up period, however, participants, still had a better sprint capacity (67%) and shuttle run test outcome (33%) at the end of the follow-up period compared with the start of the functional power-training period. **Chapter 5** described the results of the study on the achievement on individual treatment goals and parent-reported mobility. Outcome measures were goal attainment scaling (GAS) of individual daily-life walking activity related treatment goals, mobility performance measured with the Functional Mobility Scale (FMS), and the parent-reported Mobility Questionnaire (MobQues). After the power-training 86% of the children achieved or exceeded their goal compared to 14%

in the usual care period. The probability for improvement by one point or more on the FMS-500 meters after functional power-training was 10 times higher compared to the usual care period. At FMS-500m 52% of the children used a walking frame, wheelchair or support by their parents at the start of the functional training period, whereas at the end of the functional power-training period only 13.6% of the children were dependent on a walking aid, wheelchair or support by their parents. No changes were found in the FMS-5 and FMS-50 meter categories. Improvement on the MobQues was significantly larger after power-training compared to usual care (7.9% (95% CI 2.7–13.0, p=.005)). Improvements in treatment goals and parent-reported mobility were maintained in the follow-up period. These results indicated that strengthening interventions are effective in improving muscle strength *and* walking ability in young children with CP if they involve high-velocity strengthening exercises incorporated in functional movements of sufficient intensity (maximal effort, 3 sets of 6 to 8 repetitions), frequency (3 times a week) and duration (14 weeks).

The relationships and the clinical implications between the changes in lower-limb muscle strength and the changes in walking capacity during the 14-weeks periods of usual care, functional power-training and follow-up, are described in **chapter 6**. The multivariate relationships between within-subject changes in muscle strength (isometric strength of gastrocnemius (GASTR), soleus, knee extensors, hip abductors (HA)) and walking capacity were evaluated. The results showed that changes in HA were strongly associated with changes in sprint capacity, changes in GASTR en HA (least affected legs) were associated to changes in walking endurance, and changes in GASTR (most-affected leg) were associated to changes in walking speed. The results of this study implied that walking capacity, especially sprint capacity, can be improved by increasing strength of these muscles, with functional power-training, in this population. Further research about the working mechanism on, e.g., the level of muscle morphology is needed to understand the underlying physiology of the increase of muscle strength after functional power-training.

In conclusion, young children with CP can improve their muscle strength and walking ability with functional power-training, indicating that this type of training is important to be considered in rehabilitation treatment. **Chapter 7** presented an extensive discussion of the studies described in this thesis and their clinical implications. The main findings indicated that with increasing movement velocity and functionality in strength training programs it is possible to improve muscle strength, walking capacity and mobility performance in children with CP.

Samenvatting

Kinderen met cerebrale parese (CP) ervaren vaak dat ze hun leeftijdsgenootjes niet goed bij kunnen houden tijdens het lopen en rennen. Eén van de belangrijkste oorzaken van de ervaren problemen in de loopvaardigheid is de verminderde spierkracht waar de kinderen met CP mee te maken hebben.
Krachttraining om de spierkracht te verbeteren en daarmee de loopcapaciteit, is een veel gebruikte behandeling bij deze kinderen. Wetenschappelijk onderzoek naar de effecten van krachttraining laten echter wisselende resultaten zien. Sommige onderzoeken laten wel een verbetering zien van spierkracht in een aantal van de getrainde spieren maar geen verbetering in de loopcapaciteit van kinderen met CP. Recente wetenschappelijke studies suggereren dat krachttraining uitgevoerd op een hogere snelheid of in een functionele beweging effectiever zou kunnen zijn dan de traditionele vormen van krachttraining. Vandaar dat we een training hebben opgezet, functionele power training genoemd, waarin we deze twee vormen van trainen hebben samengevoegd: 1) krachttraining met een hoge bewegingssnelheid; 2) uitgevoerd in functionele bewegingen zoals het lopen, rennen en traplopen. Dit is beschreven in **hoofdstuk 1**.
Het belangrijkste doel van dit promotieonderzoek was het evalueren van de effectiviteit van de functionele power training op het verbeteren van spierkracht, loopsnelheid, sprintsnelheid, uithoudingsvermogen en de door ouders gerapporteerde mobiliteit van hun kind met CP. Daarvoor wilden we eerst onderzoeken of het mogelijk was om de spierkracht op een betrouwbare manier bij jonge kinderen met CP te meten. Daarnaast wilden we onderzoeken of eventuele veranderingen in spierkracht de eventuele veranderingen in loopvaardigheid konden verklaren.

Hoofdstuk 2 beschrijft de resultaten van het onderzoek naar de haalbaarheid en test-hertest betrouwbaarheid van het meten van de spierkracht in de benen van jonge kinderen met CP. Tevens berekenden we met de resultaten van dit onderzoek een optimale testprocedure per beenspier. Isometrische spierkracht van de heupabductoren, knie-extensoren en de plantair flexoren (kuitspieren) is getest met een handdynamometer (HHD). De dynamische spierkracht van de kuitspieren is getest met een test waarbij de kinderen op 1 been staan en gevraagd worden zo vaak mogelijk los te komen met hun hiel van de grond (standing heel-rise test (SH)). Beide spierkrachttesten werden afgenomen op 2 verschillende dagen. Twintig kinderen met spastische CP, die allen kunnen lopen (3–5 jaar (n=10) en 6–10 jaar (n=10)), zijn meegenomen in dit onderzoek.
ICC (Intraclass Correlation Coefficient) en SDD (Smallest Detectable Difference) werden berekend om daarmee het optimale testschema te vinden om spierkrachtsverschillen te kunnen meten in kinderen met CP. De resultaten van dit onderzoek lieten zien dat het haalbaar was voor jonge kinderen met CP om deze spierkrachttesten uit te voeren en

dat de kinderen de instructies goed begrepen, ook de jongste kinderen in de groep. De ICC-waarden van alle spiergroepen waren boven de 0.77 bij eenmalige testafname, wat een goede betrouwbaarheid aangeeft. Alhoewel de SEM (Standard Error of Measurement) en de SDD's hoog waren bij eenmalige testafname – vooral bij de 3–5-jarige kinderen – hadden de isometrische spierkrachttesten acceptabele SDD's (9–30% verschil te meten bij een herhaalde meting van spierkracht) als we de gemiddelde waarden van 2 à 3 verschillende testdagen en 2 tot 3 repetities nemen per spiergroep.
De SH test had grote SDD's (tenminste 40–128% verschil in aantal repetities moet er aanwezig zijn bij het kind om met behulp van deze test een spierkrachtverschil aan te kunnen tonen voor de 3–5-jarigen en 23–48% voor de 6–10-jarigen). De resultaten van deze studie kunnen gebruikt worden om het individuele testschema te bepalen voor de isometrische spierkrachttesten voor jonge kinderen met CP, afhankelijk van de grootte van de verwachte spierkrachtverandering, de spiergroep en de leeftijd van het kind.
Het kiezen van het meest optimale testschema is een balans tussen de SDD en de haalbaarheid van het herhalen van de testen op verschillende dagen.
De SH testen kunnen alleen gebruikt worden bij jonge kinderen met CP om hele grote verschillen in dynamische spierkracht van de kuitspieren aan te tonen als men het gemiddelde neemt van deze test gemeten op drie verschillende testdagen.

Hoofdstuk 3 beschrijft het onderzoeksprotocol van de studie naar de effectiviteit van de functionele power training op het gebied van spierkracht, loopvaardigheid en door ouders gerapporteerde mobiliteit van jonge kinderen met CP. Dit is uitgewerkt door de veranderingen in spierkracht, loopvaardigheid en mobiliteit binnen de kinderen in een periode van 14 weken functionele power training te vergelijken met 14 weken reguliere zorg voorafgaand aan de trainingsperiode en met een periode van 14 weken volgend op de trainingsperiode. Beoogd werd om 20 kinderen met uni- of bilaterale CP, in de leeftijd van 4–10 jaar, te includeren in deze studie.
Het onderzoeksprotocol geeft een gedetailleerde beschrijving van de nieuw ontwikkelde functionele power training en de meetinstrumenten om de eventuele veranderingen in kaart te brengen.
De power oefeningen in de training worden verzwaard met gewichten en uitgevoerd op 50 tot 70% van de maximale onverzwaarde loop- en sprintsnelheid van de kinderen. Primaire uitkomstmaten zijn sprintcapaciteit (15 meter Muscle Power Sprint Test) en in hoeverre het individuele behandeldoel behaald wordt (gemeten met de Goal Attainment Scaling). Secundaire uitkomstmaten zijn loopsnelheid (1-minuut wandeltest), uithoudingsvermogen (10 meter shuttle run test), grof-motorische vaardigheden, spierkracht van de onderste extremiteiten en de door ouders gerapporteerde mobiliteit van hun kind.

Hoofdstuk 4 en hoofdstuk 5 beschrijven de resultaten van de functionele power trainingsstudie. Veranderingen in spierkracht en loopvaardigheid binnen de deelnemende kinderen over een periode van 14 weken functionele power training werden vergeleken met 14 weken reguliere zorg voorafgaand aan de trainingsperiode en een periode van 14 weken volgend op de trainingsperiode.
Tweeëntwintig kinderen met een gemiddelde leeftijd van 7,5 jaar en een spastische CP (13 bilaterale CP en 9 unilaterale CP) deden mee aan de studie. Veranderingen in alle uitkomstmaten waren significant groter (beter) na de functionele power trainingsperiode dan na de reguliere zorgperiode.

In **hoofdstuk 4** staat beschreven dat grote verbeteringen te zien zijn bij de kinderen in de loopcapaciteit (13–83% verbetering) en in spierkracht (18–128% verbetering), terwijl er in de reguliere zorgperiode bij de kinderen geen veranderingen te zien zijn. Verder zagen we dat de verbeteringen in spierkracht en loopsnelheid werden vastgehouden door de kinderen in de 14 weken na de trainingsperiode. Wel werd er een significante daling van sprintcapaciteit en uithoudingsvermogen gemeten in de 14 weken na de trainingsperiode bij de deelnemende kinderen. Ze hadden echter nog steeds een verbetering van 67% in hun sprintcapaciteit en 33% verbetering van hun uithoudingsvermogen aan het einde van de follow-upperiode in vergelijking met de start van de trainingsperiode.

In **hoofdstuk 5** staat beschreven in hoeverre de behandeldoelen van de kinderen werden behaald. Tevens staan in hoofdstuk 5 de resultaten beschreven van de door ouders gerapporteerde mobiliteit van de kinderen. De behandeldoelen werden behaald en/of overtroffen door 86% van de kinderen tijdens de functionele power training. Dit in tegenstelling tot de periode van de reguliere zorg, de 14 weken voor de trainingsperiode, waarin maar 14% van de kinderen hun behandeldoel behaalde.
Bij de start van de trainingsperiode was 52% van de kinderen afhankelijk van een rollator, rolstoel of ouders bij het verplaatsen over een afstand van 500 meter. Aan het einde van de trainingsperiode was nog maar 13,6% van de kinderen afhankelijk van een loophulpmiddel bij het verplaatsen over een afstand van 500 meter.
Met de mobiliteitsvragenlijst werd een significante verbetering gemeten in mobiliteit in dagelijkse activiteiten na de trainingsperiode vergeleken met de reguliere zorgperiode. De verbeteringen die gevonden werden in mobiliteit na de trainingsperiode, bleven behouden in de 14 weken na training.
De behaalde resultaten van deze trainingsstudie geven aan dat krachttraininginterventies effectief kunnen zijn in het verbeteren van spierkracht én loopvaardigheid mits de krachttraining op hoge snelheid en in functionele bewegingen wordt uitgevoerd met

genoeg intensiteit (maximale inzet van de kinderen, 3 verschillende oefeningen met 6 tot 8 repetities), frequentie (3 keer in de week) en trainingsduur (14 weken).

Hoofdstuk 6 beschrijft de klinische implicaties en de multivariate relaties tussen de veranderingen in kracht van de verschillende spiergroepen met de veranderingen in loopcapaciteit tijdens de reguliere zorgperiode, de functionele power trainingsperiode en de follow-upperiode.
De resultaten van deze analyses lieten zien dat de verandering in spierkracht van de heupabductor sterk gerelateerd is aan de verandering in sprintcapaciteit. Daarnaast was de verandering in spierkracht van de gastrocnemius (lange kuitspier) en de heupabductor van het minst aangedane been gerelateerd aan de verandering in loopduur (uithoudingsvermogen). Als laatste zagen we dat veranderingen in spierkracht van de gastrocnemius van het meest aangedane been gerelateerd is aan verandering in loopsnelheid.
Deze resultaten impliceren dat loopcapaciteit verbeterd kan worden door verbetering van spierkracht van de bovengenoemde spiergroepen met functionele power training bij jonge kinderen met CP. Verder onderzoek is nodig om het werkingsmechanisme van deze veranderingen in spierkracht na de functionele power training op spierweefselniveau en de onderliggende fysiologische en anatomische processen beter te begrijpen.

In **hoofdstuk 7**, de discussie, volgt een uitgebreide beschrijving van de functionele power trainingsstudie en de daarmee samenhangende klinische implicaties. De belangrijkste bevinding is dat met het verhogen van de bewegingssnelheid in de krachttraining en door de krachttraining uit te voeren in een functionele beweging, het mogelijk is om spierkracht, loopcapaciteit en mobiliteit van kinderen met CP te verbeteren.

Kort samengevat: jonge kinderen met CP kunnen hun spierkracht en loopvaardigheid verbeteren door functionele power training. Dit geeft aan dat deze manier van trainen een belangrijke toevoeging is aan de revalidatiebehandeling van jonge kinderen met CP.

Dankwoord

Terugkijkend naar de start van deze reis in de wetenschapswereld kom ik als eerste uit bij de kinderen en hun ouders die ik als kinderfysiotherapeut ben tegengekomen. Specifiek Janna en Vigo. Deze twee mooie persoonlijkheden met prachtige ouders hebben mij echt tot denken gezet. Het stellen van vragen over wat ik aan het doen was in mijn behandeling. Waarom ik deed wat ik deed? Of dit niet beter en efficiënter kon? Dát was de werkelijke start van dit onderzoek.

Bij het opzetten en uitvoeren van het onderzoek vond ik:

» Mensen die de tijd wilden nemen om me te bevragen: Waarom linksom en niet rechtsom? Komt het dan wel goed met..? Hoe zie je deze gedachten in vergelijking met de gedachten van…? Na dit soort gesprekken kon ik met frisse energie weer verder lezen en mijn ideeën verder uitwerken.

» Mensen die in me geloofden en met me mee wilden denken hoe ik mijn ideeën tot uitvoering kon brengen. De ideeën kon toetsen bij de experts in het vakgebied. Is dit inderdaad een wetenschappelijk en maatschappelijk relevante vraag?

» Mensen die me hebben gesteund bij tegenslagen. Mij de energie en kracht gaven om toch door te zetten, risico's te nemen en te geloven in mezelf.

» Mensen die geloof in me hadden om hun kind aan mij toe te vertrouwen in de trainingen en metingen. Ontroerend, vereerd en een heel bijzonder gevoel geeft mij dat, nu nog steeds.

» Mensen die me in balans wisten te houden tijdens deze reis. Zorgden voor afwisseling, rust, nieuwe energie en zo veel meer. Kortom zeker een reis van velen die tot dit resultaat heeft geleid.

Andreas, je bent samen met mij dit avontuur aangegaan en door jouw liefde, creativiteit, energie, geduld en vasthoudendheid is het ook daadwerkelijk tot een eind gekomen. Je praktische en soms dwingende manier om uurtjes, weekenden en weken vrij te plannen zodat we de natuur in konden wandelen, kajakken of fietsen en om naar prachtige muziekstukken te luisteren in het Concertgebouw en Muziekgebouw aan 't IJ waren zo ontzettend nodig en fijn!

Jules Becher, Sonja de Groot, Annet Dallmeijer en Eugene Rameckers, jullie waren als onderzoeksteam altijd erg betrokken. Jullie verschillende expertises en benadering van de wetenschappelijke wereld heeft me heel veel geleerd.

Jules, ondanks je drukke bestaan stond je altijd open om door te praten over ingewikkelde vraagstukken. Deze openheid en bereidheid om mee te willen denken heeft ervoor gezorgd dat we dit zijn gestart.
Sonja en Annet, wat een bewondering heb ik voor jullie kennis, snelheid en gemak waarmee jullie in deze wetenschappelijke wereld leven en bewegen. Sonja, je enthousiasme voor mijn ideeën zorgde ervoor dat ik Annet haar brein kon uitknijpen over mijn gedachten tijdens een mountainbiketocht ergens op de Utrechtse Heuvelrug. Jullie waren bereid tijd vrij te maken voor mij en me dit hele traject te begeleiden met alle ups en downs. Annet, ik ben heel blij voor je dat je tijd vrij hebt gemaakt om een jaar in Chili te verblijven. Wel heel vreemd om dit af te sluiten zonder jouw aanwezigheid.
Eugene, je ervaring als fysiotherapeut met het trainen van kinderen met CP en je bereidheid om mee te denken over de inhoud van de training is een deel van het succes van de functionele power training. Heel fijn om samen te puzzelen hoe we de oefeningen zo konden maken dat we onze ideeën en theorieën erin kwijt konden. En heel leuk om dit samen met jou aan anderen over te kunnen dragen!

Thomas Janssen, Adam Shortland, Jeroen Vermeulen, Vincent de Groot en Olaf Verschuren were the members of the reading committee. Thank you for investing your expertise and time in judging my thesis. Janke de Groot and Annemieke Buizer thank you for being part of my thesis ceremony.

Er zijn twee stichtingen die ik graag hier wil bedanken: de Duyvenzs-Nagel Stichting (DNS) en Stichting Mitialto. DNS heeft er met het uitreiken van de DNS Fellowship voor gezorgd dat ik de eerste stappen durfde te zetten om mijn plannen uit te werken. Arie Prevo, destijds voorzitter van DNS, heel veel dank voor je belangstelling. Waar ik je ook maar tegenkwam wist je je details te herinneren over mijn onderzoek waarop je dan door wist te vragen.
Daarnaast wil ik heel graag de mensen van Stichting Mitialto bedanken voor hun steun en belangstelling voor het project. De financiële steun voor drie jaar was een hele belangrijke en kwam op een cruciaal moment. Dit gaf me het zetje wat ik op dat moment nodig had om echt door te zetten en er vol voor te gaan. De belangstelling voor het project door te komen kijken naar een training bij Reade en door ons uit te nodigen in Eindhoven om over de voortgang te komen vertellen heb ik als heel fijn en bijzonder ervaren.

Arjan Baan, bedankt voor de tijd die je hebt genomen om naar me te luisteren en bedankt voor het willen delen van je mensenkennis en je managementvaardigheden. Toen ik aan dit traject begon was ik vooral bezig met de inhoudelijk kant van mijn onderzoeksvraag. Dat er heel veel organisatie en management bij kwam kijken had ik nog niet bedacht. Jij

hebt me echt de leuke kanten hiervan laten inzien. Je hebt me geleerd om goed te blijven kijken naar het belang van anderen om met mijn ideeën mee te willen gaan en hoe dit strategisch in te kunnen zetten. Heel veel dank voor je vertrouwen in me en het mee willen denken hoe we dingen voor elkaar konden krijgen.

Caroline Doorenbosch, met jou heb ik samen de eerste studentenbegeleiding gedaan voor dit project. Gelukkig hadden we meteen twee talentvolle studenten, Fenna en Mariëtte. Met je enthousiasme, oprechte interesse en gevoel voor humor heb je me de leuke menselijke kanten van de wetenschapswereld laten zien.
Yvette, Maaike, Eline, Janneke en Marloes, mijn kamergenoten op de VU. Dank voor jullie gezelligheid, steun en de zoektochten naar onder andere de lekkerste koffie in de buurt. Marjolein, Marjolein en Laura bedankt voor het meten en uitwerken van gangbeeldgegevens van de kinderen. Wat een werk is dat en wat ben ik blij met jullie expertise.
Eline, collega-kinderfysiotherapeut en collega-onderzoeker, wat fijn dat je ongeveer hetzelfde pad liep als ik. Hierdoor hebben we veel ups en downs kunnen delen en wist ik dat bepaalde onzekerheden erbij hoorden. Ik hoop dat we samen nog mooie dingen gaan doen in onze zoektocht naar nog betere en efficiëntere manieren om kinderen en ouders te kunnen ondersteunen in hun weg naar zelfstandigheid.

Petra, Margje, Esther, Linda, Maartje, Lieseke, Cindy, Danny en alle andere collega's van de De Parel en Drostenburg die zo hard hebben meegewerkt aan deze studie. Heel veel dank voor jullie inzet en bereidheid om hele programma's om te gooien en mee te doen. Elk project heeft zijn "early adapters" nodig.

Nicolette, Tanja, Laura, Janneke en alle andere lieve collega's van het kinderteam. Hier weet ik niet goed waar te beginnen omdat ik zoveel steun van jullie heb gehad. Jullie hebben alles meegemaakt en doorstaan! Bewondering heb ik voor jullie inzet, geduld, nieuwsgierigheid om dit allemaal samen met mij voor elkaar te krijgen. Wat een ongelooflijke passie voor het werk wat we doen. De borreluren na afloop van een werkdag en de weekendjes weg hielpen zeker mee om tegenslagen te verwerken en successen te vieren.

Heel veel studenten hebben hun bijdrage geleverd aan dit werk. Studenten van Bewegingswetenschappen, Geneeskunde, Kinderfysiotherapie, Bewegingstechnologie, Fysiotherapie, Sportkunde en Industrieel Product Ontwerpen.
De vier enthousiaste jongens Alwin, Alwin, Djurre en Simon, die als trainers de kinderen in de MegaPower training tot een hoger niveau wisten te coachen, waren meteen een groot succes. Daarmee was voor mij de twijfel of ik dit wel van deze jonge mensen kon vragen weggenomen en heeft dit me vooral heel veel plezier opgeleverd. Dank jullie allemaal voor

jullie geweldige inzet! Simon, jij was bereid om als onderzoeksassistent aan te blijven na je studie Bewegingswetenschappen. Zonder jouw inzet was dit boek nog niet af geweest. Je goede organisatie- en computervaardigheden heb ik met beide handen aangegrepen.

Ilse en Margriet, jullie zijn mijn paranimfen en ik ben heel blij dat jullie mij ondersteunen bij de verdediging. Ilse, ik ken je al heel lang (Dames '90) en we hebben samen hard geroeid, veel lief en leed gedeeld, en hele mooie reizen gemaakt. Je voelt als familie voor me, iemand waar ik altijd terecht kan en waar ik avonden lang wijntjes mee kan drinken. Margriet, met jou heb ik mooie hoogstandjes bereikt. Ik ben heel blij dat ik jou ben tegengekomen in mijn leven. Samen hebben we lekker getraind, veel gelachen en de laatste jaren vooral veel koffietjes gedronken om weer helemaal bij te kletsen. Je openheid, manier van doorvragen, rust en passie voor het leven is een fijne energie om in de buurt te hebben.

Beste ouders van de MegaPower kinderen, heel veel dank voor jullie inzet, vertrouwen en bereidheid om mee te doen aan dit onderzoek. Gwenne, dank voor al je inzet in dit project. Ik heb er echt van genoten om samen met jou te zoeken naar leuke, spannende verhaallijnen voor de kinderen. Met je creativiteit, enthousiasme en organisatietalent maakte je de prachtigste ijsmachines, geluksdubbeltjes en speurtochten.

Papa en Mama, jullie hebben me geleerd om nieuwsgierig te blijven en jullie hebben me het vertrouwen gegeven dat alles mogelijk is zolang je maar door blijft denken, zoeken en gaan. Nog steeds zijn staan jullie altijd klaar voor me als ik weer met een gek idee of een vraag bij jullie aanklop. Papa heel veel dank voor je bereidheid om mee te denken en om mijn logica te willen begrijpen. Ook heel erg bedankt voor je geweldige knutselwerkstukken die je hebt gemaakt voor dit project en die veelvuldig gebruikt zijn. Mama heel veel dank voor je bereidheid om mee te lezen. Bedankt voor jullie onvoorwaardelijke liefde en vertrouwen!
Mijn zus Anne, Jasper, Lotte en Mees. Door de drukte zien we elkaar niet vaak maar voelt het altijd goed als we elkaar wel zien. Anne, het was leuk dat we door het inzetten van jouw studenten meer contact hadden. De logeerweekenden van Lotte en Mees zorgden voor de nodige afleiding. Als die twee op bezoek kwamen was de keuze makkelijk gemaakt om alle onderzoeksactiviteiten weg te leggen en alleen maar leuke dingen te ondernemen. Ik kijk ernaar uit om Amsterdam weer verder te verkennen met Lotte en Mees.

En dan dank ik nu nog alle stoere helden en heldinnen die zich onvoorwaardelijk hebben ingezet met bloed, zweet en tranen in de MegaPower trainingen van de afgelopen jaren. Dank jullie wel allemaal, wat kunnen jullie ongelooflijk hard rennen!

About the author

Curriculum vitae

Liesbeth van Vulpen was born on Queens Day the 30th of April 1971 in Utrecht, The Netherlands. After finishing secondary school in 1989 at Thorbecke Scholengemeenschap in Zwolle, she attended the Academy of Physical Therapy at the Hanze Hogeschool in Groningen, which she finished in 1995. Liesbeth started her professional life as a physical therapist (PT) working in Sogndal Kommune, Norway for 4 years where her interest in pediatric PT started. In 1999 she moved back to the Netherlands to study Pediatric PT (1999–2002) – Modulaire Opleiding voor Kinderfysiotherapie, Nijmegen, the Netherlands – while working at Children's Rehabilitation Centre De Waarden in Dordrecht, the Netherlands.

In 2000 Amsterdam became her home base and she started to work as a pediatric PT at a private practice, soon combining it with working as a pediatric PT at Reade, Rehabilitation Centre Amsterdam, the Netherlands. Her interest in gait analysis and research grew while working with children with cerebral palsy and she started to work as a research assistant at the Clinical Gait and Movement Laboratory in Amsterdam Rehabilitation Research Institute | Reade, Amsterdam, besides her work as a pediatric PT.

To gain more knowledge on evidence-based medicine and how to get answers on her questions regarding scientific strength training principles in children with cerebral palsy, she started in 2008 with the Master Evidence-Based Practice at the University of Amsterdam. In 2011 she graduated as a clinical epidemiologist (MSc).

In 2010–2011 she won the Duyvensz-Nagel Stichting research Fellowship in Reade, which was the start of her PhD research. She conducted her PhD research on functional power training in children with cerebral palsy at the Amsterdam Rehabilitation Research Institute | Reade in collaboration with the Department of Rehabilitation Medicine, VU University Medical Centre, Amsterdam Movement Sciences, which resulted in the present doctoral dissertation and worked as a pediatric PT at Reade, centre of rehabilitation and rheumatology, Amsterdam, during her PhD research.

Liesbeth is married with Andreas Vlasman.

International publications

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Improved parent-reported mobility and achievement of individual goals on activity and participation level after functional power-training in young children with cerebral palsy: a double-baseline controlled trial. Eur J Phys Rehabil Med 2018 Mar 7 (Epub ahead of print).

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Improved walking capacity and muscle strength after functional power-training in young children with cerebral palsy. Neurorehabil Neural Repair 2017;31:827-41.

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Effectiveness of functional power training on walking ability in young children with cerebral palsy: study protocol of a double-baseline trial. Pediatr Phys Ther 2017;29:275-82.

Van Vulpen LF, De Groot S, Becher JG, De Wolf GS, Dallmeijer AJ. Feasibility and test-retest reliability of measuring lower-limb strength in young children with cerebral palsy. Eur J Phys Rehabil Med 2013 Dec;49(6):803-13.

Presentations

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Effectiveness of functional power-training on individual goals and parent-reported outcomes in young children with Cerebral Palsy: A double baseline controlled trial. Oral presentation at the annual meeting of the American Academy for Cerebral Palsy and Developmental Medicine, 2017, Montreal, Canada.

Van Vulpen LF, Kranendonk D, Verberne S, Rameckers EEA. Functional power training to improve walking capacity in young children with cerebral palsy. A new challenge? Instructional Course at the European Academy of Childhood Disability, 2017, Amsterdam, The Netherlands.

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Effectiveness of functional power training on walking ability in young children with cerebral palsy. Oral presentation at the International Cerebral Palsy Conference and European Academy of Childhood Disability Conference, 2016, Stockholm, Sweden. Dev Med Child Neurol 2016;58(Suppl. 6):46.

Van Vulpen LF, De Groot S, Rameckers EA, Becher JG, Dallmeijer AJ. Effectiviteit van functionele power training op de loopvaardigheid van jonge kinderen met een Cerebrale Parese. Oral presentation at the conference for Nederlands Vereniging voor Kinderfysiotherapeuten, 2015, Utrecht, The Netherlands. Pediatr Phys Ther 2016;28(4): E14-E16.

Van Vulpen LF, De Groot S, Becher JG, Dallmeijer AJ. Muscle Strength is related to the one-minute walk test in young children with cerebral palsy. Poster presentation at international congress Rehabilitation: Mobility, exercise and sports, 2014, Groningen, The Netherlands.

Van Vulpen LF, De Groot S, Becher JG, Dallmeijer AJ. Reliability of measuring lower-limb strength and walking performance in young children with Cerebral Palsy. Poster presentation at the conference of the Nederlands Vereniging voor Kinderfysiotherapeuten, 2012, Zwolle, The Netherlands.

Van Vulpen LF, De Groot S, Becher JG, Dallmeijer AJ. Muscle Strength is related to the one-minute walk test in young children with cerebral palsy. Oral presentation at the 4th International Cerebral Palsy Conference, 2012, Pisa, Italy.

Rameckers EA, Van den Dikkenberg N, Van Vulpen LF, Snijders B, Scheijmans C, Ketelaar M. Methodical steps in task oriented therapy for children with cerebral palsy. Instructional course at the 4th International Cerebral Palsy Conference, 2012, Pisa, Italy.

Van Vulpen LF, De Groot S, Becher JG, Dallmeijer AJ. Reliability of measuring lower-limb strength and walking performance in young children with Cerebral Palsy. Poster presentation at the annual meeting of the Dutch Academy for Childhood Disability, 2011, Nijmegen, The Netherlands.