CHAPTER II

LITERATURE REVIEWS

This literature review comprises of two sections. The first section provides a comprehensive review of CP. The second section provides an intervention for strengthening muscle and describes outcome measurement in related to muscle weakness in CP.

1. Cerebral palsy (CP)

1.1 Definition of CP

CP is a chronic neurologic disorder caused by a static lesion to the immature brain that is characterized by deficits in movement and postural control. The motor impairments in CP are complex such as muscle weakness, spasticity, dystonia, contractures, bony deformities, coordination problems and loss of selective motor control that lead to activity limitations including gait, functional mobility and independence. CP can be classified according to topographic distribution of motor involvement, with the spastic form of the disorder the most prevalent, such as spastic diplegia, hemiplegia and quadriplegia (Figure 1) (1).

Spastic diplegia is the principle type of CP. It is characterized by impaired motor control resulting from a centrally mediated imbalance of muscle strength and tone in the lower extremities which are more severely affected than the upper extremities (1, 5).

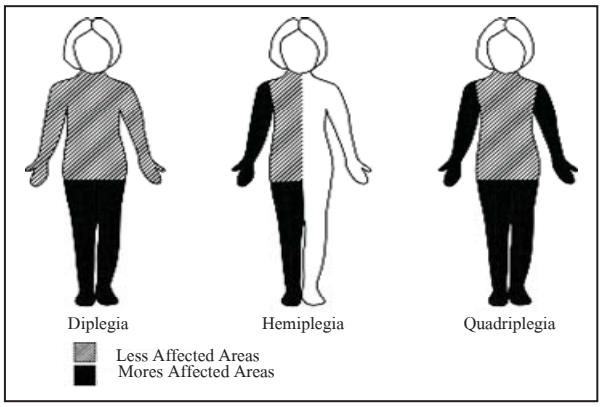

Figure 1 Children with spastic diplegia, hemiplegia and quadriplegia (14)

Without intervention, and often even despite intervention, this imbalance becomes more pronounced over time including muscle weakness and atrophy, soft tissue contracture and joint deformity. Although the majority of children with spastic diplegia are able to walk, their gait patterns differ from typical children. Most ambulatory children with spastic diplegia frequently walk with excessive flexion of their hips, knees and ankles throughout the stance phase. This movement abnormality is often called crouch gait (Figure 2) (6, 8-9).

Figure 2 Children with spastic diplegia walk with crouch gait (7)

1.2 Crouch gait in CP

Abnormalities of the knee which affect its range of motion in gait can seriously impair walking. Crouch gait/posture is one of the most prevalent and troublesome movement abnormalities among children with spastic diplegia. It is characterized by excessive knee flexion in combination with hip flexion and ankle dorsiflexion throughout the stance phase of gait cycle (6-7). Therefore, insufficient knee extension at terminal swing in children with spastic diplegia has been concerned because it affects foot position at initial contact and is linked to limit normal peak knee extension at terminal swing (15). The study of Arnold and co-workers (16) demonstrated that children who walked with crouch gait had significantly larger knee flexion angles than typically developing (TD) children during gait cycle and a larger hip flexion and ankle dorsiflexion were also found (Figure 3-5).

Generally, children who walk with crouch gait typically exhibit a shorter stride length and slower than typically developing children (6). Importantly, crouch gait is problematic because it increases patello-femoral force, impedes toe clearance, and dramatically increases the energy requirements of walking (17). Without intervention, crouch gait typically worsens over time. Hence, if it is uncorrected, the crouch gait can lead to chronic knee pain, skeletal deformities and loss of independent ambulation (7, 17).

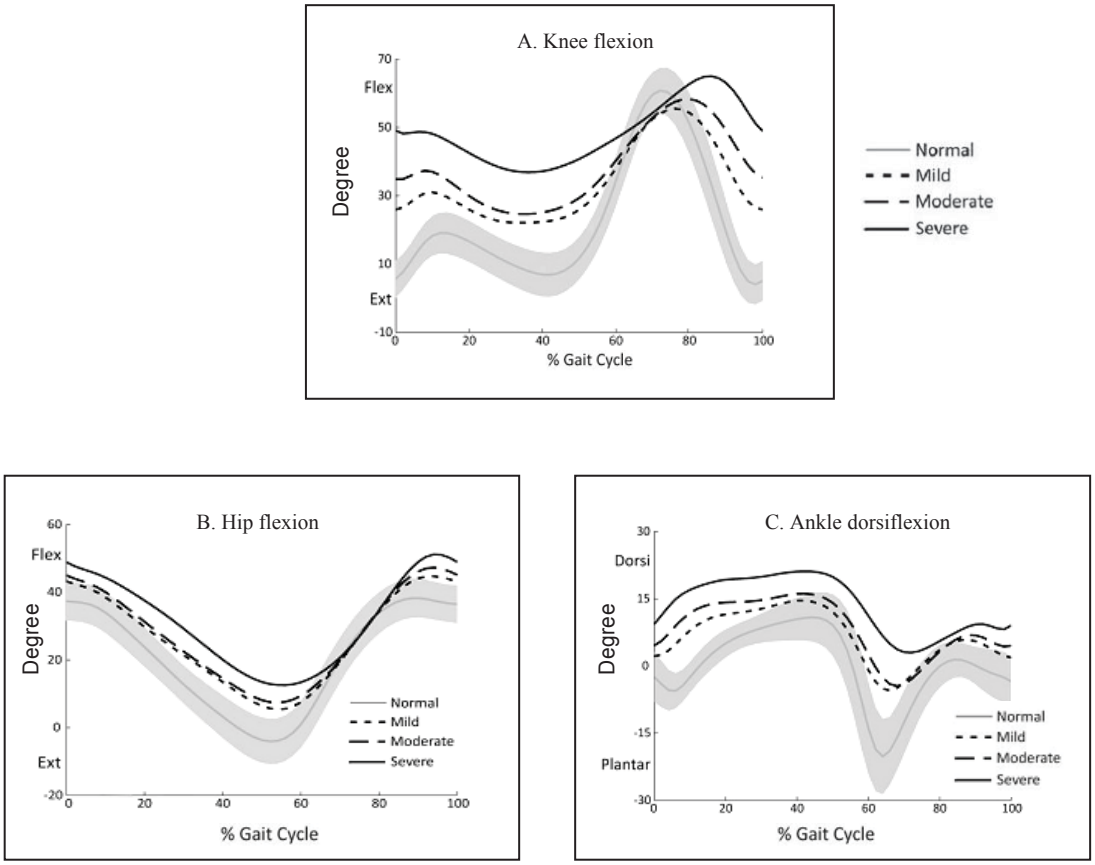

Figure 3-5 Average joint kinematics for typically developing children and children with CP who walk with crouch gait. The three crouch groups (mild, moderate and severe) had significantly greater than normal knee flexion (A), hip flexion (B) and ankle dorsiflexion (C) when averaged over the gait cycle (16)

A host of possible factors has been linked to the development of crouch gait including muscle tightness, weakness, spasticity, skeletal deformity and motor control deficits. Therefore, treatments to improve crouch gait show varying success. However, examining the cause of progressive crouch gait and appropriate corrective treatment is difficult (16). The biomechanical causes of the excessive hip flexion and knee flexion in children with spastic diplegia are often unclear and make it challenges

to determine the most appropriate treatment (17). Possible causes of crouch gait include hamstrings tightness or/and spasticity, psoas tightness or/and spasticity and hip extensors or knee extensors or ankle-plantarflexors weakness or/and excessive length (1, 6, 8, 17). Conservative treatments such as stretching of spastic knee flexors, strengthening of knee and hip extensors or orthosis fail to address this problem in most cases (6). The most popular method of treatment of crouch gait is surgical lengthening of the hamstrings which reduces knee flexion (18). Unfortunately, it is difficult to predict which patients will get benefit from hamstrings surgery. Results of hamstrings lengthening surgery are irreversible. Some individuals achieve dramatic improvements in knee extension, stride length and walking efficiency following surgery while others show little improvement or get worse (7). Therefore, quadriceps strengthening and hamstrings lengthening to correct skeletal deformities may be also performed to balance the knee and increase the capabilities of muscles to extend the knee in children who walk with crouch gait (8-9, 18).

1.3 Knee function and crouch gait in CP

During normal gait, the knee function is a key to stance stability, and several muscles contributing to knee control contract at selected intervals within the gait. The propose is to provide the stability and mobility need for ambulation. During stance, the quadriceps is the most direct source of extensor control and knee extensors act to decelerate knee flexion. During swing, both knee flexors and extensors contribute to limb progression. Among fourteen muscles acting on the knee, the quadriceps is the dominant muscle group. The four vasti heads of the quadriceps cross only the knee

joint whereas rectus femoris cross both the knee and hip joint (19). The basic function of the quadriceps muscle is to powerfully extend and straighten the leg. Additionally, it plays an important role in many activities of daily life such as standing, walking, up and down stairs, and running, that require less than 135 degrees of knee flexion (20). During walking, however, the quadriceps is only used to restrain the shock-absorbing flexion during the loading response and other mechanisms are employed to attain optimum extension in single stance. The knee uses a larger arc of motion during swing than any other joints. Approximately 60 degrees of flexion are needed to assure toe clearance of the floor. However, the necessary knee function is attained by multiple mechanism rather than just direct knee muscle action. Pre-swing ankle plantar flexors muscle action, hip flexion and tibial inertia are the major determinants of initial swing flexion. The local knee flexors have a minor role. Consequently, knee function involves the entire limb in both stance and swing (19). Generally, normal gait has several attributes including a stability in stance, foot clearance in swing, pre-positioning of the foot in terminal swing, an adequate step length and conservation of energy. The first four of these components can be negatively affected by lower extremity muscle weakness (4). In children with CP, the importance of muscle strength can be seen in the direct relation to motor function. Crouch is the one characteristic gait of children with spastic diplegia (19). Hamstrings tightness or/and spasticity, psoas tightness or/and spasticity and hip extensors or knee extensors or ankle-plantarflexors weakness or/and excessive length are toughed as a cause of crouch (1, 6, 8, 17). Specifically, strength of the knee extensors has been correlated with gross motor mobility and gait velocity (5, 8). In addition, it has been negatively correlated with crouch degree at heel strike and the

score on the energy expenditure index (21). Therefore, children who have stronger quadriceps strength are more likely to perform better on gross motor skills, walk faster, ambulate with less knee crouch, and have greater energy economy when they walk (5, 8-9, 21). Walking ability is a major concern for the families of children with CP and parents often ask about and focus on their child's walking ability. Hence, improving or maintaining this ability is often considered to be the primary focus of most therapeutic interventions addressing the motor problems seen in children with spastic diplegia (8). Because of muscle strength is thought to be directly related to gross motor function and gait velocity, especially knee extensors muscle which has an important role in knee joint control and walking ability (5, 8, 19, 21). In addition, the quadriceps muscle is also the one of three primary muscle groups (i.e., vasti, hip extensors and ankle plantarflexors) that maintains erect posture for supporting the whole body against the gravity, thus, several researches have addressed on muscle weakness management and suggested that children with stronger knee extensors are more likely to perform better on gross motor skills and walk faster with less knee crouch and greater energy economy (5, 8-9, 19, 21-22). Specifically, Damiano and co-workers (8-9) found that children with spastic diplegia could increase quadriceps muscle strength through simple heavy resistance exercise, improved the degree of crouch at initial floor contact during the freely selected speed walking, and increased in stride length during free and fast speeds walking. Moreover, they also found that the weakest point for quadriceps was at the 30 degrees of knee flexion (Figure 6).

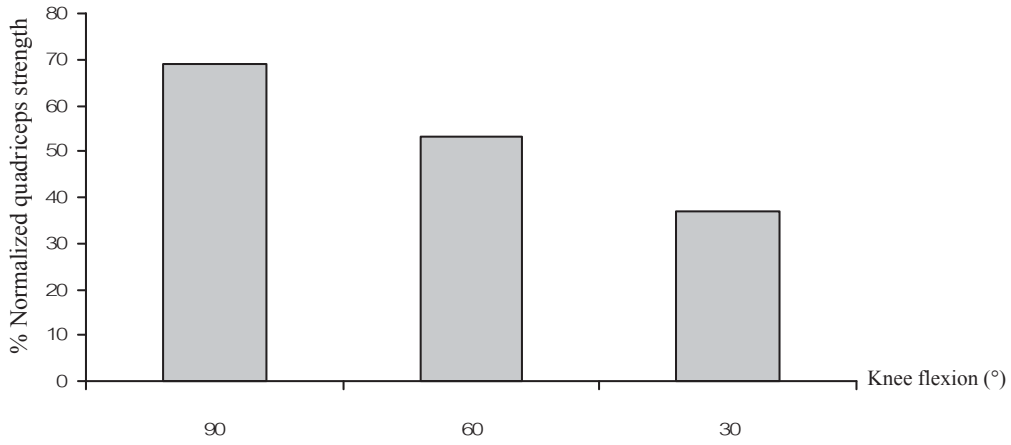

Figure 6 Percent normalized quadriceps strength in children with CP with respect to knee joint angle during sitting (Adapted from Damiano and co-workers) (9)

The study of Kramer and Macphil (21) and Damiano and co-workers (23) presented that knee extensors muscle strength had positive correlations with the Gross Motor Function Measure (GMFM) and gait velocity. Additionally, the most important finding from Kramer and Macphil (21) study was that knee extensors strength was directly related to efficient walking and gross motor mobility. They also suggested that a lack of knee extensors strength could be a reason why adolescents with CP were limited in their standing, walking, running and jumping activities. As a result, several studies focus on quadriceps strengthening because of its important role in maintaining upright posture in standing and walking, as well as in other functional activities such as squatting and stair climbing.

1.4 Muscle weakness in CP

Muscle weakness in CP is multifactorial including pathologic motor unit recruitment, agonist-antagonist cocontraction, lost of selective motor control, reducing muscle cross sectional area, volume and rigidity due to collagen infiltration (24). Damiano and Wiley (5) demonstrated that children with spastic diplegia were weaker than age matched typical development children. Similarly, Stackhouse and co-workers (25) compared the maximum voluntary isometric contraction (MVIC), antagonist co-activation, and fatigability between children with CP and typical development children. They found that the typical development children were stronger, had higher MVIC and a lower rate of antagonist co-activation than children with CP. In addition, Rose and Mcgill (26) investigated torque and motor unit recruitment between CP patients and controls. They concluded that CP patients could not recruit the higher threshold motor units necessary for maximum contraction, and they were not able to modify the firing rates of low threshold motor units. Specifically, Ross and Engsber (27) demonstrated that people with CP were weaker than controls in all muscle groups. Importantly, they also presented that there was no relationship between antagonist muscle weakness and the level of spasticity. Hence, the opinion that spastic antagonist led to decrease muscle strength in CP patients was not sustained. For the muscle fiber type distribution in people with CP, it was also found the changes in muscle fiber type were variation, decreasing in number of type I fibers (slow twitch fibers) and increasing in number of type II fibers (fast twitch fibers) was reported in CP patients, hence, compatible with higher fatigability (24). However, both muscle atrophy and hypertrophy with either type I fiber dominance or type II fiber dominance and with increased variation of muscle fiber size as well as

variation dominance of fiber types were also reported (26). Additionally, Ross and co-workers (28) indicated that the type I fiber dominance was in the ambulatory children and the type II fiber dominance was in non-ambulatory children.

Muscle weakness in CP may have several explanations according to above mentions. There are also problems attributable to biomechanical conditions such as muscle shortening and musculoskeletal deformities. These lead to changes in lever arms and thus reduce the output of muscle force in term of torque, resulting in lever arm dysfunction and muscle weakness (6). Importantly, weakness interferes with function and leads to limited activity and participation (5, 23).

2. Intervention for strengthening muscle

2.1 Conventional strengthening program

In general, strength gains have been attributed to both neural adaptation and muscle hypertrophy which related to neural and muscular factors (29). In short term training, neural adaptations related to learning, coordination, and the ability to recruit prime movers to play a major role in the strength gain (30). Adaptive alterations in nervous system function that elevate motor neuron output largely account for the rapid and large strength increases early in training, often without an increase in muscle size and cross sectional area. In addition, neural adaptations with resistance training may result from influencing effects of greater efficiency in neural recruitment patterns, increased central nervous system activation and improved motor unit synchronization, lowering of neural inhibitory reflexes and inhibition of Golgi tendon organs. Alternatively, in long term training programs, an increase in size of the prime movers plays the major role in strength development (29, 30). The effect of a long

term resistance training on muscle fiber generally relate to adaptations in the contractile structures. These usually accompany substantial increases in muscular force and power through a given ROM and increasing in muscle force with exercise training provides the primary stimulus to initiate the relatively slow process of skeletal muscle growth, or hypertrophy (29). The relative roles of neural and muscular adaptations in strength improvement with resistance exercise training for individuals as shown in Figure 7. (30).

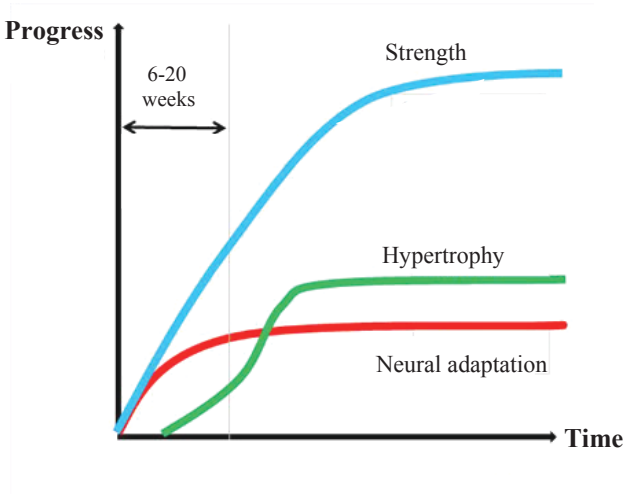

Figure 7 Relative roles of neural and muscular adaptations in strength improvement with resistance exercise training (29)

2.2 Strengthening muscle in CP

Muscle weakness is a primary impairment in children with CP. Researches have shown that muscle force production can be improved in children with CP and that improved strength can translate into functional gains (23-24). The principles used for strength training, in term of weight progression and specificity of training, are similar to those for people without disabilities (31). Historically, many physical therapists

held the opinion that muscle strengthening could lead to increase in spasticity, and was therefore contraindicated in CP. Another common belief was that children with CP would not be able to gain strength from resistance training due to loss of selective motor control, and weakness was not a major component of motor dysfunction in CP. Thus, treatment was focused on inhibiting spasticity, and excessive effort was avoid as it believed to increase spasticity and impair motor control (9). However, a systematic review of strength training in children with CP by Mockford and Caulton (32) reported that strength gain can be achieved in children with CP and without serious side effects and no increased in spasticity. In addition, Ross and Engsberg (27) investigated the relationship between spasticity and strength in 60 individuals with spastic diplegia and 50 individuals without disability. They measured spasticity using a KinCom dynamometer that stretched the knee flexors and ankle plantarflexors at different speeds and recorded passive resistive torques. Besides, muscle strength of the knee flexors/extensors and ankle dorsiflexors/plantarflexors was also measured using the KinCom to measure maximum contraction through the ROM at a speed of 10º/sec. The group with CP was significantly weaker and had greater resistance torque (spasticity) compared with the non-disable group and there was no relationship between spasticity and strength with in a muscle group and no correlation between spasticity of one muscle (agonist) and strength of the opposing (antagonist) muscle. Ross and Engsberg (33) indicated that strength was highly related to motor function and explained far more of variance than spasticity in children with mild spastic diplegia. Muscle strengthening is an important part of rehabilitation in patient with CP. Not only can children with CP become stronger but they can also increase strength at a rate similar to persons with weakness who do not have a brain insult

(31). Importantly, children who participated in muscle strengthening programs had a better quality of life and improved function (32, 24). The systematic review study of Mockford and Caulton (32) showed that strengthening exercise protocols varied considerably across studies. Muscle groups were targeted using free weight, strengthening machines and isokinetic exercise. Length of program in most previous studies was 6 weeks or more with frequently of 2-3 times per week at least. Isometric strength was selected as an outcome measure in most studies. In addition, gait analysis, motor skill, functional test and energy expenditure were also used as outcome measures of strength training program.

Interestingly, for contribution of quadriceps muscle weakness to the degree of crouch, Damiano and co-workers (8) used free ankle weights to strengthen the bilateral quadriceps in an attempt to decrease the amount of knee crouch during gait. Fourteen ambulatory children with spastic diplegia were participated in their study. All exhibited the characteristic crouch gait pattern with a minimum of 10 degrees of knee flexion when standing or walking in the absence of a static contracture at the knee. Each participant exercised 3 times per week for 6 weeks using free ankle weights at load of approximately 65% of maximum isotonic force production. The maximal voluntary contraction of the quadriceps muscle was measured before, at the mid point, and immediately following the exercise program at 30, 60 and 90 degrees of knee flexion. Gait analyses were performed before and after the strengthening program to determine whether quadriceps muscle strengthening influenced gait. The results showed that children with spastic diplegia could increase quadriceps strength through heavy resistance exercise. In addition, significantly improved knee extension during walking was also found in an improvement of the degree of crouch at initial

floor contact at the freely selected speed and an increase in stride length at free and fast or speeds. In addition, another study of Damiano and co-workers (9) in the same participants and training program, a normal comparison group of 25 children was also tested under identical conditions. When compared quadriceps strength between typical and the CP group, the results showed that before this short term resistance exercise, the CP group was weaker than the control group, but at the end of the training period, quadriceps strength in the CP group was improved as same as the control. Although, quadriceps strength increased significantly at all three angles of knee flexion, the results indicated that the weakest of quadriceps muscle was 30 degrees. In addition, quadriceps weakness was shown to be a factor in crouch gait in children with spastic diplegia. This research concluded that the children with spastic diplegia could strengthen their quadriceps muscle using a simple exercise program without any increase in spasticity. And recently, Unger and co-workers (34) determined the effect of a strength training program targeting multiple muscle groups using basic free weights and resistance devices 1-3 times per week, for 8 weeks at school setting. Programs were individually designed for specific ambulant adolescents with spastic CP. Three-dimensional motion analysis was used to investigate the participants' gait and identified a reduction in crouch compared to an increase in crouching in the control group. They recommended the use of similar programs with inexpensive equipment to encourage muscle strengthening in adolescents with CP.

Although several studies have shown that resistance training program can increase muscle strength and improve gross motor function in children with CP, more specifically, strengthening quadriceps muscle showed increasing in quadriceps

strength and improving the degree of crouch in children with spastic diplegia. However, most participants in the previous studies have mild in severity and are able to ambulate without assistive devices; therefore, they may have adequate strength to perform resistance exercise. Children who have insufficient strength or ambulatory with assistive device may not be able to get full benefits of conventional resistance training program as mentioned above.

2.2.1 Outcome measurement in relation to muscle strengthening in CP

2.2.1.1 Muscle strength

Evaluation of muscle strength can be done in various ways (32, 35). The most common and easy method is manual muscle testing (MMT). This technique is based on the patient performing a maximum voluntary isometric contraction (MVIC) of a muscle against the examiner's hand and is easy to perform but inaccurate. The patient needs to cooperate with the examiner and performs a maximal contraction of one muscle group only, without causing a stretch reflex. This is a task that many children with CP find very difficult due to cognitive limitation or co-contraction of antagonists or agonists. The MMT offers a subjective measure of strength based on a 0-5 scale that is largely dependent on the providers' educational and clinical experience. In addition, the sensitivity of conventional MMT to detect changes in muscle strength is poor, especially in grades 4 to 5. Experience and skill among clinicians is the inter-rater difference in manual muscle testing evaluation (35). Moreover, the strength measured in a certain muscle length cannot be extrapolated to other lengths (35-36). This evaluation method will remain an important part of the clinician's evaluation for population with CP, but its use in research should be considered as doubtful (35).

Conversely, the isokinetic testing machines offer good stabilization of the patient, accurate and repeatable measurement and no examiner bias. This is probably the best method for research, but its clinical use is limited due to the time required, the necessary cooperation and understanding of the patients, cost, and lack of portability. Isokinetic muscle testing is proved to be reliable in patients with CP (36). This method can be used in most age groups, and performing properly can yield reliable results. This method is done with laboratory device with the resistance arm pivoting around an axis aligned with the segment tested. Resistance can be set to different velocities, and there is a registration of torque through the whole range of motion. Various types of muscle contraction can be measured (concentric, eccentric and isokinetic). However, this device may be difficult to adapt for children because of the size and the time it takes to adjust for testing of different muscles. All measurement methods require patient cooperation and understanding, and low selective control is a hindrance to all measurements techniques (24, 35-36).

Alternatively, a hand held dynamometer (HHD) is a portable device that can be used to obtain more discrete, objective measures of strength during manual muscle testing than can be achieved via MMT. A HHD has been shown to be a less expensive, portable, easy, lightweight, and requires little training for proficient application to measure muscle strength. The force transducer of the instrument is placed on the examine limb and the patient is asked to perform a maximum contraction. It provides a measurement of MVIC. The MVIC is a standardized, objective, quantitative and sensitive tool for the measurement of muscle strength (35). There was reported that the used of hand-held dynamometry as a primary strength measurement tool for healthy, strong individual is not recommended (24, 35). In

children with CP, the HHD has a good reliability and suitable for clinical use for measuring the strength of maximum isometric muscle contraction in lower extremity muscle groups (36).

2.2.1.2 Muscle spasticity

Spasticity is a common finding, present in over 80% of all children with CP. It is one feature of upper motor neuron syndrome that may affect functionality, limit daily living activities and diminish quality of life in children with spastic CP (1). It can be defined as a velocity dependent increase in muscle tone. In clinical practice, for measurement of muscle spasticity, there are many different assessment methods for spasticity varying from clinical ordinal scales to complex electrical or orthotic equipments such as pendulum test, electromyography and isokinetic dynamometry (37-38). However, these methods are mostly used for research studies and it is hard to elicit cooperation in these children. Alternatively, the method that most widely used in practice is; measuring the resistance of spastic muscles to quantify muscle tone such as Modified Ashworth Scale (MAS). The MAS is performed by passively moving a joint through its range of motion at a standard speed and rating the resistance of stretched muscle on a 6-point scale. It is technically suited to assess hypertonia than spasticity (39). Nevertheless, hypertonia and spasticity are closely associated and the scale is widely accepted as a clinical tool appropriate to assess spasticity. The MAS has been recently modified from original five-point scale and accepted as clinical scale of spasticity (10, 37-39). Recently, the MAS was reported that it had poor to good inter-rater reliability in children with hypertonia and it had

moderate to good intrarater reliability in these population, therefore, assessment by the same investigator should perform (39).

2.2.1.3 Range of motion (ROM)

Range of motion (ROM) is defined as the amount of movement that occurs at the joint. There are several methods in clinical practice for measurement the ROM. Goniometry is an essential tool for assessing a patient with muscular, neurological, or skeletal disability. Goniometric measurements may be used to quantify functional loss, physical disability, progression of a disease process, or effectiveness of a treatment program (40). To be useful as a clinical evaluation tool, however, goniometric measurements must be reliable and fairly easy to perform. In clinical environment, two sizes of 180º and 360º goniometers are the most common devices for measuring ROM. It is important for the investigator to know that joint positions and ROM can be measured consistently and without measurement error. The reliability of joint position and ROM using the universal goniometer depends on the joint being assessed but has generally been found to be good to excellent (20, 40). Although the universal goniometer is more reliable than visual estimation of joint ROM, intrarater and interrater reliability should be determined for each clinical facility (40).

One method in clinical practice for measuring knee joint ROM that related to characteristics of crouch is extensor lag (41). Children who walk with a crouch gait pattern may not have the ability to extend their knees at the end range of motion. Muscle groups such as quadriceps may become functionally long, due to biomechanical malalignments causing a stretch weakness at the end range of motion.

Assessment of the active ROM versus passive ROM of the knee joint can give insight into true muscle strength. Typically, knee extensor lag or quadriceps lag is a clinical sign with often profound functional relevance for patients who are unable to fully extend the knee (42-43). It is as a measure of inadequate full range of quadriceps strength. Muscle lag is as an inability to actively move a joint to its passive limit. In more detail, the passive limit should be the passive limit that can be achieved without producing significant discomfort, and without exerting more than mild against resistance from joint stiffness or other soft tissue tightness. The active limit should be determined with the patient positioned so that the moving segment is restricted by gravity but no other external load (42). In general, the lag may results from muscle atrophy, muscle weakness, muscle tightness, pain, effusion, or reflex inhibition (42-43). However, it may be fully passively extended by the clinician. Typically, to complete the last 15 degrees of knee extension, a 60% increase in force of the quadriceps muscle is required (43). Hence, if quadriceps is not strong enough to fully extend the knee, it is often accompanied by lag. Quadriceps lag can be examined in difference positions including supine, long sitting or sitting. The magnitude of quadriceps lag is the angular limit of active knee extension minus the angular limit of passive knee extension. In clinical setting, the common device such as a universal goniometer can be used to assess the lag angle of knee extension ROM. When full active extension is present, there is no lag (41-43).

2.3 Electrical stimulation (ES)

Historically, ES has been used for thousands of years for a variety of purposes (44). Generally, it has been used to establish strength, establish ROM, establish neuromuscular reeducation, manage pain, reduce edema, reduce spasticity, and as an aid in the healing of bone fractures and pressure sores on healthy or pathology population (44-45). For strengthening muscle, a muscle can be stimulated by passing an electrical current directly across it or its motor nerve. This technique, called electrical stimulation training, has been proven effective in a clinical setting. It is used to reduce the loss of strength and muscle size during periods of immobilization and to restore strength and size during rehabilitation. In addition, it also has been used experimentally in training healthy subjects because it can increase muscle strength, however, the gains reported no greater than those achieved with more conventional training (12, 30). The parameters for ES in general clinical application are reported in Table 1 (45).

Table 1 Parameters for electrical stimulation in general clinical application (45)

Type of current	Pulsatile or burst-modulated alternating current
Amplitude	Maximum tolerate to produce maximum range movement
Pulse duration	100-300 μs.
Waveform	Subject preference
Frequency	20-100 pulses or burst/s
Duty cycle	Start with on : off 2 s: 10 s or 5 s: 15 s
Type of contraction	Isometric
Contractions/session	10-20 at maximum tolerable intensity or 15 min/session, 2-3 times daily
Frequency of sessions	3-7 times/week

For children with CP, ES continues to grow because it has potential as a passive and non-invasive therapy, which is proposed as a useful modality in CP due to the lack of selective muscle control required for specific strengthening programs since it can target isolated muscles. Directly loading the muscle through specific strengthening programs or sufficiently high intense ES seems to be the directly way to gain muscle strength in this population (46-47). In addition, it may be particularly useful in expanding the functional outcome of other therapeutic treatments that address different components of the motor disorder (10). Although it has the advantages of being non-invasive, simple, inexpensive and minimal side effects, pain or discomfort in some cases may be occurred because the motor nerve is most susceptible to stimulation. As a result, proper electrode size, suitable type of electrode, appropriate electrode position, optimal stimulation parameters and close monitoring are quite important especially in pediatric population (12, 44). Generally, the goal of the ES in CP patients is to increase muscle strength, decrease spasticity and improve motor function. It is often used via a portable unit that can be used in the home-setting by parents or the patient (10, 11). It has been applied in three different ways including, first, the stimulation can be applied functionally or known as functional electrical stimulation (FES). This term is used when the aim of treatment is to enhance or, produce functional movement. A second way, threshold electrical stimulation (TES) or previously called therapeutic electrical stimulation, it does not require active movement by the child but it is applied the stimulation at low intensity, subcontraction level, for several hours during the night and used when the goal of treatment is to decrease spasticity and/or increase muscle strength. Finally, ES can be applied therapeutically for shorter duration and at sufficient high intensities to

produce muscular contraction or known as neuromuscular electrical stimulation (NMES). This form of ES is commonly used when the main purpose of treatment is to increase muscle strength and enhance motor function. It may be applied to the muscle either during movement or without movement occurring. However, in some cases, TES may be an attractive alternative treatment, especially in children who do not tolerate NMES well (11, 48-49). Although NMES is the most commonly used form of ES in CP population as an alternative muscle strengthening, its clinical utility remains a topic for debate and there are few studies which have demonstrated that NMES might have a place in the treatment of this population (11, 13). According to a reviewed study about the effects of ES on muscle strength and motor function in CP population, Kerr and co-workers (11) identified 18 articles that applied TES or NMES to the various muscles and the results demonstrated both statistically significant and insignificant improvement in muscle strength and motor function. It seemed that there were more evidences to support the use of NMES than TES. However, the findings of the studies must be interpreted with caution because they generally had insufficient statistical power to provide conclusive evidence for or against these modalities. Kerr and co-workers (11) highlighted that the majority of ES studies have poor methodology; therefore, they suggested that more rigorous study designs, follow up and homogeneous subjects are required for the unequivocal support of the use of ES in CP population. Among several previous studies that applied NMES on different muscles, two studies focused on quadriceps muscle weakness in CP patients. In a case report, Daichman and co-workers (10) determined the effects of NMES home program on impairment and functional skills in a 13 years old child with spastic diplegia who ambulate with posterior walker. In this study, NMES was applied to the

right quadriceps muscle for 6 weeks. The NMES home program was administrated every other day for 10 repetitions and 3 sets via a portable battery powered electrical stimulator by the parents. The stimulation was completed with the child in supine with the knees position in approximately 40 degrees of flexion over a bolster. The intensity was set by the trainer to produce full knee extension as much as possible at the highest intensity the child could tolerate. The NMES protocol used in this study was designed according to the overload principle. The author applied the principles of strengthening, including overload and time for muscle repair between muscle training sessions, to increase strength of the quadriceps muscle group. The parameters used in this study are presented in Table 2.

Table 2 Stimulation parameters used in the study of Daichman and co-workers (10)

Waveform	Balance symmetrical biphasic pulsed current
Pulse duration (μs)	300
Frequency (pps)	35
On: Off time (s)	10 : 50
Ramp up : down (s)	2 : 0

In the study of Daichman and co-workers (10), quadriceps strength was assessed using the HHD, hamstrings spasticity was assessed using the MAS and KinCom isokinetic dynamomerter, gait spatiotemporal parameters were assessed using GAITRite and functional motor performances were assessed using PEDI. After intervention, right quadriceps strength was increased, hamstrings spasticity was decreased. In the untreated left quadriceps, strength remained unchanged but spasticity increased. Positive changes were seen in gait spatiotemporal parameters

including walking velocity, step length, step time, cadence, double support time and the size of the base of support. In addition, the PEDI showed development of new skills, including the ability of climb stairs with less assistance. Therefore, the author concluded that NMES was an effective therapeutic technique to improve strength and motor function of a child with spastic diplegia. However, the use of a single subject in this study led to the limited generalization. Conversely, a randomized placebo-controlled trail of Kerr and co-workers (13) was carried out to investigate the efficacy of NMES in strengthening the quadriceps muscle of both legs in children with CP. Sixty children including diplegia, quadriplegia, dystonia, ataxia and non-classifiable types were randomized to NMES, TES or placebo group. However, more than half of children in this study were able to walk without aids. The NMES home program was applied for 1 hour daily, 5 days per week at the highest intensity the child could tolerate via a portable battery powered electrical stimulator by the parents. In all children this produces an observable muscle contraction equivalent to an isometric contraction in supine with the knees being minimally flexed over a pillow. For training protocol, both legs were stimulated concurrently. In the TES group, the stimulation was applied for 8 hours per night, 5 nights per week, at a sensory threshold level. For placebo group, treatment was applied similar as TES group but no stimulation was delivered through the electrodes. All treatments were carried out at home for 16 weeks. The parameters used in the study of Kerr and co-workers (13) are presented in Table 3.

Table 3 Stimulation parameter used in the study of Kerr and co-workers (13)

Waveform	Balance symmetrical biphasic pulsed current
Pulse duration (μs)	300
Frequency (pps)	35
On: Off time (s)	7 : 12
Ramp up : down (s)	2 : 1

For outcome measures, peak torque of the left and right quadriceps muscle, gross motor function and impact of disability were assessed using isokinetic dynamometer, GMFM and lifestyle assessment questionnaire-cerebral palsy (LAQ-CP), respectively at baseline, end of treatment (16 weeks) and at a 6 week follow-up. After intervention, the results showed that no statistically significant difference was demonstrated between NMES or TES versus placebo for quadriceps strength or function. However, statistically significant differences were observed between three groups for impact of disability at the end of treatment, but only between TES and placebo at the follow up. The author highlighted that a shortfall in their experimental design and an exceedingly high inter-participant variability contributed to masking the effect of the stimulation and they also suggested that further research is required to show whether NMES and/or TES may be useful as an adjunct to therapy in children with CP who have a difficulty in resistive strengthening program.

According to above issues, although there were evidences determined the effects of NMES on muscle strength and motor function, previous studies on the efficacy of this intervention is undetermined because evidences remain insufficient statistical power because most of NMES studies in previously were non-empirical studies which lacked adequate statistical power and differentiation in the physical abilities of the

participants, hence, further well control study design with homogeneous participants is important and necessary required to determine whether NMES may be useful to CP population. More specifically, the contribution of quadriceps muscle weakness by NMES strategy to the degree of crouch has not been well documented. Therefore, the purpose of this study was to determine the effects of NMES on knee extensor muscle in individuals with spastic diplegia who ambulate with crouch gait in a trial with control design for seven weeks of training and two weeks of follow-up. We hypothesized that NMES had potential to increase quadriceps strength, reduce quadriceps and hamstrings spasticity and improve quadriceps lag and angles of hip, knee and ankle joints during standing at the end of training and follow up. This study may provide the knowledge that may be used as an adjunct therapy for children with CP who have a difficulty in resistive strengthening program.