STRETCH REFLEX THRESHOLD AS A MEASURE OF SPASTICITY OF THE PLANTAR FLEXOR MUSCLES BEFORE AND AFTER TAILORED STRENGTH TRAINING INTERVENTION IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

Sanna Hosio

Master's Thesis in Exercise Physiology

Faculty of Sport and Health Sciences

Autumn 2020

University of Jyväskylä

Supervisors: Heikki Kyröläinen & Taija Juutinen

CONTENT

ABSTRACT

Hosio S. 2020. Stretch reflex threshold as a measure of spasticity of plantar flexor muscles before and after tailored strength training intervention in children and adolescents with cerebral palsy. Faculty of Sport and Health Sciences, University of Jyväskylä, Master's thesis in Exercise Physiology, 48 pp., 2 appendices.

Children and adolescents with cerebral palsy have various symptoms. One of the most disturbing and disabling for locomotion is spasticity. The measurements and treatments have no clear cause-effect standard due to the fact that the definition of spasticity is ambiguous. Strength training is the most effective training method to increase strength and muscle mass. It is also known to affect the nervous system in various ways most often leading to improvements in performance. Stretch reflex excitability is the most commonly used way to measure spasticity.

The purpose of this study was to find out whether the spasticity measured with tonic stretch reflex threshold (TSRT) from the soleus (SOL) and medial gastrocnemius (MG) muscles is affected by strength training intervention in children and adolescents with cerebral palsy (CP). A total of 10 subjects were measured and all of them were included for further analysis. Measurements were done before and after three months strength training intervention. Spasticity was tested with an ankle dynamometer, which induced stretches at four different velocities. Surface EMG was recorded from the SOL and MG muscles.

In statistical analysis Wilcoxon signed-rank test was used to study differences between pre- and post- measurements and to determine whether there were similarities between SOL and MG muscles. Friedman's test was used to study if there were variance with the age and different velocities.

Results support the hypothesis that strength training does not have a negative impact on spasticity in the CP population. In three cases the results showed small changes in TSRT values, but the changes were only ± 1 degrees in the angle. The TSRT measurement produces more reliable outcomes when measuring the stretch reflex from MG. There is no significant difference when using TSRT in different ages, and therefore it can be considered as an appropriate measurement for children and adolescents.

Keywords: cerebral palsy, strength training, spasticity, stretch reflex threshold test

TIIVISTELMÄ

Hosio S. 2020. Stretch reflex threshold as a measure of spasticity of plantar flexor muscles before and after tailored strength training intervention in children and adolescents with cerebral palsy. Liikuntatieteellinen tiedekunta, Jyväskylän yliopisto, liikuntafysiologian pro gradu -tutkielma, 48 s, 2 liitettä.

CP-vammaisilla lapsilla ja nuorilla on useita oireita. Liikkumisen kannalta yksi haittaavin ja invalidisoivin on spastisuus. Spastisuuden määritelmä on epäselvä ja sen vuoksi mittaamisen ja hoitojen valinnassa ei ole yhtä selvää standardia. Voimaharjoittelu on tehokkain harjoitusmenetelmä voiman ja lihasmassan lisäämiseksi. Nykyään tiedetään myös sen useista tavoista vaikuttaa hermostoon ja sen seurauksena suoritusten paranemiseen. Venytysrefleksin herkkyyden mittaaminen on yleisin tapa mitata spastisuutta kun käytetään Lancen (1980) määritelmää spastisuudesta.

Tämän tutkimuksen tarkoituksena oli selvittää, onko voimaharjoitteluinterventiolla vaikutusta spastisuuteen CP-vammaisilla lapsilla ja nuorilla, kun mittarina käytetään toonisen venytysrefleksin kynnystä (TSRT) soleuksesta ja mediaalisesta gastrocnemiuksesta. Yhteensä 10 koehenkilöä osallistui mittauksiin ja kaikkien heidän tulokset analysoitiin. Mittaukset tehtiin ennen ja kolmen kuukauden voimaharjoitteluintervention jälkeen. Spastisuutta testattiin nilkan dynamometrilla, jossa venytykset tehtiin neljällä eri nopeudella. EMG-vasteet mitattiin soleuksesta ja mediaalisesta gastrocnemiuksesta.

Tilastollisessa analyysissä käytettiin Wilcoxon signed-rank testiä tutkimaan, oliko tuloksissa eroja ennen interventiota ja sen jälkeen, ja lisäksi oliko muutokset lihasten välillä yhtäläisiä. Friedmanin testiä käytettiin tarkastelemaan, oliko tuloksissa vaihtelua eri-ikäisillä tai eri nopeuksilla.

Tulokset tukevat hypoteesia, ettei voimaharjoittelulla ole negatiivisia vaikutuksia spastisuuteen CP-vammaisilla lapsilla ja nuorilla. Kolmessa tapauksista oli pieni muutos TSRT-mittausten välillä, mutta kulman muutos oli vain ± 1 astetta. TSRT mittaus näyttää tuottavan luotettavampia tuloksia, kun venytysrefleksiä mitataan MG:sta. Iällä ei ole merkittävää vaikutusta TSRT -mittauksen tuloksiin ja siksi sen voidaan katsoa olevan sopiva menetelmä lapsille ja nuorille.

Avainsanat: CP-vamma, voimaharjoittelu, spastisuus, stretch reflex threshold test

1 INTRODUCTION

Children with cerebral palsy (CP) experience various impaired muscle functions in their daily life activities, such as reduced selective motor control, spasticity and muscle weakness (Bax et al. 2005). These impairments often lead to difficulty with certain aspects of walking, such as walking speed and endurance or stair climbing. Because of the increasing interest in studying muscle strength management in these children (Dodd et al. 2003, Lee et al. 2007, Liao et al. 2007), it has been suggested that muscle weakness has a strong association with mobility limitations (Ross et Engsberg 2007). Progressive resistance exercise training is a well-established strength training method, in which the intensity progresses with time. This stimulates strength gains that are greater than those associated with normal growth and development. (Faigenbaum 2009.)

In the current research, it appears that strengthening programs, in general, may be effective in increasing strength in people with CP. However, because of the methodologic limitations of the literature, it is still impossible to make definitive recommendations. (Dodd et al. 2002.) The assumptions about the benefits of strength training to people with spasticity have been argued and previously it was seen more in a negative aspect. In recent studies, it has been shown repeatedly that resistance training does not increase spasticity and it has lots of possible positive outcomes for people with neurological diseases, including muscle strength and locomotion. (Kirk et al. 2016.)

Spasticity is a common impairment observed after neurological lesions. Spasticity was defined by Lance in 1980, but in literature, research field and clinically the definition has wider aspects and it includes different variations about muscle stiffness. From a research and treatment point of view, the lack of a precise definition of spasticity interferes to compare studies of spasticity and outcome after treatment interventions. (Gjelsvik & Syre, 2106. 83) Therefore it is recommended to differentiate pathophysiology which causes hypertonia and stiffness in

neurological patients. It is the basis for better treatment and quality of life. (Gjelsvik & Syre 2016, van den Noort et al. 2017.)

Spasticity has been measured by using different kinds of scales like the Modified Ashworth or Tardieu Scale but their reliability and validity are not good (Fleuren et al. 2009, Levin and Calota 2008, van den Noort et al. 2017). The tonic stretch reflex threshold (TSRT) is a new method that is developed to measure spasticity. It is in agreement with the traditional definition of spasticity proposed by Lance and finds its basis in motor control theory. (Levin and Calota, 2008.)

The aim of this study is to measure if there is an association between tailored strength training and spasticity in children and young CP population. In previous studies the strength training has had lots of variance with intensity and quantity and also a measurement that is used has been some of the scales with poor validity and reliability. Therefore this is the first study when strength training protocol is tailored and based on certain recommendations, and the measurement that is used is in agreement with the definition of spasticity.

2 CEREBRAL PALSY

Cerebral Palsy (CP) is caused by brain injury or brain malformation that occurs before, during, or immediately after birth while the infant's brain is under development. How the brain injury affects a child's motor functioning and intellectual abilities is highly dependent on the nature of a brain injury, where the damage occurs, and how severe it is. A child's muscle control, muscle coordination, muscle tone, reflex, posture and balance can be affected. It can also impact a child's fine motor skills, gross motor skills, and oral motor functioning. (Bax et al. 2005, Paulson & Vargus-Adams, 2017) The Gross Motor Function Classification System (GMFCS) is a 5 level clinical classification system that describes the gross motor function of people with cerebral palsy on the basis of self-initiated movement abilities (figure 1.).

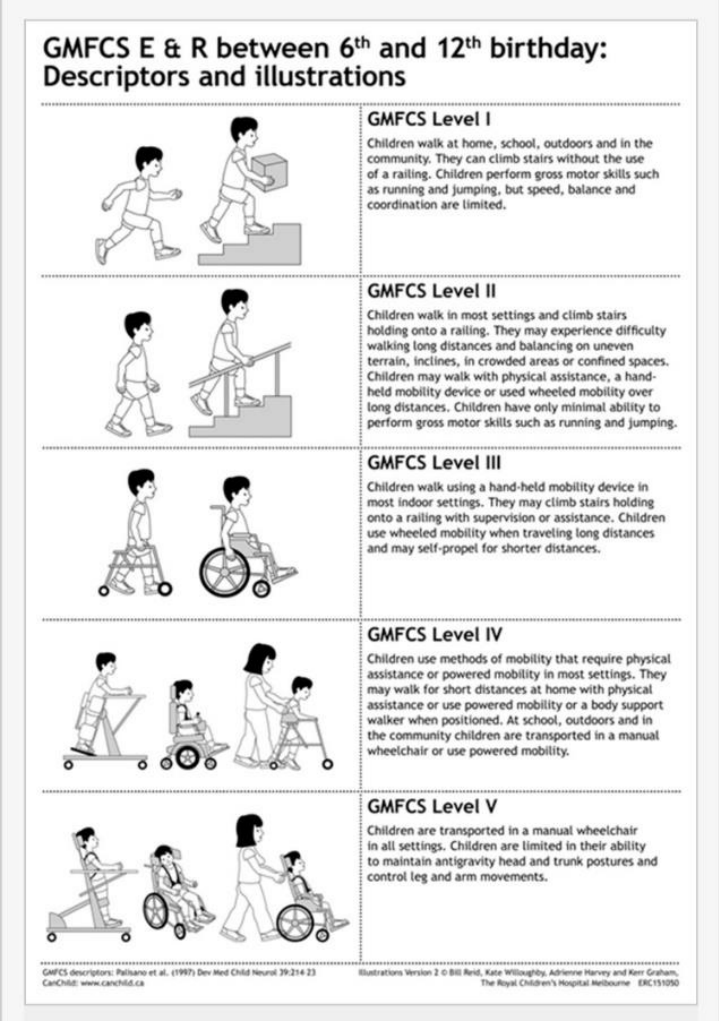


FIGURE 1. Gross Motor Function Classification Scale, GMFCS scale. (Paulson & Vargus-Adams, 2017)

Cerebral palsy (CP) is the most frequent cause of motor disability among children in Europe representing 700 000 citizens. The prevalence of CP in Europe has been stable over the last 30 years and ranges between 1.5 and 3.0 cases per 1000 live births. In Finland, according to the Finnish CP Association, 100-120 children per year have diagnosed cerebral palsy and it is the most common disorder among children. Approximately 30% of CP children have hemiplegia, 50-60% of all CP patients have diplegia affecting mostly the lower limbs and 10-15% tetraplegia. In 2004, there were estimated economic costs for the families and government in the US, and the average loss is around one million per child with CP. (CDC 2004.)

The motor disorders of individuals with CP are complex (Armand et al. 2016, Ryan et al. 2017). They are related to primary deficits such as muscle spasticity, muscle weakness and loss of selective motor control, and secondary deficits such as muscle contractures and bony deformities. The main dysfunctions are related to motor disorders during posture and movement causing limitations in daily life activities and locomotion (e.g. walking, figure 2). Around 75% of CP children are ambulatory. (Armand et al. 2016.)

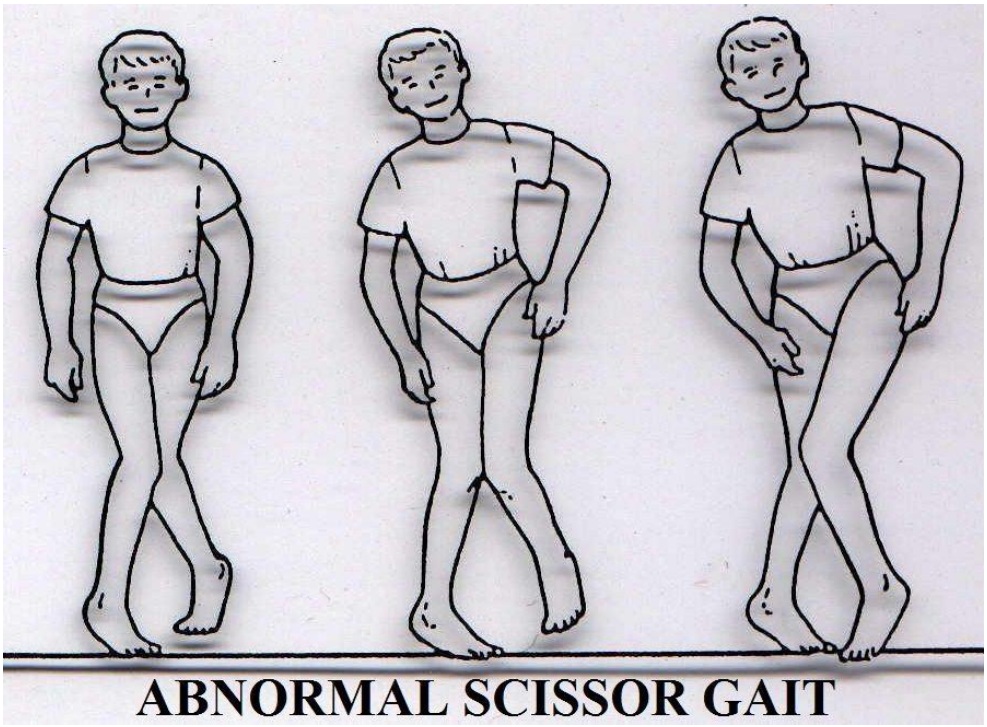


FIGURE 2. Typical gait pattern in spastic diplegia among CP children and adults. (https://www.abclawcenters.com/practice-areas/types-of-birth-injuries/cerebral-palsy/birth-injuries-and-spastic-cerebral-palsy/what-is-spastic-diplegia/)

Walking is essential for multiple activities. This is why it is often considered to be one of the most important activities in daily life (Armand et al. 2016). Challenges in gait might become a major problem to integrate with other children, participate in social activities and hence reduce the quality of life (Willerslev-Olsen et al. 2014.)

3 SPASTICITY

The term spasticity is frequently used differently in practical fields and research. Most researchers use the operational definition proposed by Lance (1980), but clinicians use the term spasticity more broadly. (Grey et al. 2008.) Lance defined (1980) spasticity as a motor disorder characterized by a velocity-dependent increase in tonic stretch reflexes with exaggerated tendon jerks, resulting from hyperexcitability of the stretch reflex, as one component of the upper motor neuron syndrome (Silva et al. 2017, Lance 1980). Burke (1988) and Brown (1994) have described it as a syndrome, developed as a result of the changes in plasticity in the CNS after an injury. In literature, it is written that the incidence of spasticity depends on the cause of the upper motor neuron (UMN) lesion (Brashear & Elovic 2011). It is a common disorder followed by UMN lesions like stroke, spinal cord injury (SCI), traumatic brain injury, or cerebral palsy (CP), and multiple sclerosis (Santos et al. 2017). Spasticity affects approximately 20% of people with stroke, 47-70% of those with multiple sclerosis (MS), 34% with spinal cord injury, 50% of patients with traumatic brain injury, and more than 90% with CP (Lundy-Ekman, 2013). Gracies et al. 2005 (figure 3.) have shown how the adaptation and maladaptation occur in the CNS after lesion. In the practical field, it is important to separate different kinds of stiffness, hypertonia, contractures, and also spasticity, and clarify the underlying mechanisms and pathophysiology of the symptoms. It is also important to notify when the damage has occurred, because of the development of the CNS.

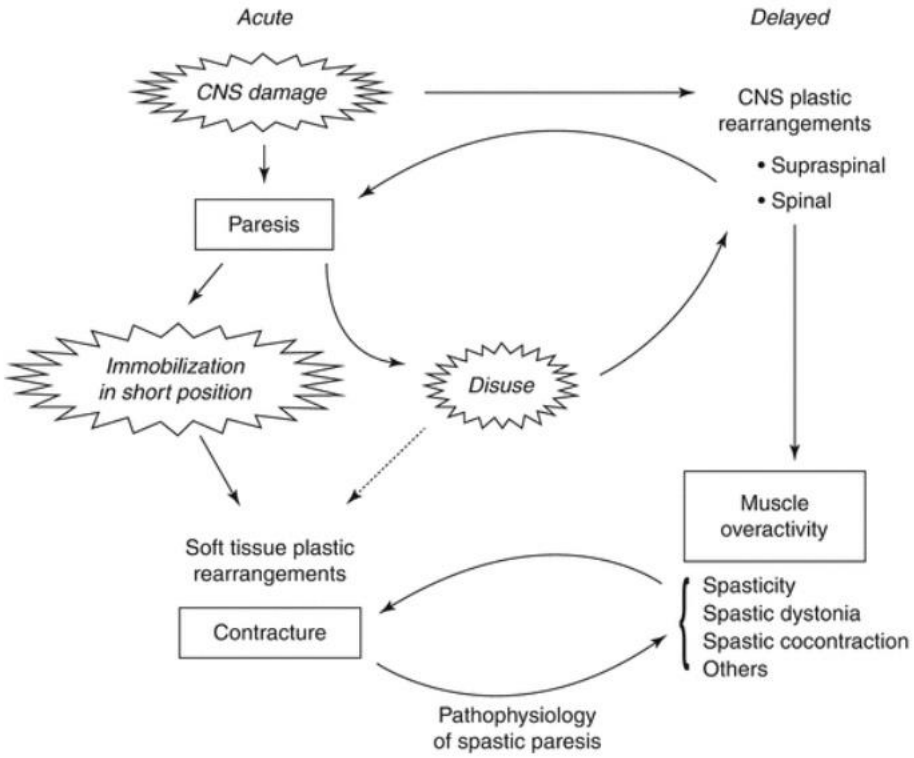


FIGURE 3. Reprinted from "Pathophysiology of spastic paresis II. Emergence of muscle overactivity", by Gracies 2005. *Muscle Nerve 31: 552-571*. The primary immediate symptom after lesion is paresis. Delayed symptoms after lesions are caused by adaptations in supraspinal and spinal plasticity. Without supraspinal inhibitory mechanisms, muscle overactivity might increase muscle contracture and enhances responses to stretch and further aggravates spastic overactivity.

Because the term spasticity is inconsistently defined and there is still no adequate definition for this phenomenon (Malhotra et al. 2009.), it is often related to neuromuscular overactivity secondary to an UMN (upper motor neuron) lesion with an overactive neural input causing excessive contraction to muscles (Lundy-Ekman, 2013). Physical manifestations of spasticity include involuntary contractions, abnormal postures and pain. In addition to functional limitations, spasticity when inappropriately treated may lead to reduced quality of life, increased pain and joint contractures (Francisco & McGuire, 2012, van den Noort et al. 2017).

The term spasticity has been increasingly used to refer to several features of the upper motor neuron syndrome. In addition to muscle hypertonus, spasticity following spinal cord injury (SCI) could also involve hyperreflexia, clonus, clasp-knife responses, long-lasting cutaneous reflexes, and muscle spasms evoked by brief nonnoxious cutaneous stimuli for example. (Elbasiouny et al. 2010.) Even today, there are gaps regarding the complete understanding of the physiopathology of spasticity (Santos et al. 2017).

According to the recently published consensus of European clinicians the term 'spasticity' should only be used next to stretch hyperreflexia (mostly in the SCI population), and other symptoms that cause 'stiffness' next to passive tissue contributions. When joint angle, moment, and electromyography are recorded, components of hyper-resistance within the framework of spasticity can be quantitatively assessed. (van den Noort et al. 2017.) Spasticity is more often found in the flexor muscles of the fingers, wrist and elbow, and the extensor muscles of the ankle and knee. Although there might be some exceptions. (Trompetto et al. 2014.) Even though there is a lack of consensus about the definition, there are also misunderstandings about treatments. The underlying pathophysiology might be different and not shared with all spasticity diagnosis. (Gjelsvik & Syre 2016, 83-86.)

3.1 Pathophysiology of spasticity

The spasticity is commonly defined as hyperexcitability of the stretch reflex responses, which are velocity-dependent. It can be identified as an increasing resistance with the increase of the joint passive rotation or as a rapid rise of the resistance above a threshold speed or joint angle value (Germanotta et al. 2017). There is also a general agreement that spasticity is caused by adaptations at the spinal cord level distal to the lesion. (Yang et al. 2014)

The consensus is that its characterization by muscle tone changes due to the exacerbation of deep myotendinous reflex caused by the increase in the speed of stretch reflex response. (Santos et al 2017.) The static stretch reflex has been attributed to secondary muscle spindle endings; primary endings of the muscle spindle are known to have dynamic or velocity sensitivity (Brashear & Elovic, 2011).

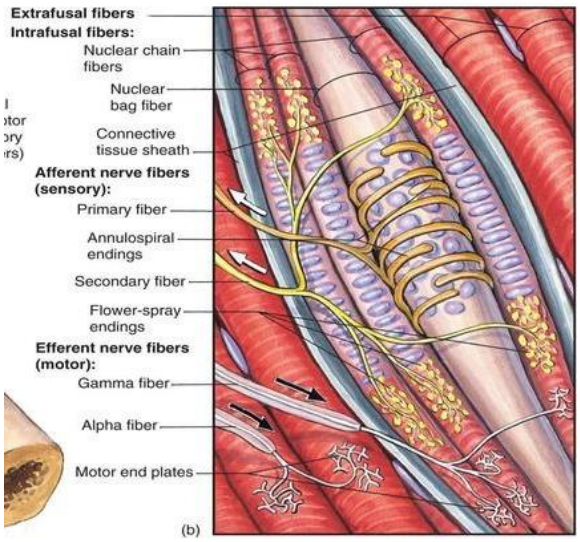


FIGURE 4. The muscle spindle sends sensory information through both group Ia and group II afferent fibers. The 1a fibers (primary afferents) wrap around the chain and bag fibers. These afferents increase their firing rate when there is a change in muscle length or rapid movement. The group II afferent fibers (secondary afferents) wrap about the chain and static bag fibers. These afferents produce a sustained response to constant muscle length and therefore provide information about the static muscle position. (Kandel et al. 2013, 794-795)

A critical assessment of experimental findings indicates that increased excitability of both motoneurons and interneurons in the spinal cord plays a crucial role in the pathophysiology of spasticity. Because hypertonia is usually a secondary outcome from lesions in the pyramidal and/or extrapyramidal pathways its pathophysiology may vary depending on the site or place of the lesion but commonly develops in the antigravity muscles (Elbasiouny, 2010.)

The central lesion, like stroke, hypoxia, or traumatic brain injury can cause the upper motor neuron syndrome (UMNS). This leads to disturbances in the balance of supraspinal inhibitory and excitatory inputs directed to the spinal cord and a state of disinhibition of the stretch reflex. (Trompetto et al. 2014.)

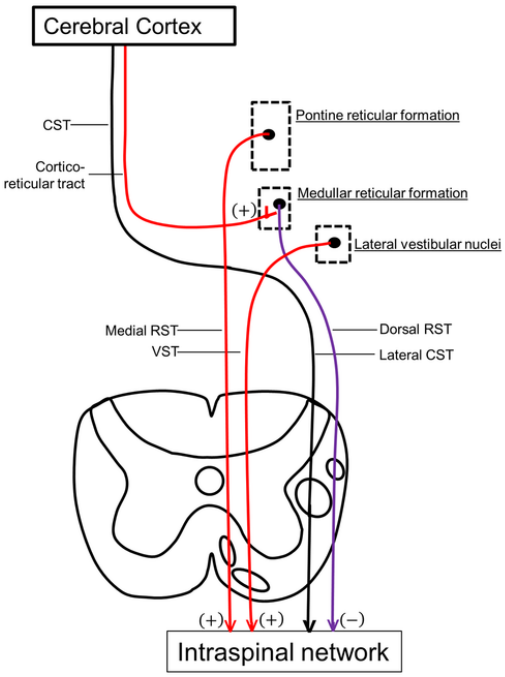


FIGURE 5. Illustration of supraspinal control of spinal stretch reflex (Sheng 2015).

In the early phase of recovery muscle strength and motor function is mainly attributed to cortical plastic reorganization. Reticulospinal (RS) hyperexcitability as a result of maladaptive plasticity (figure 1), is the most plausible mechanism for spasticity after stroke. (Sheng, 2017.) The early belief was that spasticity is the result of injury to the corticospinal tract. Today, we know that spasticity is not caused by injury to the corticospinal tracts, but rather closely related to corticoreticulospinal and vestibulospinal tract injuries. (Sheean 2002, Gjelsvik & Syre 2016, 85.)

3.1.2 The monosynaptic reflex

The stretch reflex is considered as an involuntary reflex that is essential in postural control and movement (Shemmell et al. 2010). Functioning of the stretch reflex loop and the state of the motoneuron pool is usually studied with H-reflex which is muscle's response to artificially produced stretch. H-reflex is elicited with electrical stimulus to the afferent nerve. When the given stimulus is strong enough, it will cause an action potential in the motoneuron and a muscle response can be measured. (Enoka 2008, 257, Kandel et al. 2013, 808-809.) The H-reflex can be used to assess the excitability of spinal alpha-motoneurons, while also reflecting

transmission efficiency (i.e., presynaptic inhibition) in Ia afferent synapses (Aagaard et al. 2002).

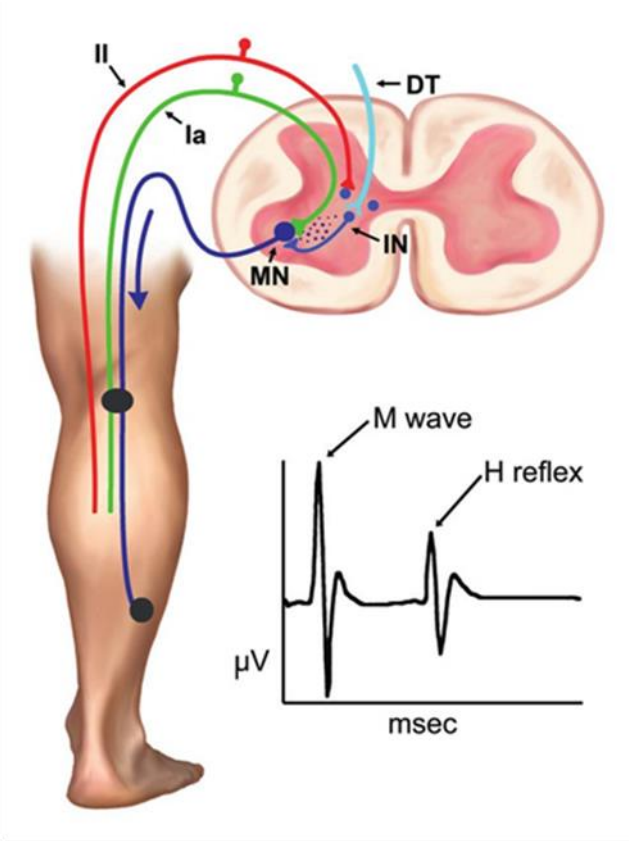


FIGURE 6. The H-reflex pathway. DT= descending tract, IN= inhibitory interneuron, MN= motoneuron, Ia= group Ia afferent fibers, II= group II afferent fibers, small dots in the medial ventral spinal cord represent Renshaw cells. Stimulation of the posterior tibial nerve at the popliteal fossa below motor threshold results in excitation of Ia afferents and activates motor neuron in spinal cord which is then recorded with latency from soleus as H-reflex. M-wave is an initial response for stimulus in muscle (Yates et al. 2010, Knikou et al. 2008).

In typically developed people stretch reflexes are mediated by excitatory connections between Ia afferent fibers from muscle spindles and α-motoneurons innervating the same muscles from which they arise. A passive stretch of the muscle excites the muscle spindles, causing Ia fibers to discharge and send inputs to the α-motoneurons through mainly monosynaptic, but also oligosynaptic pathways. The α-motoneurons in turn send an efferent impulse to the muscle, causing it to contract. In typically developed people the surface EMG shows that when a resting muscle is stretched with the velocities used in the clinical practice to assess muscle tone, it

doesn't produce any reflex contraction (Trompetto et al. 2014.), i.e. reflex responses are velocity-dependent.

3.1.2 Stretch reflex pathway

One of the clinical conditions related to spasticity is excess reflex activity. Changes in structure and function of the CNS may affect via different mechanisms in stretch reflex networks. It might change the excitability of motor neurons, reduce 1a presynaptic inhibition, change the inhibitory mechanisms from the efferent pathway, alter reciprocal inhibition, reduce recurrent inhibition and also increase the strength of withdrawal reflexes by increasing the flexor reflex pathways. The changes in CNS can also increase receptor sensitivity and neural drive from muscle spindles, causing more easily excited stretch reflex threshold. (Gjelsvik & Syre 2016, 84. Knikou et al. 2008.)

3.2 Spasticity measurements

Spasticity can be classified as an impairment based on the International Classification of Functioning, Disability and Health (ICF). The available measurements for assessing spasticity can be classified as neurophysiological or biomechanical measures. (Malhotra et al. 2009.) The defining characteristic of clinical spasticity is excessive resistance of muscle to passive stretch, a resistance that intensifies as the examiner increases the rate of stretch in subsequent stretch maneuvers. Slow stretch may offer modest resistance, but fast stretch can result in a suddenly intensified resistance that may even catch the examiner off guard. Increased EMG activity of the stretched muscle accompanies different clinical rates of stretch. (Brashear & Elovic, 2011.)

Tonic stretch reflexes, on the other hand, respond to the level of a stimulus and lead to sustained muscle contraction for the duration of the stimulus. For example, a passive stretch of a muscle, that is, progressive lengthening of the muscle, generates a change in the level of the stimulus (muscle length), and in spastic patients, once the threshold of excitation is reached, sustained reflex contraction is produced until a stretch is released. (Brashear & Elovic, 2011.)

The velocity of stretch, when using machines, does not simulate what clinicians experience because clinicians, unlike machines, cannot maintain a constant stretch velocity when spastic tension runs high. (Brashear & Elovic, 2011.) The evidence seems to favor a reduction in the threshold of the tonic stretch reflex, namely, less afferent input is necessary to trigger stretch reflex activity in the presence of an UMN lesion. Mechanisms of reduced presynaptic and postsynaptic inhibition and impaired recurrent inhibition in the Renshaw system are likely to contribute to the handling of afferent input by a reorganized spinal cord (Brashear & Elovic, 2011.)

3.2.1 Biomechanical approach

When approaching via biomechanics, joint position, and angular speed, reactive/resistive torque quantifies muscle behavior especially during movements with controlled speed. Isokinetic devices and speed control motors measure the stretch reflex resistance during passive movements. The advantage is that these devices allow the standardization and control of velocities and amplitudes while evoking the stretch reflex. (Santos et al. 2017.) Biomechanical measurements of spasticity using a servo-controlled motor-driven device can provide a more objective measure of resistance to passive stretch. (Francisco & Byrne 2012.)

3.2.2 Neurophysiological approach

Electrophysiological tests, such as the H-reflex, and H/M ratio, have been used to quantify spasticity but tend to correlate poorly with the degree of spasticity. (Francisco & Byrne 2012.) The neurophysiological approach for assessing spasticity investigates the neuromuscular electrical activity of agonist, antagonist, or both muscles. It is used during active or passive movements acquired by surface electromyography (EMG). This mechanism of evaluation involves the excitability analysis of motoneurons and tendon reflexes, observed by signs of EMG. (Santos et al. 2017.)

It has been suggested that reduced depression of transmitter release from 1a afferents following previous activation (post-activation depression, PAD) is involved in the pathophysiology of spasticity. Originally PAD was described by Eccles and Curtis (1960), who noticed that the size of 1a excitatory postsynaptic potentials (EPSPs) in intracellular recordings was frequency-

dependent, with relative facilitation at short intervals (<50ms) and with depression at longer intervals (>1s). (Grey et al. 2008)

3.2.3 Clinical scales

Modified Ashworth Scale (Appendix 1.) is the most commonly used clinical scale to estimate spasticity (Blanchette et al. 2016) but nowadays when awareness in the research field has increased it should be called hypertonia what is measured with MAS (Lundy-Ekman, 2013, Van den Noort et al. 2017). Some features make these scales attractive for clinicians, such as their ease of use in clinical settings, without specific equipment, and the short time needed to administer them. The original Ashworth Scale is a subjective 5-point ordinal scale for grading resistance felt by the evaluator during passive stretching. To render the Ashworth Scale more sensitive, few modifications were proposed. (Blanchette et al. 2016.)

The validity of the Ashworth Scales is still questionable despite all of the attempts to improve them (Blanchette et al. 2015, Brashear & Elovic 2011, Kim et al. 2011). It measures resistance to passive movement, which is one feature of spasticity (Blanchette et al. 2015). These scales assess the perceived resistance to passive movement, which is influenced not only by reflex responses in the stretched muscle but also by changes in passive resistance of non-contractile and contractile properties, as well as changes in the resistance of the shortening antagonist muscle. (Dietz & Sinkjaer, 2017.) The fact that non-reflex components are not discriminated from reflex components compromises the construct validity of Ashworth Scales. From critical point of view, scores on Ashworth Scales do not take into account the velocity dependence of the response. Because Ashworth Scales measure the perceived resistance to passive movement, which varies with the velocity of the stretch, the failure to control stretch velocity negatively affects the reliability of these scales. (Blanchette et al. 2016.)

The Tardieu Scale has been suggested as a more valid alternative for spasticity assessment. Unlike Ashworth Scales, the Tardieu Scale takes into consideration the velocity-dependent aspect of the stretch reflex response. The most recent versions of the Tardieu Scale compare the passive resistance when the muscle is stretched at a slow velocity, and when the muscle is

stretched at a fast velocity. The maximal angle reached during a slow stretch and the angle at which a catch or clonus is felt during a fast stretch are determined. The latter angle is then subtracted from the former angle, and the resulting value reflects the dynamic, or velocity-dependent, a component of spasticity. (Blanchette et al. 2016.)

The resistance perceived during stretching also is characterized by a subjective 6-point ordinal scale. Although the construct assessed by the Tardieu Scale is more closely related to Lance's definition of spasticity, this scale also focuses on the perceived resistance to passive movement, which is a consequence of spasticity and may not reflect the neurological origin of spasticity. There is a consensus among researchers that neurophysiological measures should be at least part of a spasticity evaluation given that they provide information about the pathways that are altered in spasticity. (Blanchette et al. 2016.)

These two scales mentioned above are mainly used in the field. There are still lots of different scales used among clinicians. Although, their validity is poor and leads to misunderstandings about the phenomena. In summary, it is good to be aware of these scales used by clinicians. Lots of research has been done about their reliability and validity. If we consider these scales and the definition of spasticity by Lance (1980), there is a conflict about what is measured. With these scales it is possible to evaluate hypertonia or stiffness, but not spasticity. (Van Noort et al.2017.)

3.2.4 Tonic Stretch Reflex Threshold Test (TSRT)

The tonic stretch reflex threshold (TSRT) evaluates the excitability of the motoneurons resulting from supraspinal and segmental influences. It represents the joint angle at which the motoneurons and their muscles of the joint are recruited and when the muscle is contracting during stretches with different velocities. The stretches are produced by a dynamometer and the muscle activity is measured by EMG from the surface of the target muscles. (Calota et al. 2008, Marques et al. 2019.)

Levin and Feldman (1994) proposed a method to quantify spasticity based on the indirect measurement, called tonic stretch reflex threshold (TSRT). This measure is in agreement with the traditional definition of spasticity proposed by Lance and finds its basis in the motor control theory. Regulation of this threshold is essentially related to the supraspinal control and the excitability of the reflex activity. TSRT was proposed as a tool to evaluate spasticity in stroke and cerebral palsy populations (Silva et al. 2017.)

The TSRT has very good inter-evaluator reliability and is a physiologically valid measurement of spasticity (Blanchette et al. 2016). The TSRT identifies the minimal joint angle at which abnormal motoneuronal recruitment begins when the muscle is at rest and there is no motion. The theoretical construct of the TSRT has been validated as a measure of spasticity in the upper limb muscles of patients with chronic stroke. In recognition of the clinical reality, TSRT testing has been implemented in a portable device requiring a minimal amount of equipment (Montreal Spasticity Measure). (Blanchette et al. 2016.)

The correct EMG onset detection is fundamental for the method because it determines the values of angle and velocity that correspond to the dynamic DSRT's. If there is "basal activity" with a patient's muscles, it can give false results in EMG activity while measuring TSRT. (Nakagawa et al. 2014.) The TSRT index seems to be the more promising approach (Calota and Levin 2008, Mullick et al. 2012), as it more accurately reflects Lance's definition of spasticity than other clinical tests (Phadke et al., 2015).

TSRT has been evaluated with stroke (Calota et al. 2008, Calota & Levin 2009) and CP (Jobin & Levin, 2000) populations, and the results were showing moderate to good values of reliability in both cases. (Germanotta et al. 2017.) The intraclass correlation coefficient (ICC) for all participants during visits 1 and 2 was 0.73 ($p<0.001$), indicating a good test-retest reliability of the measure (Jobin & Levin, 2000). They found moderately good reliability for subjects with moderate to high spasticity (intra-evaluator: 0.46-0.68, and inter-evaluator: 0.53-0.68) (Calota et al. 2009) and high inter-evaluator reliability (ICC = 0.85) for plantar flexor spasticity in adult post-stroke patients (Blanchette et al 2016).

3.3 Spasticity in cerebral palsy

Muscle spasticity is characterized by exaggerated stretch reflexes and affects about 85% of the children with cerebral palsy (van der Krogt et al 2010). In spastic CP the lesion affects the corticospinal (CS) and corticobrainstem tracts during the perinatal period, interfering with the development of the spinal cord and brain. In a normally developed nervous system, a single CS axon synapses with a spinal lower motor neuron (LMN), and in the early years, it activates agonists, antagonists and synergists. Through normal development of the locomotor system, the weaker synapses are eliminated and by the age of 4 years, the CS axon will synapse only the agonists. With CP population and damages in the CS tracts during development, it causes some elimination of competition for synaptic sites during a critical period. It causes the persistence of inappropriate connections and abnormal development of spinal motor centers. The persistence of inappropriate connections causes abnormal cocontractions, the simultaneous activation of antagonists that interferes with muscle activation, and performing the task. (Lundy-Ekman, 2013.)

3.4 Rehabilitation and spasticity treatments in CP

CP patients may present many multiple combinations of impairments and they can be managed with different kinds of treatments. Physiotherapy is the most common treatment in CP and it might include variations of different kinds of methods to maintain range of motion and adequate muscle length, maintain and restore strength, and to improve balance and coordination. (Armand et al. 2016.)

For spasticity management, there is a large spectrum of therapies that can have effects locally or globally and permanent or reversible. Botulinum is locally used for spasms and it should be reversible. Nowadays, there is still a lack of knowledge about the long-term implications of botulinum concerning denervation atrophy. Researchers recommend being cautious with botulinum, especially with kids, who are walking. (Armand et al. 2016.)

Selective dorsal rhizotomy (SDR) has a permanent effect and more globally impacting spasticity in lower limbs. It is a complex neurosurgical procedure, where abnormal rootlets in

the spinal cord are removed. (Armand et al. 2016., Enslin J.M.N et al. 2019, Lundy-Ekman 2013.) There are also other treatments for spasticity e.g. medication (baclofen), orthoses, and musculoskeletal surgeries which in some cases can prevent further symptoms and restore functional ability (Armand et al. 2016).

It has concluded in a systematic review (Katalinic, et al. 2010, Pin et al. 2006) that regular stretching does not produce clinically important changes in spasticity or activity limitation in people with neurological disorders. The coexistence of paresis and overactivity in a voluntary drive may confuse clinicians, in particular as to treat one dysfunction (overactivity) in specific muscles, some treatment strategies may include partial but deliberate worsening of the other (weakness), for example, the use of injections of neuromuscular blocking agents. (Gracies 2005.)

4 STRENGTH TRAINING

It is already well acknowledged that resistance training is the most effective training method to increase strength and muscle mass. It enhances overall physical function and the quality of life. It is also nowadays considered as a training method not only for athletes but also recommended for a variety of populations including people of different ages. (Walker & Häkkinen 2014.) The enhancement of motor performance caused by strength training is associated with both muscular and neural adaptations. These especially include an increased neural drive to the agonist muscles and a decreased co-activation of antagonist muscles. (Barrue-Belou et al. 2016, Seger & Thorstensson 2005.) Gains in children's and adolescents' strengths are primarily attributed to the neurologic mechanism of increases in motor neuron recruitment. It allows increases in strength without resultant muscle hypertrophy. (Stricker et al. 2020)

4.1 Neural adaptations

The human nervous system is highly adaptive in response to training. Neural adaptations occur in response to strength training and are thought to contribute to enhanced motor performance. Although the mechanisms underlying these adaptations have been tried to elucidate, the results are ambiguous. (Vila-Chã et. al. 2012.) In normally developed people neural control of force production is reliant upon motor unit recruitment and firing rate whose combination determines the final signal presented to the muscle. Many regulators influence both/either the recruitment and firing rate of motor units. Afferent feedback through sensory tracts from muscle contraction influences forthcoming signals at the cortex and spinal cord. At the spinal level, there are wide networks of interneurons to excite or inhibit the afferent information coming from muscle spindles. Also, Renshaw cells via supraspinal control, act as an inhibitor to the feedback system that responds to activation of the α -motoneuron. These regulators allow the neural system to continuously monitor and modulate force production. (Walker S. 2018.) It is suggested, that the increase in motoneuronal output induced by resistance training may comprise both supraspinal and spinal adaptation mechanisms (i.e., increased central motor drive, elevated motoneuron excitability, reduced presynaptic inhibition) (Aagaard et al. 2002).

4.2 Muscular adaptations

Strength training leads to increases in muscle size and strength in the muscle which is trained. Muscle architecture is also a primary determinant of muscle function. The muscular adaptations have differences in the CP population and typically developing (TD) individuals. The differences in the CP population include reduced muscle volume, reduced or similar length of muscle fascicles, increased intramuscular fat, and increased tendon length e.g. achilles. From a microscopic view, there has been found longer sarcomeres and different extracellular matrix composition differences in the CP population. These changes have already started occurring in the first five years since birth and they are likely to contribute to reduced muscle strength and power compared to TD individuals throughout the lifespan. (Gillett et al. 2016.)

Spastic cocontraction refers to inappropriate antagonist recruitment triggered by the volitional command on an agonist in the absence of phasic stretch. Spastic cocontraction primarily results from an abnormal pattern of supraspinal descending drive, which can be aggravated by abnormal peripheral reflex reactions. The evidence for abnormal antagonist cocontraction during active movement in spastic paresis is pretty overwhelming. Some cocontraction (i.e., simultaneous activity in both agonist and antagonist) is common also during normal human movement, but in spastic paresis, it is present to an excessive degree. Cocontraction during any given task in healthy subjects often decreases with training. (Gracies 2005.)

Physiologically has been found to cocontraction to be activated and deactivated at a cortical level. Although cocontraction can be a normal mechanism to provide joint stability under particular circumstances, cocontraction in the UMNS refers to inappropriate antagonist activation that blunts or even reverses the agonist-driven movement. (Brashear & Elovic, 2011.) Cocontraction becomes excessive in the upper motor neuron syndrome, as measured EMG activity in agonist and antagonist while movement. In the stroke population with spastic hemiplegia, excessive cocontraction of plantar flexors has been shown to often limit voluntary ankle dorsiflexor torque beyond 90 degrees of dorsiflexion. Placing the knee in extension, which stretches the gastrocnemius muscles, aggravates these plantarflexor cocontractions. (Gracies 2005.) Increased antagonist coactivation can contribute to the measured deficits in force

production in CP. Increased antagonist coactivation in children with CP can occur during ambulation and standing balance. (Stackhouse et al. 2005.)

4.3 Strength training in CP patient population

Muscle weakness is a common impairment in children with cerebral palsy. Weakness has been attributed to incomplete recruitment or decreased motor unit discharge rates, inappropriate coactivation of antagonist muscle groups, secondary myopathy, and altered muscle physiology (Stackhouse et al 2005). Some findings have been noticed in CP-related weakness and the causes may lie within the morphology of single muscle fibers and whole muscle. The most common findings are an increased incidence of muscle-fiber atrophy, increased intramuscular fat and connective tissue in the most involved muscle groups, and an increase in the percentage of histochemically identified type I muscle fibers, all of which may contribute to weakness. (Stackhouse et al 2005.)

Training can increase strength and may improve motor activity in people with CP without adverse effects (Dodd et al. 2002, Gillett et al. 2018). Kim & Park (2014) reviewed that strengthening interventions are useful for increasing muscle strength in individuals with CP, especially with children and youth. The optimal exercise intervention should perform three times per week and 40-50 minutes/session. Even if the evidence has shown lots of positive outcomes about strength training in CP populations (Dodd et al. 2002, Gillett et al. 2018), there is still lack of knowledge how it affects the neural adaptations in spinal and supraspinal level and to motor activity and functional capacity. Spasticity and stretch reflex excitability describes the properties and function of stretch reflex pathways in spinal cord level. This study concentrates on possible changes in spasticity after strength training intervention.

5 PURPOSE OF THE STUDY AND STUDY QUESTIONS

The purpose of the present study is to evaluate effects of the tailored strength training to stretch reflex excitability and/or hyperreflexia (spasticity) on the calf muscles in people with spastic cerebral palsy and if there are differences between the investigated muscles (SOL and MG). In addition, it was interesting to see if the variance between different ages and velocities occurs in TSRT.

1. Does strength training have effects in calf muscles (SOL and MG) spasticity in the CP population?

Hypothesis: Strength training has minimal or no effects on spasticity in the CP population.

Background theory: Strength training is not effective in children and adolescents with cerebral palsy (Scianni et al. 2009). Progressive resistance training improves muscle strength in young people with CP (Damiano 2014). It is possible to increase muscle strength in adults with CP through progressive resistance training, but it does not increase spasticity. (Kirk et. al 2016.)

2. Is the change in spasticity (TSRT) similar in SOL and MG in the CP population after strength training intervention?

Hypothesis: Strength training affects differently to spasticity in MG and SOL because of the difference in neural pathways.

Background theory: The training induces more neuromuscular changes in a voluntarily active gastrocnemius. Soleus is more active in postural control and activated from subcortical areas, while gastrocnemius is more active in voluntary contractions and activated cortically via corticospinal pathways (Mochizuki et al. 2006).

3. Do DSRT values have more variance in different ages?

Hypothesis: The age of the subjects varied from 9-21 years and it was expected that in the younger population, there is more variance between stretches at the same velocities than older ones because of more immature supraspinal inhibition strategies.

Background theory: Objective measures are becoming prominent in spasticity assessment, to overcome limitations of clinical scales. Previous studies have demonstrated the validity and reliability of TSRT in spasticity assessment in adults (Germanotta et al. 2017). The TSRT technique is a potential outcome variable for measuring the efficacy of treatments aimed at decreasing spasticity in children with CP (Jobin & Levin 2000).

6 METHODS

6.1 Subjects

This study included 10 CP male or female who were 9-21 years old. They had diagnosed of spastic hemiplegia or diplegia-type CP and were classified as Gross Motor Function Classification System level I to III.

TABLE 1. Subject description. GMFCS, gross motor function classification scale (see figure 1.)

N	AGE	GMFCS (n)	GENDER (n)
3	<10y	1 (2) 3 (1)	M (3)
3	10-15y	1 (3)	M (2) F (1)
4	>16y	1 (2) 3 (2)	M (4)

The participants were not included if they: a) have had lower limb surgery and/or pharmacological treatments (e.g. intrathecal baclofen, botulinum toxin) in the past six months; b) have had selective dorsal rhizotomy; c) are utilizing serial casting on the lower limbs; d) have participated in a resistance training program for the lower limbs in the last six months; e) is unable to provide sufficient cooperation in the intervention and/or testing sessions. CP population was measured before the intervention (pre 1 and pre 2) and after (post 1).

6.2. Study design

This study was based on a multiple baseline design. It was composed of pre -test and the post-test immediately before and after the intervention (Figure 1). The multiple baseline design solves some ethical problems concerning RCT studies and enables possibilities to each participant to compare with himself at different time points (i.e. repeated measures).

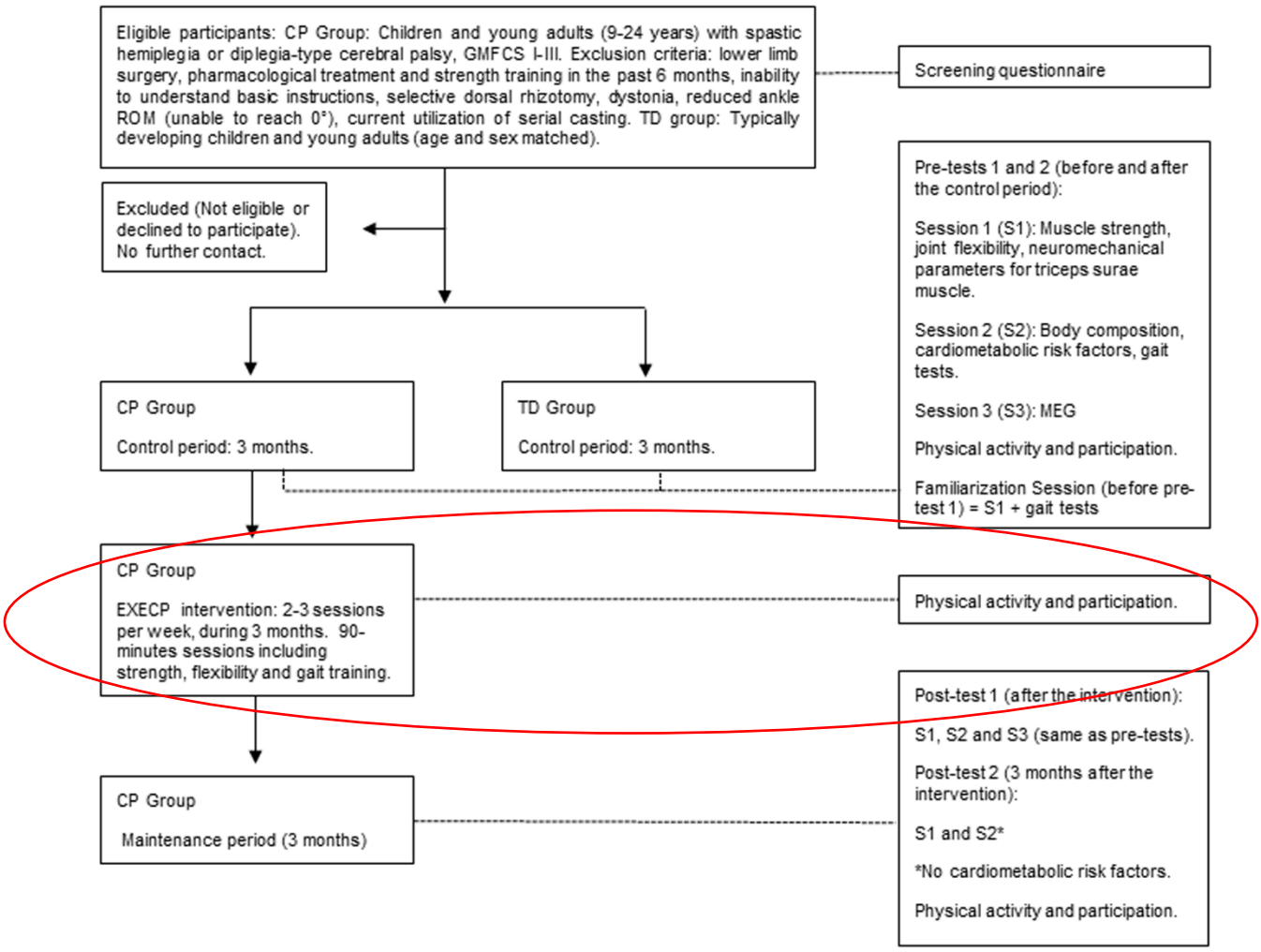


FIGURE 7. Flowchart for the whole EXECP study, the red circle includes the Pre2 and Post1 measurements and intervention. CP = cerebral palsy; TD = typically developing; GMFCS = gross motor function classification system; ROM = range of motion; MEG = magnetoencephalography (Valadao Pedro et al. 2019, unpublished material.)

Legal guardians have signed the informed consent for children under 18 years, and written assent from children was also required. The study was done according to the declaration of Helsinki recommendations and it was approved by the ethics committee of the Central Hospital of Jyväskylä (ethical approval received on April 13, 2017; Dnro 8U/2017).

6.3 The training protocol

The EXECP intervention took place at the Neuromuscular Research Center, University of Jyväskylä or in other gyms near the subject's environment. It consisted of two to three training sessions per week for 12 weeks. The number of weekly sessions varied on the level of physical activity (PA) of each participant: those engaged in regular weekly PA chose to perform 2 or 3 sessions, while sedentary participants trained 3 times per week. Training sessions lasted 90 minutes and were separated by at least 48 hours. Ten exercises were selected for the intervention, five single joint and five multi-joint (Appendix 1.). To ensure optimum training quality all sessions were performed individually and supervised by a strength and conditioning coach or a physiotherapist with full understanding of the training protocol (Appendix 2.).

In EXECP project the training load was devised to increase muscle strength and mass, complying with the American College of Sports Medicine and National Strength and Conditioning Association guidelines (Garber et al. 2011; Faigenbaum et al. 2009) and CP specific guidelines (Verschuren et al. 2016). The intervention was divided in 3 blocks of 4 weeks (TABLE 2).

TABLE 2. Training description for the intervention (Pedro Valadao 2019, Unpublished material)

Week	Volume	Repetitions	Movement Duration (s)	Rest (s)	Session A*	Session B*
1 - 4	3 sets of 8 repetitions	8 RM	3 concentric 3 eccentric	60	1 – Seated calf raise 2 – Seated dorsiflexion	1 – Seated machine knee flexion 2 – Seated machine knee extension
5 - 8	3 sets of 8 repetitions	8 RM	1 concentric 3 eccentric	90	3 – Standing calf raise 4 – Hip flexion	3 – Hip flexion 4 – Standing calf raise
9 - 12	4 sets of 6 repetitions	6 RM.	! concentric 2 eccentric	90	5 – Seated horizontal leg press 6 – Roman chair trunk extension 7 – Squat	5 – Seated horizontal leg press 6 – Isometric hollow rocks 7 – Squat

! = ballistic muscle action. * = each session has a minimum of 7 exercises and a maximum of 10 (i.e. all exercises).

6.4 Data collection

Data was collected before (PRE2) and after intervention (POST1). Spasticity of the plantar flexors was measured with the TSRT (Blanchette et al. 2016, Calota & Levin 2009). The participants were seated with the knee joint fully extended and the ankle dynamometer induced stretches (from 20° of plantarflexion to 0°) on the triceps surae muscle at different velocities: 50, 100, 200 and 300°/s (figure 8).

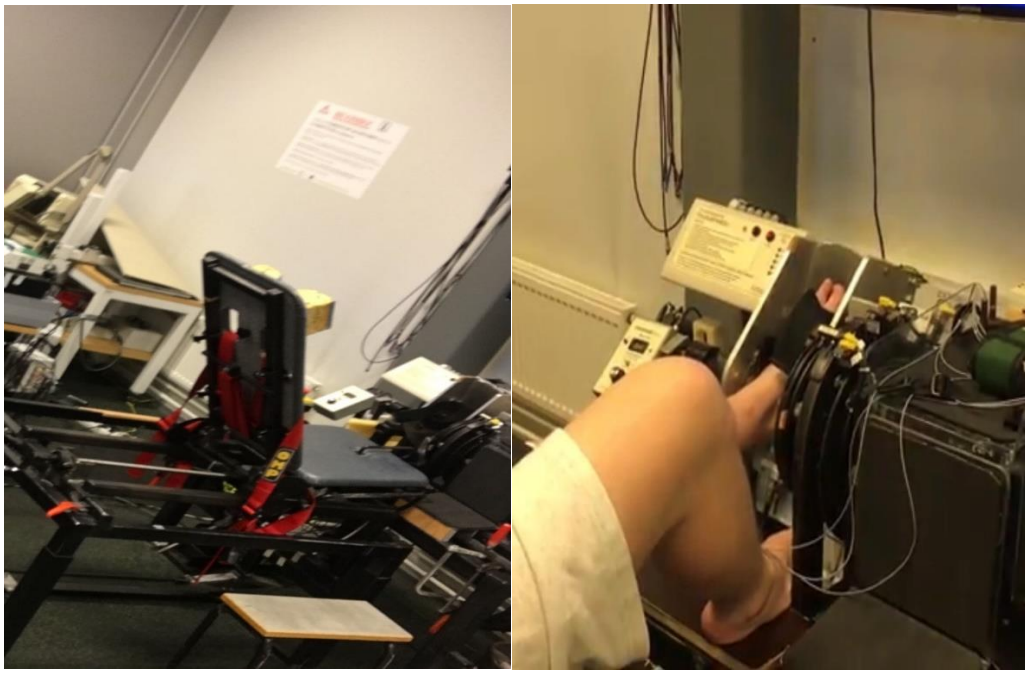


FIGURE 8. TSRT - The seat and dynamometer in Biomechanics Lab, University of Jyväskylä.

The stretch velocities, consisting of 10 stretches at each velocity, were delivered randomly. EMG data were used to determine the joint angle, in which stretch reflex onset of soleus and/or medial gastrocnemius occurred (i.e. dynamic stretch reflex threshold, DSRT). The participants were instructed to relax and they were wearing noise-blocking headphones. TSRT was measured on the most affected limb of the CP group and in the same leg of the age/sex-matched control. The control group, which was matched by age and gender was used to ensure the reliability and repeatability of the TSRT test in the EXECP project, and to inhibit systematic bias (Piiparinen M. 2020).

EMG activity was recorded from the soleus (SOL), medial gastrocnemius (MG), and TA muscles with self-adhesive electrodes (Blue Sensor N, Ag/AgCl, 0.28 cm2; Ambu, Ballerup, Denmark), and a ground electrode placed on the tibia. Spike 6.10. Ink was used for analyzing and doing some bandpass filtering (few resting values) EMG and the EMG onset from stretches at different velocities. Because of the inability to finish proper signal processing to EMG data, it was decided to include stretches that had 10% or less of MVC maximal values in the analysis. This was done to guarantee that the muscle was at rest before stretching. An interval between stretching was 2.6-2.9 s. Noraxon, a portable device, was used in a few cases to record the EMG signals. The data, what was recorded with Noraxon, had to subtract because of the delay in processing.

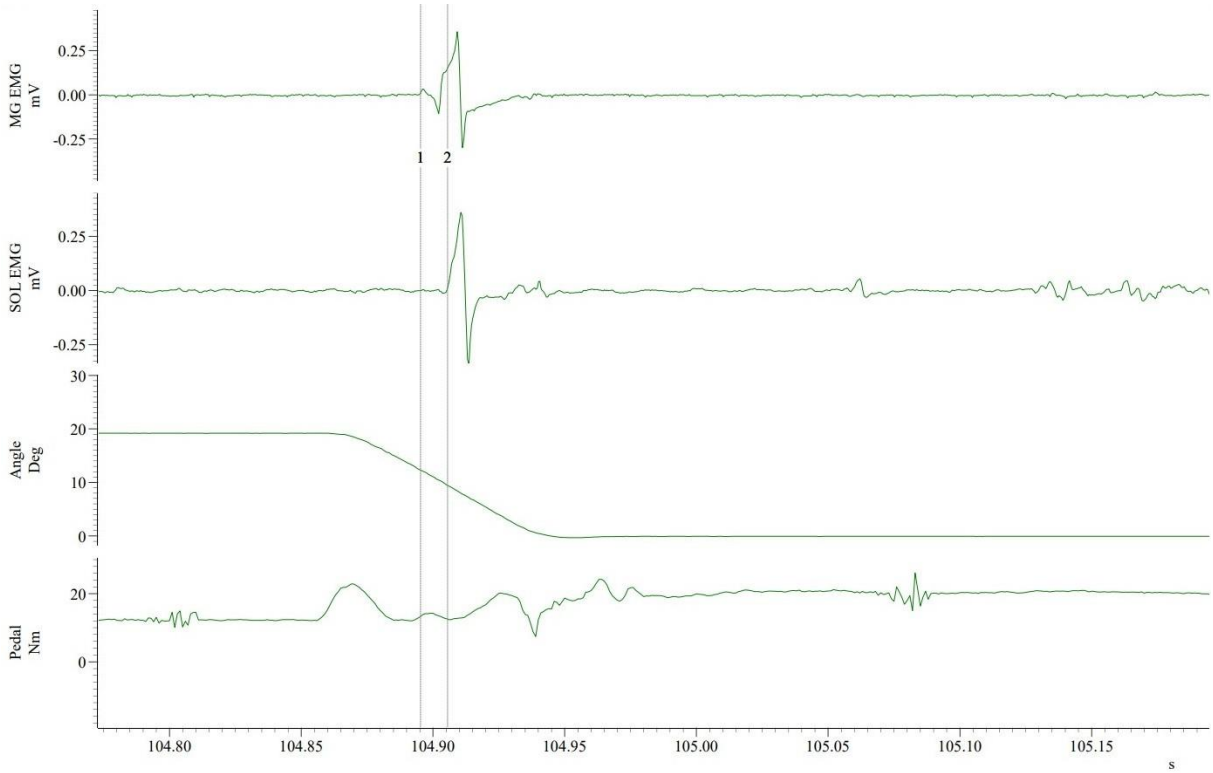


FIGURE 9. TSRT data collection in Spike2 6.10. Ink. (screenshot). MG=medial gastrocnemius, SOL=soleus, Angle=stretch angle (from 19.8 to 0), the cursor (1) is the onset of the EMG activity in the MG muscle and (2) is the onset of the EMG activity in the SOL muscle.

6.5 Statistical analysis

Statistical analysis were done with SPSS and excel was used for graphics. Wilcoxon signed-rank test was used to study differences between pre- and post- measurements and if there were similarities between these two muscles. It is a nonparametric test and its goal is to determine if two or more sets of pairs are different from one another in a statistically significant manner. Friedman's test was used to study if there were variance with the age and different velocities.

A regression line based on a first-order linear equation using all DSRT's was calculated on a stretch velocity-joint angle plot. From the equation, the coefficient of determination, slope, and intercept with the x-axis was attained. The TSRT is the angle at which the regression line intersects with the x-axis, low values indicate a high level of spasticity. The correlation coefficient (r) was used to evaluate the quality of the linear regression. The absolute value of r was interpreted, in agreement to (Dancey and Reidy, 2007), and this interpretation was also used in Germanotta's (2017) study. There is no correlation, if $r \leq 0.1$, mild/modest correlation, if $0.1 < r \leq 0.3$, moderate correlation, if $0.3 < r \leq 0.6$, strong correlation, if $0.6 < r < 1$ and perfect correlation, if $r = 1$ (Table 3.).

7 RESULTS

The results are presented and compared individually. Some results are also presented in group comparison. All the available TSRT values are collected together in table 3. Figure 10 presents an example of how the regression line is formed from the DSRT values. Also, the equations are presented in the figure, where the intercept of the x-axis is calculated and the TSRT angle is solved. The correlation coefficient, R^2, was used to evaluate the quality of the linear regression.

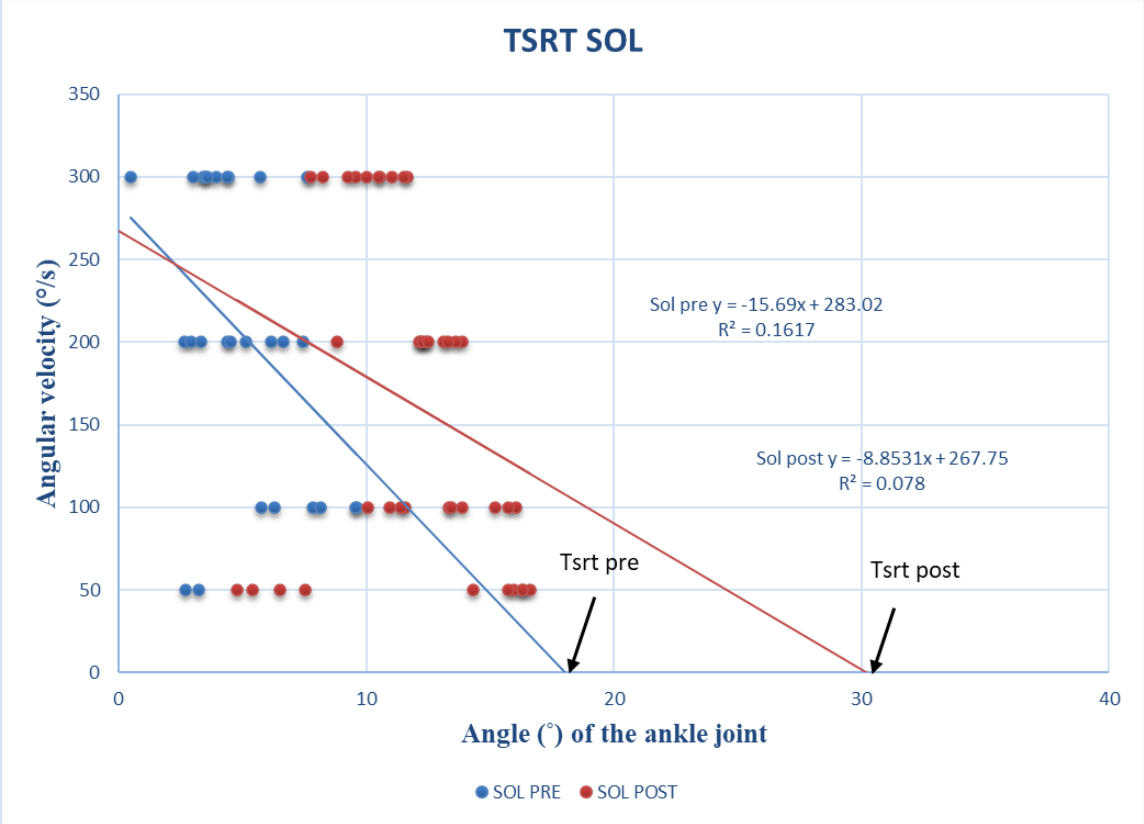


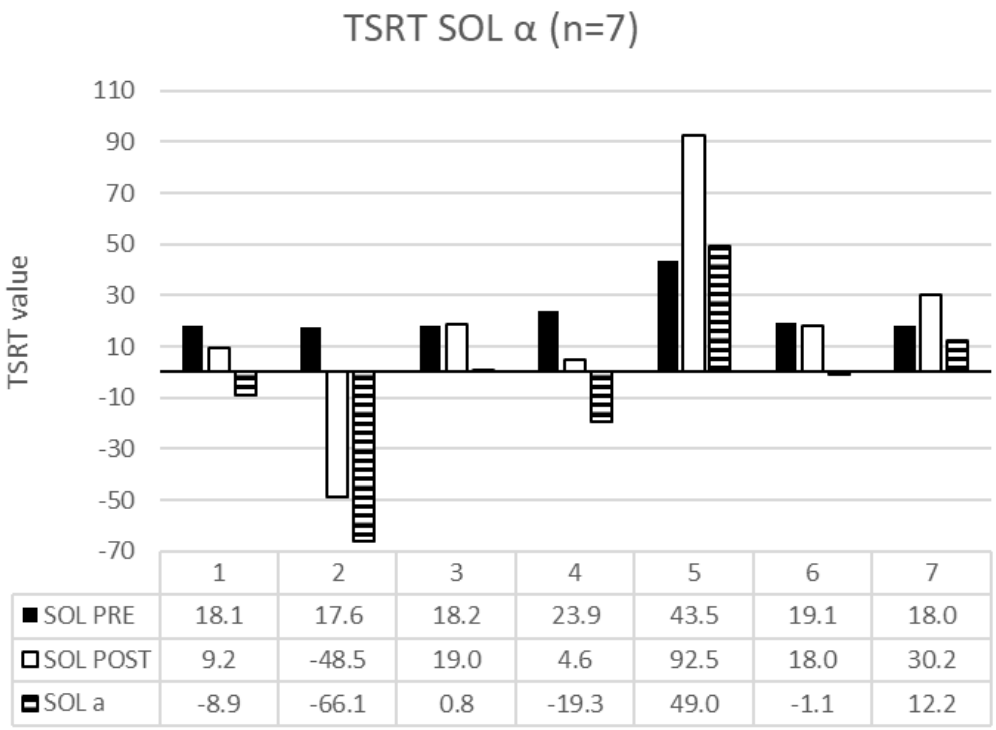
FIGURE 10. An example of one subjects SOL dynamic stretch reflex threshold (DSRT) of the soleus (SOL) values and their regression lines and equations. The tonic stretch reflex threshold (TSRT) is the angle at which the regression line intersects with the x-axis. These equations were solved for x when y = 0. The R^2 values for linear regression show poor or no correlation coefficient ($R^2 < 0.3$)

TABLE 3. TSRT values and correlation coefficient (R^2) for each participant. The R^2 values present the quality of the linear regression; no correlation mild/modest correlation, moderate correlation, strong correlation. * TSRT values with strong correlation.

	SOLEUS		SOLEUS		MG		MG	
PARTICIPANTS	PRE		POST		PRE		POST	
	TSRT	r	TSRT	r	TSRT	r	TSRT	r
1	18.12	0.09	9.23	0.79	23.61	0.33	18.15	0.95
2	17.55	0.58	-48.50	0.04	18.57	0.99	19.02	0.99
3	18.20	0.98	18.97	0.79	18.91	0.92	18.50	0.99
4	23.93	0.18	4.59	0.33	17.19	0.91	20.97	0.17
5	43.50	0.03	92.50	0.04			19.90	0.15
6	19.08	0.36	17.99	0.83			17.85	0.88
7	18.03	0.16	30.24	0.08			18.17	0.52
8	18.71	0.80			18.65	0.71	29.57	0.15
9	29.75	0.09			24.35	0.33		
10			9.75	0.41	20.54	0.48	11.00	0.36
Avg	22.99	0.36	16.85	0.41	20.26	0.67	19.24	0.57
Std	8.67	0.35	38.61	0.35	2.73	0.29	4.78	0.38

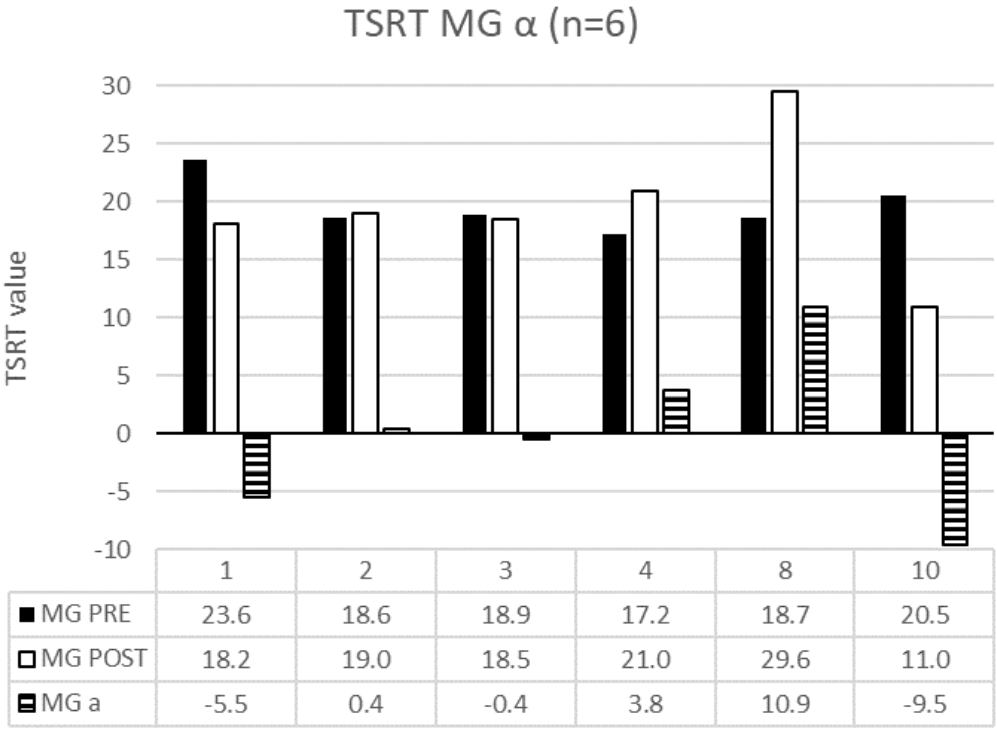
There are both positive and negative changes in the TSRT values (Table 3.). When comparing the changes with the Wilcoxon Signed Rank test, it shows no significant change in SOL (0.499, n=7) and MG (0.917, n=6) when the significance level is 0.05. Only three measures got strong correlation coefficients for the quality of the linear regression and their changes in the TSRT values were small (±1). These three values are marked with * (Table 3).

In figures 11 a and b, the changes between these two muscles (SOL and MG) before and after training in the TSRT values were compared. It shows lots of variation in the results, but regarding the results with the values where correlation is good, only one subject was observed. It shows a small difference between SOL and MG (figure 12 a and b). The other participants were not compared because of only a mild/modest correlation of the linear regression and therefore the TSRT values are not reliable.



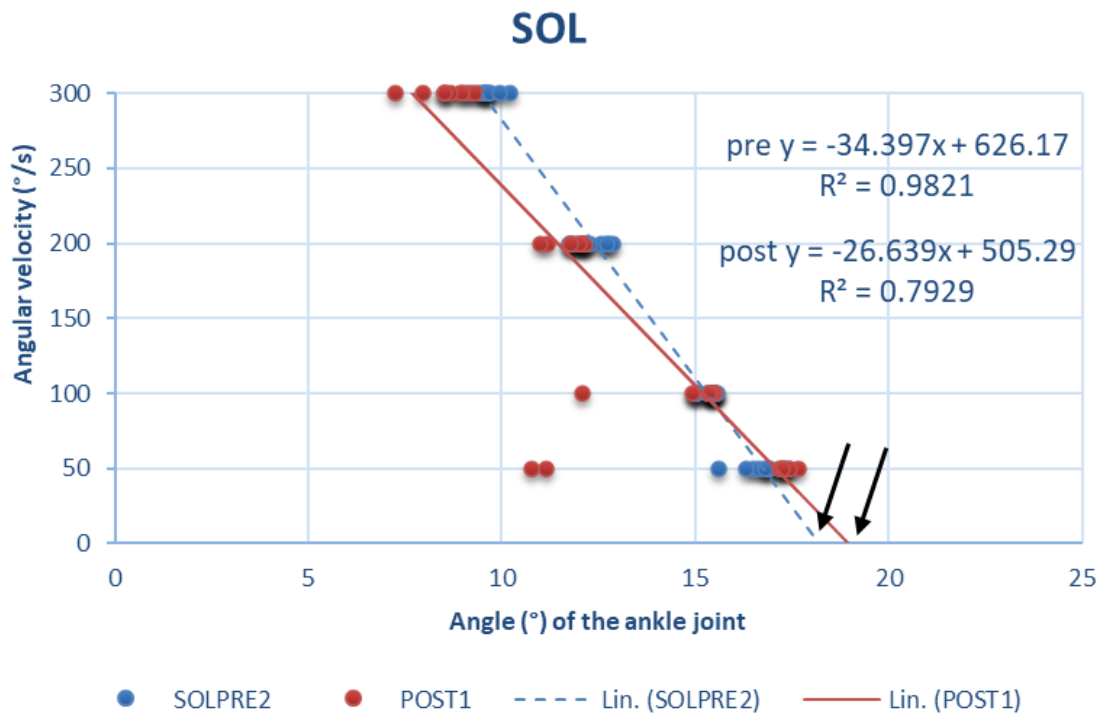
	1	2	3	4	5	6	7
SOL PRE	18.1	17.6	18.2	23.9	43.5	19.1	18.0
SOL POST	9.2	-48.5	19.0	4.6	92.5	18.0	30.2
SOL a	-8.9	-66.1	0.8	-19.3	49.0	-1.1	12.2

FIGURE 11A. TSRT values in SOL before and after the intervention and the change (dashed bars) in 7 participants. If the change in TSRT value is negative, it means spasticity is increased.



	1	2	3	4	8	10
MG PRE	23.6	18.6	18.9	17.2	18.7	20.5
MG POST	18.2	19.0	18.5	21.0	29.6	11.0
MG a	-5.5	0.4	-0.4	3.8	10.9	-9.5

FIGURE 11B. TSRT values in MG before and after the intervention and the change (dashed bars) in 6 participants. If the change in TSRT value is negative, it means spasticity is increased.



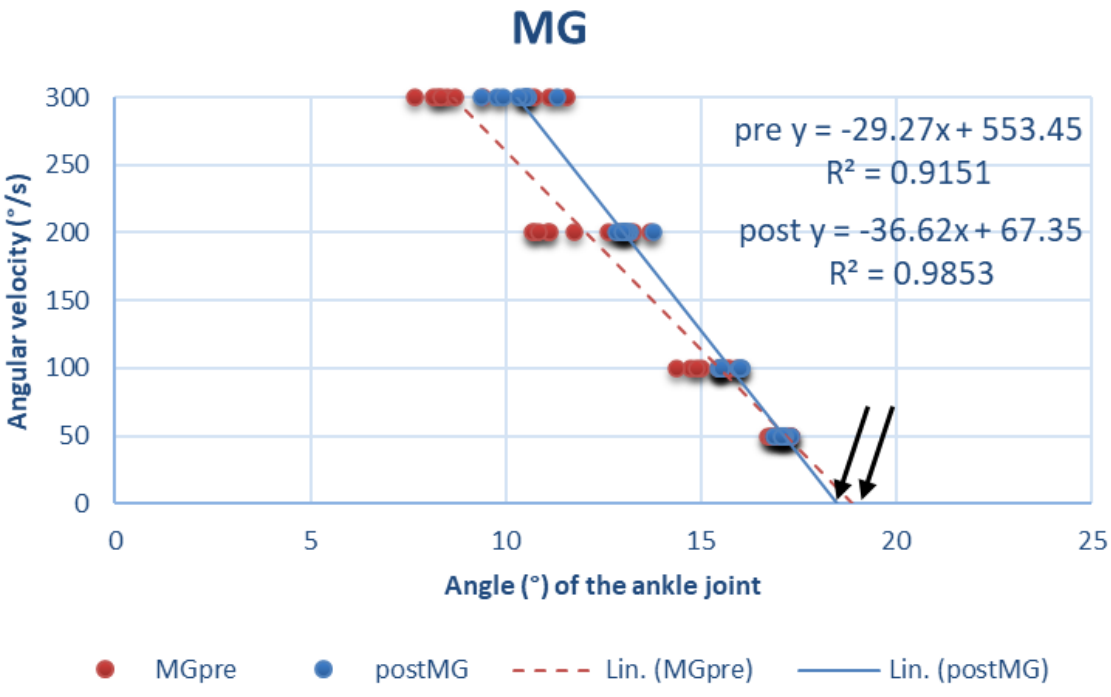
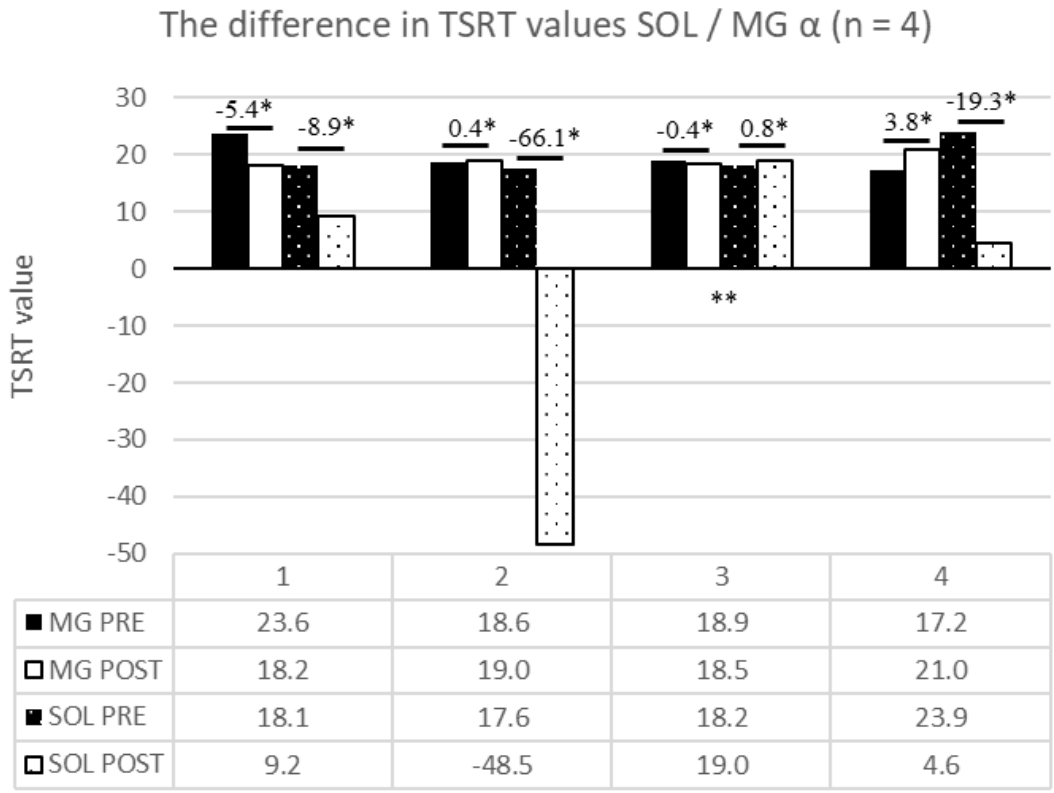


FIGURE 12 A and B. The participant with TSRT values before and after intervention from both muscles (SOL and MG). In this case, the TSRT values show strong correlation

coefficient ($R^2 > 0.6$). SOL TSRT angle pre/post 18.2/18.9 and MG TSRT angle pre/post 18.9/18.5.

When comparing the change (α) in SOL and MG individually, it shows no significant difference between these two muscles after the intervention (Figure 12.). Four participants had these all values and the comparison was made individually. The median of differences between SOL (α) and MG (α) was tested with the Wilcoxon Signed Rank Test. There was no significance (0.273) when the significance level is 0.05.



	1	2	3	4
MG PRE	23.6	18.6	18.9	17.2
MG POST	18.2	19.0	18.5	21.0
SOL PRE	18.1	17.6	18.2	23.9
SOL POST	9.2	-48.5	19.0	4.6

FIGURE 12. The change (*) in MG and SOL (dashed bars) before and after the intervention. ** = strong correlation (> 0.6) in values = TSRT value is reliable.

To examine if DSRT values have more variance in different ages, standard deviations of reflex responses were analyzed at given velocities and plotted them as a function of age. If the

correlation is negative (-), it represents more variance in younger participants and if the correlation is positive, it represents more variance in older participants. If the correlation is near zero, there is no correlation with the age and the variance in DSRT values. Both Pearson's and Spearman's correlation were calculated. Spearman's correlations coefficient is non-parametric and more suitable for a small sample size as in our study. The results show that there was a moderate correlation between SOL's DSRT values and age only at one velocity, SOL POST 200ms (Table 5 and Figure 13a). In MG's DSRT values at faster velocities (200°/ms and 300°/ms), there was found a moderate correlation with velocity and age. The velocities and standard deviation are shown in figures and the dashed line presents the slowest angular velocity 50°/ms. The slowest velocity differentiates with greater variance in DSRT values in the both muscles (SOL and MG).

TABLE 5. Comparison of standard deviation of DSRT values and age. *p > 0.3 = moderate correlation (Dancey & Riley 2007).

Velocity	Pearson's	Spearman's
SOL POST 200	0.162	0.315 *
MG POST 200	- 0.430	- 0.308 *
MG POST 300	- 0.596	- 0.496 *

The velocities and standard deviations were tested with Friedman’s test and it showed statistically significant values (p=0.041) with SOL’s PRE -measurements (Table 6). Pairwise comparison showed a statistically significant difference between the faster velocities and the slowest (50°/ms) velocity in SOL muscle (Table 6 and figure 13 a). There were no same trends and significant results with MG’s values or SOL POST -measurements, but some similarities are seen in figure 13b with the slowest velocity and standard deviation.

TABLE 6. Pairwise comparison of the velocities with SOL's PRE measurements.

Velocity (°/ms)	Sig.
SOL PRE 300 vs SOL PRE 200	0.846
SOL PRE 300 vs SOL PRE 100	0.699
SOL PRE 300 vs SOL PRE 50	0.012 *
SOL PRE 200 vs SOL PRE 100	0.846
SOL PRE 200 vs SOL PRE 50	0.020 *
SOL PRE 100 vs SOL PRE 50	0.033 *

Statistically significant difference * p = 0.041.

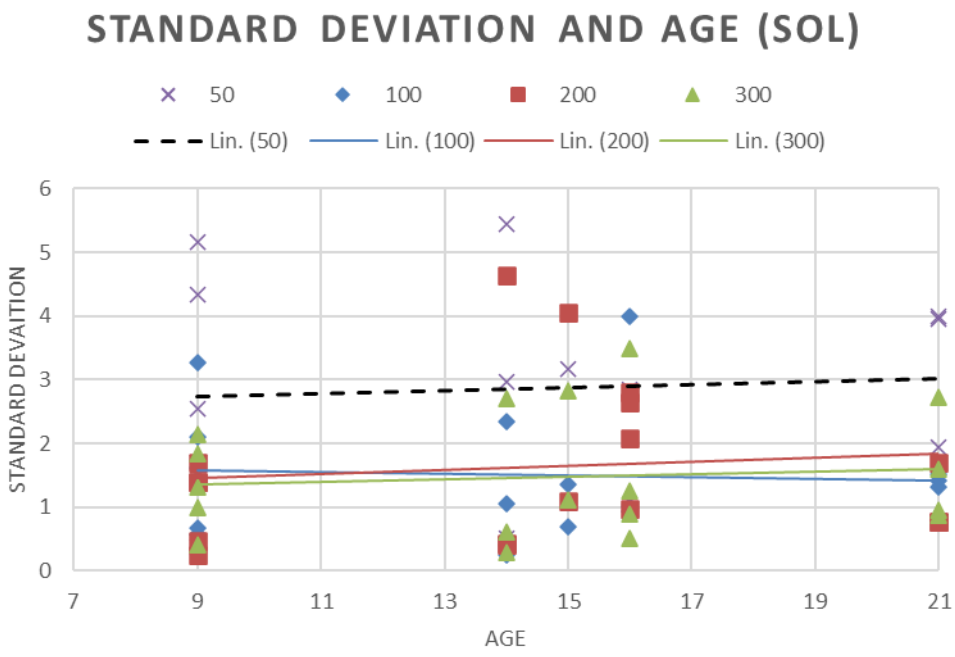


FIGURE 13 A. Standard deviation in different velocities and age comparison in the SOL muscle. All stretches are marked with different symbols. The lines represent the average of the standard deviations of each velocity. The dotted line represents the slowest velocity (50°/ms) that was statistically significant when comparing with other velocities in SOL muscle.

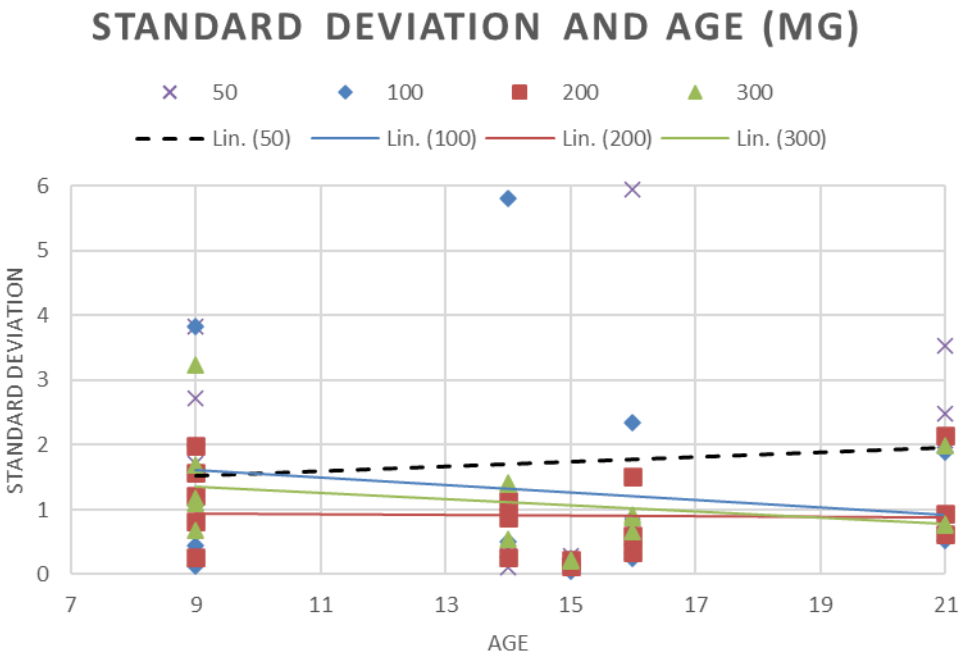


FIGURE 13B. Standard deviation in different velocities and age comparison in the MG muscle. All stretches are marked with different symbols. The lines represent the average of the standard deviations of each velocity. The dotted line represents the slowest velocity (50°/ms).

8 DISCUSSION

The present study found that strength training intervention with young and adolescents CP population had no significant associations to spasticity when measuring it with the tonic stretch reflex threshold in a rest position. The quality of linear regression and the correlation coefficient, which was used to calculate the TSRT values, remained weak in most cases.

The first research question was to find out whether there were strength training effects in calf muscles spasticity in the CP population. In three cases the results showed a strong correlation (< 0,6) (Germanotta et al. 2017) and the values showed small positive and negative, but not significant changes in TSRT. The changes in these three cases were ± 1 degrees in the angle and were not statistically significant.

The second research question was to compare whether the change in spasticity (TSRT) is similar in SOL and MG after strength training intervention in the CP population. Because the TSRT values were reliable only in the pre and post measurements of one case, making conclusions to one direction or another does not make sense based on this study.

The present study compared the TSRT measure with different ages and also as a secondary outcome with different velocities. It showed a moderate correlation with a few faster velocities in MG and SOL, but they were not statistically significant.

In spite of the limitations of the TSRT method, our findings about the the effects of strength training on spasticity do appear to be in line with the evidence from previous studies: three months intervention or shorter periods of training had no significant impact to neural networks or stretch reflex excitability (Jenkins et al. 2012).

The purpose of this study was to compare, if there were any changes in spasticity between soleus and medial gastrocnemius and whether the change is similar after strength training. The muscles (SOL and MG) are commonly seen as plantar flexors, but they also have other functions like controlling the postural control and body sway (SOL) and flexing the knee (MG) (Mochizuki et al. 2006, Platzer 2013). Mochizuki et al (2006) has found that the soleus muscle, common modulation of motor unit discharge is greater during postural tasks than during voluntary isometric tasks and can be observed in both bilateral and unilateral motor unit pairs. (Mochizuki et al. 2006.)

The results indicate that the correlation coefficient with MG's TSRT values is better than when using SOL as a target muscle. SOL behaves more as a postural muscle and the activation to this muscle is through the vestibulospinal pathway. This is not conscious behavior and the strength training effects depend on in what position the training is performed. Regarding the TSRT measurement and different muscles (SOL and MG), it shows that in this study MG gave more moderate or good values, and correlation coefficients of linear regression were stronger. It is a more superficial muscle, but also more active muscle in voluntary contractions. Somatosensory inputs appear to be processed in conjunction with vestibular inputs. which are active during a standing posture, but are relatively inactive during seated plantar flexion. (Horak et al. 2001, Mochizuki et al. 2006.) In this study, most of the exercises in the training program were performed in a sitting position or with extra support for the upper limbs. It decreases body sway and minimizes requirements for postural control. Therefore the activity in SOL is minimal. In future studies it is needed to consider if training should include more practice with postural demands and SOL activation, to evaluate the effectiveness of training to spasticity in these muscles.

In this study, the standard deviation of the DSRT values of different ages showed no significant differences. This indicates that when the measure outcomes have good, reliable values, it can be used by both children of different ages and adults. This study shows that at the slowest velocity (50 ms/°), there was a bigger variance in all ages. In research point of view, if the correlation coefficient of the linear regression is low because of high variability in DSRT values, also the TSRT values show no meaningful results. Therefore the use of the slowest stretches should be considered in future. Clinical tools such as the Modified Ashworth Scale and Modified Tardieu scale are not reliable and accurate (Bar-On et al. 2013, Van den Noort et al. 2017) and therefore, new methods to measure spasticity accurately are needed. TSRT has shown good validity and reliability (Calota et al. 2008, Calota & Levin 2009, Germanotta et al. 2017, Jobin & Levin, 2000) and therefore it is recommended for clinical use when measuring spasticity. In this study, the TSRT values had modest or no correlation and therefore the TSRT angles did not describe the true values of spasticity. This could be ensued because of unexperience of handling the EMG data and the problems with signal processing. Other possible problems concerning the validity in this study is discussed later in the Discussion section.

Neural adaptations after strength training have been studied with healthy adults (Aagaard et al. 2002, Jenkins et al. 2017, Vila-Cha et al. 2012) and even though muscle hypertrophy may be the same with different loads, the strength is not. Jenkins et al. (2012) found in their study, that 80% of 1RM loads were needed to induce neural adaptations and gain more strength. It is proposed that neural adaptations are primarily caused by an increase in the ability to maximally excite the motor neuron pool (i.e., agonist activation). This may be secondary to an increase in descending excitatory drive and better activation of the corticospinal tract. Alterations in evoked motoneuron potentials indicated an enhanced neural drive in descending corticospinal pathways. (Aagaard et al. 2002.) The difference in results may be attributed to various training protocols or methodological approaches. (Vila-Cha et al. 2012.) According to these neural adaptations, it seems that strength training is appropriate when improving agonist activation through a voluntary contraction. In CP, muscle cocontraction and spasticity, therefore, decrease because of better corticospinal drive via descending tracts.

This was the first study where TSRT measurement was used to see if intervention affects spasticity. In this study, the stretch reflex was measured in resting position and the results are in line with previous studies. There were no significant changes in stretch reflex after strength training. In future studies, it is recommended to study how the reflex responses change during actual muscle contraction and not only in resting position (Aagaard et al. 2002.). Neural adaptation occurs at both supraspinal and spinal levels and involves an enhanced drive in descending pathways from higher motor centers as well as increased motoneuron excitability and/or changes in presynaptic Ia afferent inhibition. Vila-Cha et al. (2012) found in their study that the improvements in MVC following strength training are likely attributed to the increased descending drive and/or modulation in afferents other than 1a afferents. Thus combined measures of the H reflex and V wave may provide a better understanding of the neural adaptations elicited by specific motor training programs.

The TSRT measure is primarily used in research and the use in practical fields is rare. The measure is safe and non-invasive. Spasticity as a phenomenon in neurological disorders is challenging both patients but also the professionals working in the field. Different symptoms of stiffness in neurological disorders are often generalized to spasticity. A few years ago the clinicians have concluded (Van Den Noort et al. 2017) that the term 'spasticity' should only be used next to stretch hyperreflexia. In the present study the measure when the joint angle,

moment, and electromyography are recorded, components of hyper-resistance can be quantitatively assessed and therefore the term spasticity can be used. General assumptions could not be made about the changes in spasticity because of the quality of data, but it can perform as a base study for future research.

The lack of this kind of studies, studies with poor quality or small groups in interventions have influenced the research and knowledge in this area. Subjects with neurological disorders are rarely homogeneous groups. Also, the term spasticity is challenging because of lots of variations in symptoms. CP is time-dependent but also dependent on which location the trauma has affected the brain. CP is a special group and most of all the immediate care is the priority in daily life. Current evidence suggests that these groups also need more physical activity and appropriate strength training to get more quality in their life. Systematic reviews have shown increasing evidence that strength training improves muscle strength in children with CP with no adverse effects on spasticity or ROM (Anttila et al., 2008, Scholtes et al. 2008, Dodd et al., 2002). This study is in line with previous studies and confirms that strength training has more positive outcomes than negative, and spasticity is not worsening.

A few challenges concerning the TSRT measurements were found in this study. The "basal activity" with many participants muscles before stretching was higher than recommended, and this might cause false results in EMG activity while measuring the TSRT. The interval between stretches is under discussion because 2.6-2.9 seconds might not be enough to achieve a relaxed state before the next stretch, in other words, depolarization is not ended before a new stretch. These challenges can be solved and improved in future studies. The posture when measuring the stretch reflex was sitting and the participant was guided to relax and all distractions from the environment were inhibited as good as possible. The sitting posture when the leg is straight and lower back flexed increases the tension in neural structures and other muscle-tendon properties. The posture stresses the lower part of the neuraxis and also the lumbar region with stiff fascia structures. Current knowledge of fascias and their properties are also needed to consider in this measurement when choosing the posture.

When research is done with children and adolescents, and also with a specific group (CP), there can always be multiple possibilities interfering and affecting the intervention. In this

intervention, the training loads were guided by the American College of Sports Medicine and National Strength and Conditioning Association guidelines (Garber et al. 2011; Faigenbaum et al. 2009) and CP specific guidelines (Verschuren et al. 2016). It was safe and the participants were motivated to training. Some professionals guided participants with practice, to ensure the best quality of the intervention. In the future, the data from other measurements in the EXECP project, like strength and gross motor function, will enlighten more about the benefits of this intervention in children and adolescents with CP and hopefully produces some guidelines for treatment and rehabilitation.

Limitations in this study are the measurements with EMG and the analyzes because the noise rates in measurements were out of the limits (the rest values were above 5% of the MVC values) and should be noted when interpreting the results. Because of the "basal activity" with a patient's muscles, it can give false results in EMG activity while measuring the TSRT. This could have been avoided with longer resting phase before next stretch, to limit the influence of the previous applied stretches on the response to the following. A period of 10 s is recommended between trials to ensure that muscle activity is minimal prior to the next stretch and to minimize the influence of stretch history on the response to the subsequent stretch (Mullick et al. 2012, Schmitt et al. 2000).

Conclusions and practical applications

The findings of the present study are in line with previous studies related to strength training and neural adaptations. Understanding these mechanisms could enhance the development of successful training and rehabilitation to improve strength, locomotion, and other daily life activities for people with neurological disorders. This study suggests that strength training is an appropriate training method for the CP population, and spasticity which is measured from the stretch reflex activity will not increase. This study is a part of a bigger EXECP-project and further results about changes in muscle strength, gross motor functions, and walking tests will indicate more accurately the benefits of strength training to the CP population. For clinicians and professionals, working with neurological patients, the most important message is that finding the most effective treatment for spasticity is individually assessing, measuring and based on these findings, making specific clinical reasoning before choosing the treatment. This

requires knowledge about neurophysiology and motor control and understanding the pathophysiology, how the trauma has affected the central nervous system.

REFERENCES

Aagaard et al. 2002. Neural adaptation to resistance training: changes in evoked V-wave and H-reflex responses. J Appl Physiol 92: 2309–2318.

Anttila et al. 2008. Effectiveness of physical therapy interventions for children with cerebral palsy: a systematic review. BMC Pediatr. Apr 24;8:14.

Armand et al. 2016. Gait analysis in children with cerebral palsy. Paediatrics. EFORT Open Rev 2016;1:448-460.

Bar-On L. et al. 2013. A clinical measurement to quantify spasticity in children with cerebral palsy by integration of multidimensional signals. Gait & Posture. May;38(1):141-7

Barrue-Belou et al. 2016. Neural adaptations to submaximal isokinetic eccentric strength training. Eur J Appl Physiol 116:1021–1030.

Bax et al. 2005. Proposed definition and classification of cerebral palsy. Developmental Medicine & Child Neurology, 47. 571-576.

Brashear, A. & Elovic, E. Spasticity – Diagnosis and Management. Demos Medical Pub 2011. E-book.

Brown 1994. Pathophysiology of spasticity. J Neurol Neurosurg Physchiatry. 57(7).

Burke 1988. Spasticity as an adaptation to pyramidal tract injury. Adv Neurol 47.

Calota et al. 2008. Spasticity measurement based on tonic stretch reflex threshold in stroke using a portable device. Clinical Neurophysiology 119 (2008) 2329–2337.

CDC 2004. Economic Costs Associated with Mental Retardation, Cerebral Palsy, Hearing Loss, and Vision Impairment --- United States, 2003. www.cdc.com.

Damiano D. 2014. Progressive resistance training increases strength but does not improve objective measures of mobility in young people with cerebral palsy. Journal of Physiotherapy, Volume 60, Issue 1, p. 58.

Desmedt JE & Godaux E.1978. Ballistic contractions in fast or slow human muscles: discharge patterns of single motor units. J Physiol 285:185–196

Dodd et al. 2002. A Systematic Review of the Effectiveness of Strength-Training Programs for People With Cerebral Palsy. Arch Phys Med Rehabil 83; 1157-64.

Enoka, R.M. 2008. Neuromechanics of human movement. Fourth edition. Champaign, IL: Human Kinetics.

Faigenbaum et al. 2009. Youth resistance training. Updated position statement paper from the national strength and conditioning association. The Journal of strength and conditioning Research, 23. 60-79.

Fleuren et al. 2009. Stop using the Ashworth Scale for the assessment of spasticity. J. Neurol. Neurosurg. Psychiatry 81, 46–52

Germanotta et al. 2017. Spasticity Measurement Based on Tonic Stretch Reflex Threshold in Children with Cerebral Palsy Using the PediAnklebot. Front Hum Neurosci 11.

Gillett et al. 2016. The impact of strength training on skeletal muscle morphology and architecture in children and adolescents with spastic cerebral palsy: systematic review. Res Developmental disabilities 56 ; 183-196.

Gillett et al. 2018. Functional Anaerobic and Strength Training in Young Adults with Cerebral Palsy. Medicine & Science In Sports & Exercise. Vol.50 (8), p.1549-1557.

Gracies 2005. Pathophysiology of spastic paresis II. Emergence of muscle overactivity. Muscle Nerve 31: 552–571.

Grey et al. 2008. Post-activation depression of Soleus stretch reflexes in healthy and spastic humans. Exp. Brain Research. 185 :189-197.

Hahn et al. 2012. Cortical and spinal excitability during and after lengthening contractions of the human plantar flexor muscles performed with maximal voluntary effort. PLoS One 7(11).

Kandel et al. 2013. Principles in Neural Science. 5th Ed. p. 794-795, 808-809

Katalinic et al. 2010. Effectiveness of stretch for the treatment and prevention of contractures in people with neurological conditions : a systematic review. Phys Ther. 2011.

Kirk et al. 2016. Explosive resistance training increases rate of force development in ankle dorsiflexors and gait function in adults with cerebral palsy. Journal of Strength and Conditioning Research. Vol 30 (10).

Knikou et al. 2008. The H-reflex as a probe: Pathways and pitfalls. Journal of Neuroscience Methods 171(1):1-12

Lance J.W. 1980. Spasticity: Disorder of motor control. Year Book Medical Publishers, Chicago.

Levin & Feldman 1994. The role of stretch reflex threshold regulation in normal and impaired motor control. Brain Res 657(1–2).

Lundy-Ekman L. 2013. Neuroscience. Fundamentals for Rehabilitation. 4ed. Elsevier.

Malhotra et al.: Spasticity, an impairment that is poorly defined and poorly measured Clinical Rehabilitation 2009; 23:651-658.

Marques I., Silva M., Silva A., Luiz L., Soares A & Naves E. 2019. Measurement of post stroke spasticity based on tonic stretch reflex threshold: implications of stretch velocity for clinical practice. Disability and Rehabilitation 2019, vol 41 nro 2, 219-225.

Mochizuki, G., Semmler, J. G., Ivanova, T. D., & Garland, S. J. 2006. Low-frequency common modulation of soleus motor unit discharge is enhanced during postural control in humans. Exp Brain Res, 175:584–595.

Mullick et al. 2012. Stretch reflex spatial threshold measure discriminates between spasticity and rigidity. Clinical Neurophysiology, Vol 124, 740-751.

Nakagawa A., Marques I., Silva M. & Naves E. 2014. Proposal of a real-time computational tool for the measurement of spasticity in stroke patients. Conference paper. 5th IEEE Biosignals and Biorobotics conference (BRC 2014).

Paulson A. & Vargus-Adams J. Overview of Four Functional Classification Systems Commonly Used in Cerebral Palsy. Review. Children 2017.4(4), 30.

Peterson et al. 2010. Muscle work is increased in pre-swing during hemiparetic walking Clinical Biomechanics 26, 859–866.

Piiparinen M. 2020. Toonisen venytysrefleksin kynnys CP-vammaisilla lapsilla ja nuorilla. Bachelor’s thesis. University of Jyväskylä.

Pin T., Dyke P. & Chan M. 2006. The effectiveness of passive stretching in children with cerebral palsy. Dev Medicine and Child Neurology 2006, 855-862.

Platzer Werner 2013. Color Atlas of Human Anatomy. Vol. 1 Locomotor System. p.262. Thieme.

Raine et al 2009: Bobath Concept – Theory and Clinical Practice in Neurological rehabilitation. Wiley-Blackwell.

Rossignol et al. 2006. Dynamic Sensorimotor Interactions in Locomotion. Physiol Rev 86: 89–154.

Ryan et al. 2017. Exercise interventions for cerebral palsy. Cochrane Database Syst Rev. Jun 11; 6.

Santos et al. Comparison of Modified Ashworth Scale with Systems and Techniques for Quantitative Assessment of Spasticity Literature Review. Journal of Neurological Disorders & Stroke 06/2017.

Schmitt et al. 2000. Stretch Reflex Adaptation in Elbow Flexors During Repeated Passive Movements in Unilateral Brain-Injured Patients. Arch Phys Med Rehabil 81:269-78.

Scianni et al. 2009. Muscle strengthening is not effective in children and adolescents with cerebral palsy: a systematic review. Australian Journal of Physiotherapy Vol. 55.

Seger & Thorstensson 2005. Effects of eccentric versus concentric training on thigh muscle strength and EMG. Int J Sports Med 26(1); 45–52.

Sheean G. 2002. The pathophysiology of spasticity. European journal of neurology, 9 (2002) 3–9.

Shemmell, J., Krutky, M.A., Perreault, E.J. 2010. Stretch sensitive reflexes as an adaptive mechanism for maintaining limb stability. Clinical Neurophysiology 121 (10), 1680–1689.

Shumway-Cook & Woollacott 2011: Motor Control translating Research into clinical practice (4 ed.). Philadelphia: Lippincott Williams & Wilkins

Silva et al. 2017. An improved approach for measuring the tonic stretch reflex response of spastic muscles. Computers in Biology and Medicine 80,166–174.

Stackhouse et al. 2005. Voluntary muscle activation, contractile properties, and fatigability in children with and without cerebral palsy. Muscle and Nerve 31, 594-601.

Scholtes V.A et al. 2008. Lower limb strength training in children with cerebral palsy – a randomized controlled trial protocol for functional strength training based on progressive resistance exercise principles. BMC Pediatrics 8, 41.

Stricker P.R., Faigenbaum A.D. & McCambridge T.M. 2020. Resistance training for children and adolescents. Pediatrics Jun 145 (6).

Van Den Hoorn et al. 2015. Effects of noxious stimulation to the back or calf muscles on gait stability Journal of Biomechanics. Volume 48, Issue 15, 26 November, 4109-4115

Van den Noort et al. 2017. European consensus on the concepts and measurement of the pathophysiological neuromuscular responses to passive muscle stretch. European Journal of Neurology, 24: 981–991

Valadao Pedro et al. 2019. Effects of a tailored exercise therapy intervention on physical capacity and function, neuromuscular mechanisms and cardiometabolic risk factors of children and young adults with cerebral palsy (EXECP). Unpublished material.

Vila-Chã et. al. 2012. Changes in H reflex and V wave following short-term endurance and strength training. J Appl Physiol 112: 54–63.

Walker S. 2018. Concurrent Aerobic and Strength Training : Scientific Basics and Practical Applications, edited by Moritz Schumann, and Bent R. Rønnestad, Springer, 2019. ProQuest Ebook Central. 75-86.

Walker S. & Häkkinen K. 2014. Similar Increases in Strength After Short-Term Resistance Training Due to Different Neuromuscular Adaptations in Young and Older Men. Journal of Strength and Conditioning Research Vol. 28(11).

Willerslev-Olsen M. et al. Sensory feedback to ankle plantar flexors is not exaggerated during gait in spastic hemiplegic children with cerebral palsy. J Neurophysiol 111: 746–754, 2014.

Yang et al. 2014: Post-activation depression of the lower extremities in stroke patients with spasticity and spastic equinovarus deformity. Arq Neuropsiquiatr 2015;73(6):493-498

Yates et al. 2010. Therapeutic approaches for spinal cord injury induced spasticity. Translational Neuroscience 1(2).

Zajac et al. 2003: Biomechanics and muscle coordination of human walking Part II: Lessons from dynamical simulations and clinical implications. Gait & Posture 17, 1-17.

APPENDICES

APPENDIX 1. Strength training intervention (10 exercises)

TABLE 1. Description of the strength training exercises.

	Seated Calf Raise	Standing Calf Raise	Seated Dorsiflexion	Seated Machine Knee Extension	Seated Machine Knee Flexion	Seated Horizontal Leg Press	Squat	Hip Flexion	Roman Chair Trunk Extension	Isometric Hollow Rock
Muscles targeted	Soleus, Gastrocnemius	Soleus, Gastrocnemius	Tibialis Anterior	Quadriceps femoris	Hamstrings	Gluteus maximus, quadriceps femoris, hamstrings, triceps surae	Gluteus maximus, quadriceps femoris, hamstrings, triceps surae	Iliopsoas, rectus femoris, sartorius, tensor fasciae latae, tibialis anterior	Erector spinae, multifidus	Trunk flexors, hip flexors, transversus abdominis
Inicial Position	Seated with knees at 90°. Forefoot on a 5cm step, ankle in maximal attainable dorsiflexion. Weight over the distal thigh of training leg.	Standing with hips and knees at 0°, and the forefoot on a 10 cm step in maximal attainable dorsiflexion. Holding on parallel bars for balance*.	Hips at 70-90°, knees at 0-20°, ankle in full plantarflexion. An elastic band on the forefoot resists the dorsiflexion movement.	Hips at 80°, knees at 115°. Knee and machine's lever arm center of rotation aligned. Lever arm positioned at the distal shank.	Hips at 80°, knees at 0-5°. Knee and machine's lever arm center of rotation aligned. Lever arm positioned at the distal shank.	Hips at 110-90°, knees at 90-100°. Feet and knees at hip width. 0-20° of hip external rotation is allowed.	Standing with hips and knees at 0°. Holding an adjustable support with both hands. Feet and knees at hip width. 10-20° of hip external rotation is allowed.	Supine position, arms laying by the side and both legs touching the mat. An elastic band on the forefoot resists the hip flexion and ankle dorsiflexion.	Hips and knees at 0°, the chair is 30° inclined. Padded support at pelvis height. Distal posterior part of the shank locked against a padded support.	Supine position, arms laying by the side and both legs touching the mat.
Kinesiologic Description	Unilateral or bilateral ankle plantarflexion.	Unilateral or bilateral ankle plantarflexion	Unilateral or bilateral ankle dorsiflexion.	Unilateral or bilateral knee extension.	Unilateral or bilateral knee flexion.	Unilateral or bilateral hip and knee extension, and ankle plantarflexion.	Bilateral hip and knee extension, and ankle plantarflexion	Unilateral hip flexion and isometric ankle dorsiflexion.	Isometric trunk and hip extension.	Isometric trunk and hip flexion. Knee extension.

Hip 0° = anatomical position (positive values = flexion), Knee 0° = fully extended. Ankle 90° = sole of the foot at right angles with tibial axis, > 90° = plantarflexion.
*If the exercise is too hard, leaning on the bars and helping with the arms will be allowed. If it becomes too easy, it will be done at the leg press machine.

APPENDIX 2. Instructions of training protocol for coaches and physiotherapists

The first block consists of 3 sets of 8 repetitions maximum (8RM), movement duration of 6 seconds (3s concentric and 3s eccentric), 60s of rest and no muscle relaxation between repetitions. This training load has been shown adequate to increase muscle strength and mass, being very safe as the intensity is approximately 50% of 1 repetition maximum (1RM; Tanimoto & Ishii 2006).

The second block, the volume in maintained while the intensity is increased by reducing the concentric movement duration to 1s and increasing the rest to 90s.

The third block, training volume and rest are maintained, but the sets are increased to four while the repetitions are decreased to 6, concentric muscle actions are now done as fast as possible while eccentric muscle action duration is reduced to 2s. The squat exercise due to its inherent higher intensity (i.e. body weight), will follow a different progression: one to four sets of 10 repetitions with the biggest attainable range of motion will be performed. Movement duration follows exactly like the other exercises, while the rest starts at 90 s, and will be decreased when possible to 60 s. After the entire volume is executable with 60 s rest, balance disks (Casall, Vantaa, Finland) will be placed under the participant's feet to cause instability and increase exercise difficulty.

The trainer will actively help the participant on the concentric phase of the movement in the positions in which the participant is not able to perform by himself. The exercise resistance will be selected based on the participant's strength on the optimal angles, thus the trainer will always be assisting on non-optimal angles, and never increasing the resistance. The eccentric phase will be performed unassisted, but constant feedback about movement velocity will be given. Due to the assistance during every repetition, no 8 or 6 repetition maximum test will be performed to adjust the weight, rather the trainer will frequently ask the participant to try and perform one more repetition, if successful, the weight will be increased for the next set. Whenever the participants are unable to perform a minimum concentric range of motion, the protocol will be adjusted to include an isometric muscle action of 3 seconds in each repetition. Thus, it will be a small concentric movement, followed by the isometric hold phase and then the eccentric phase. If during the intervention the concentric movement increases, the isometric part will be removed.

At the start of each training session, the trainer will ask the participant if any adverse events were experienced after the previous session. Participants will be constantly reminded to provide immediate feedback about any pain or discomfort during the training sessions.

The strength training program has two protocols (A/B), with a minimum of 7 exercises targeting lower limb and trunk muscles. The protocols are trained weekly in the order AB (2 sessions per week) or an alternating pattern of ABA/BAB (3 sessions per week).

Triceps surae muscle group was trained with seated and standing calf raises. Tibialis anterior (TA) muscle was trained utilizing a rubber band resistance against the dorsiflexion movement. Additionally, TA was trained isometrically during a hip flexion exercise, in which the participant laying supine had to flex the hip against the rubber band resistance placed on the forefoot. The thigh muscles was trained utilizing seated knee extensor and knee flexor machines. Additionally, lower limb muscles was trained using a leg press and squats holding with both hands an adjustable support. A dense foam ball was placed between the participant's knee to prevent hip adduction during leg press and squat. Exercises was done mostly unilaterally due to strength differences between limbs (80-100% of total training volume), except squat which was trained bilaterally. Trunk and hip flexors were trained isometrically with the hollow rocks exercise, in which the participant lies supine on the floor and had to lift the legs slightly above the floor (i.e. hip and trunk flexion, knee extension).