CHAPTER II

LITERATURE REVIEWS

1. Cerebral palsy

Cerebral palsy (CP) is a movement and posture disorder resulting from developing the brain defect, which brain lesion is non-progressive. It can also occur before, during or shortly following birth. The disorders can be characterized by abnormal muscle tone, posture, reflexes, motor development and coordination (1). The classical symptoms are spasticity (4), delayed motor development, paralysis, seizures, unsteady gait, and dysarthria. CP is classified by the body parts involved such as spastic diplegia, spastic quadriplegia and spastic hemiplegia as shown in Figure 1 (4, 25). However, CP can be classified according to the type of movement disorder such as spastic, ataxic, and athetoid (4).

Spastic diplegia is the most common form of CP (4). Spasticity refers to the inability of a muscle to relax which may interfere with limb manipulation and control (1). Spastic diplegia affects the leg muscles more than the arm muscles. The condition usually involves spasticity which causes difficulty in balance, movement and co-ordination (1). Nevertheless, all children are eventually able to ambulate. This skill is delayed and abnormal gait patterns (7).

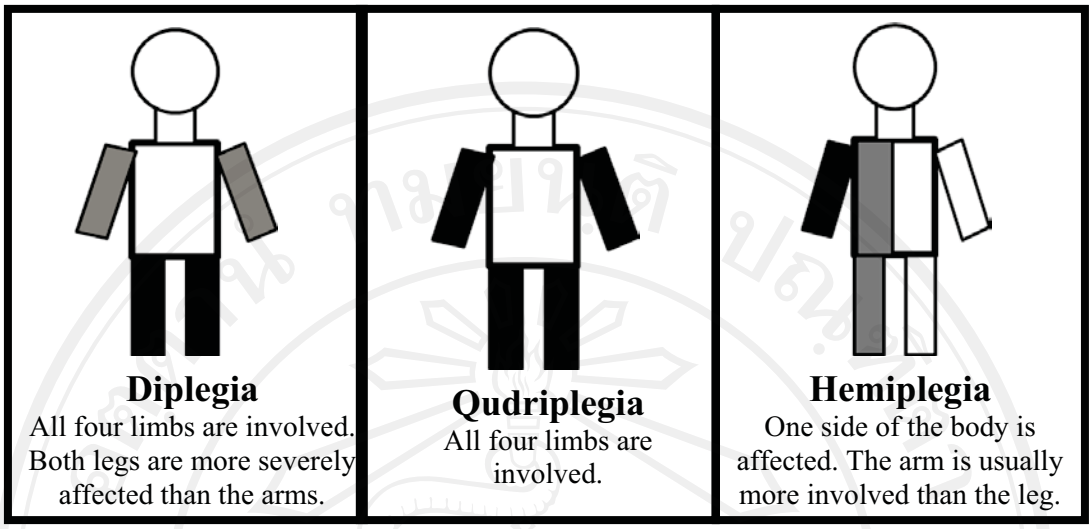

Figure 1 Children with spastic diplegia, quadriplegia, and hemiplegia

1.1 Crouch and genu recurvatum in gait in children with spastic diplegia

One of the most common gait abnormalities in children with spastic diplegia is crouch gait, which is characterized primarily by excessive knee and hip flexion (Figure 2) in stance phases and diminish knee extension during the terminal swing phases (26-27). Lack of knee extension at terminal swing in children with spastic diplegia can affect foot position at initial contact which can be linked to a limitation of precursor gait events (heel stride) that affect the normal peak knee extension behavior at terminal swing (28). Children who perform the crouch gait also exhibit an abnormally short stride length and decrease walking speed which differs from typical children (27). Possible causes of crouch gait include hamstrings tightness or/and spasticity or/and weakness and ankle-plantarflexor weakness or/and excessive length (6) but the biomechanical causes of the excessive hip flexion and knee flexion are unclear. Therefore, it is difficult to determine the most appropriate treatment. If this

crouch gait is uncorrected appropriately, it can lead to chronic knee pain, skeletal deformities, and loss of independent ambulation.

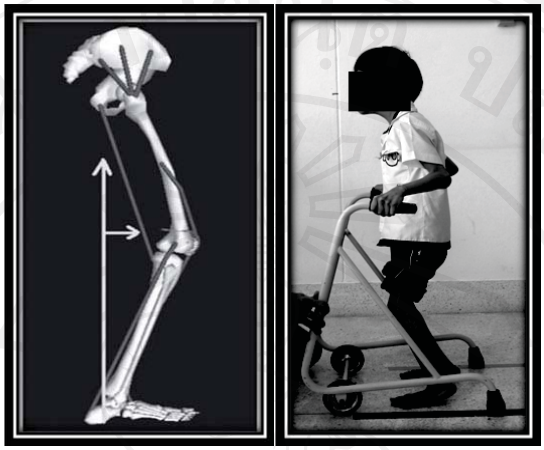

Figure 2 Crouch gait: knee flexion and the external flexion moment (29)

The principles for the correction of crouch gait are variable (29). However, there are three primary muscle groups that maintain erect posture which compose the hip extensors, vasti muscles, and soleus muscle. The hip extensors and vasti muscles act during the one third of stance phase thought to generate about 20-30% of the force necessary to maintain erect posture whereas, the soleus muscle produces about 40 - 50% (6). However, clinicians who treat gait abnormalities have documented that the vasti muscles make the important contributions to the knee extension and also induce hip extension during the stance phase of normal walking (6).

Genu recurvatum (knee hyperextension in gait) (Figure 3), which is defined as full extension or hyperextension of the knee during the stance phase of gait is common in patients with a variety of neurologically based on impairments (8). It has been reported in nearly one half of patients with cerebral palsy (8). The genu

recurvatum may be due to muscle imbalance or may be secondary to abnormalities of other joints of the lower extremity (LE) (8). The disorder is typically described to a combination of quadriceps weakness (as a compensatory strategy, ankle plantar flexor spasticity, heel cord contracture, quadriceps spasticity, and/or gastrocsoleus weakness (30). Patients with knee recurvatum have variable peak extensor torque values associated with their knee hyperextension (8). Therefore, patients with knee recurvatum are not required to have high extensor torque values. Additional, patients with weak quadriceps may also develop recurvatum because knee provides a mechanism to control an otherwise instability limb by hyperextension during the stance period of the gait cycle, with the center of gravity anterior to the knee (30).

For these patients, knee hyperextension could be considered a reasonable compensation, and treatments specifically aims to reduce knee hyperextension, such as quadriceps exercise which is very important to balance the knee or hamstring lengthening and adductor myotomy which are usually combined with a quadriceps strengthening program (29-30). Concisely, the genu recurvatum can be managed by restoration of muscle balance, proper alignment of joints and establishment of correct posture in the line of gravity (29-30).

Most children with spastic diplegia have imbalance and muscle weakness. Specifically, the group of muscles is important to maintain erect posture. Hence, the simplest of treatment must restore the extension moments and weaken the flexion moments (6). Muscle lengthening and tendon transfer surgery are employed to correct excessive knee flexion by reducing tightness in muscles that flex the knee. However, the benefits of surgery had been reported either to improve or worsen knee

extension, stride length and walking efficiency (31). Therefore, the strengthening exercise and surgery to correct skeletal deformities may be also performed to increase the capabilities of muscles to extend the knee.

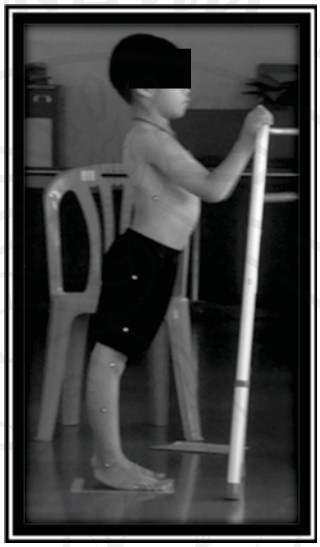

Figure 3 Genu recurvatum: hyperextension of the knee

Muscle weakness in spastic CP is associated with reduced motor drive (32). The most common finding in CP are an increased incidence of muscle fiber atrophy, whereas this appear to be more selective in fast-twitch muscle fibers, increased intramuscular fat and connective tissue in the most involved muscle groups, and increased percent of slow-twitch muscle fibers (11, 22). The muscle weakness resulting from inability of agonist to produce force, constraint by antagonist, inappropriate length due to contractures, changes in muscle fiber properties, and poor selective muscle control (19), specially the problems attributable to biomechanical conditions such as muscle shortening, musculoskeletal deformity, and muscle weakness. Consequently, the produce force of muscle is reduced. Finally, the muscle weakness leads to limit function (33).

The quadriceps muscle weakness has been shown to be related to a crouch gait position (17). Damiano et al (7) found that children with spastic diplegia have the weakest quadriceps at 30 degree of knee flexion and weaker than normal children (7). Strengthening of quadriceps muscles has been shown to lead to a decrease in the knee flexion angle, suggesting the potential for energy cost also be affected and directly related to quickly walking (10, 17).

Moreover, children with spastic diplegia can increase muscle strength and improve the degree of crouch at initial contact during the freely selected speed, and increase in stride length during free and fast speeds after heavy resistance exercise on quadriceps muscle (13). Also, they found that the weakest point for quadriceps was 30 degree of knee flexion (Figure. 4 Adapted from Damiano et al).

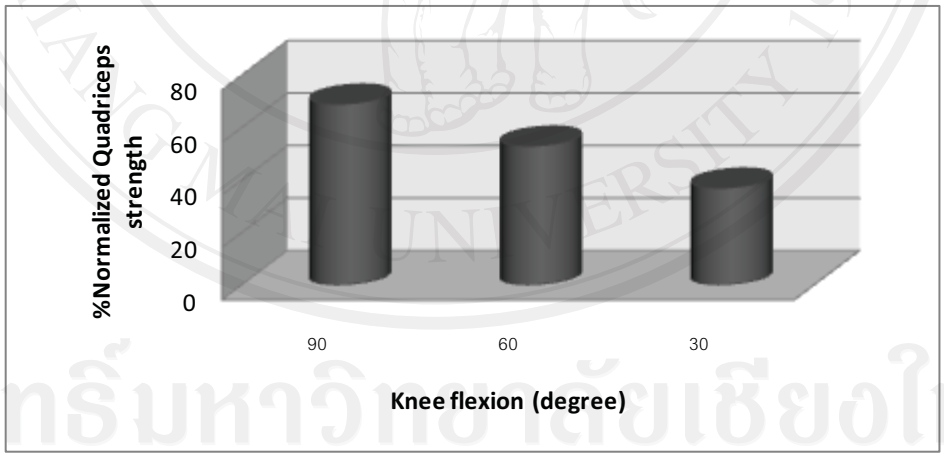

Figure 4 Strength deficits in children with CP with respect limb to position (13)

1.2 Knee function and gait

The Quadriceps femoris muscle is the strongest and most powerful muscle in the human body. This muscle has four heads, which has rectus femoris, vastus

medialis, vastus intermedius, and vastus lateralis. The basic function of the quadriceps femoris muscle is to powerfully extend and straighten the leg. In addition, quadriceps femoris muscle plays a crucial role in many daily activities such as standing, up and down stairs, walking, and running, which require less than 135° of knee flexion (34). During normal gait, the vasti muscles activity begins in terminal swing, rapidly increasing intensity early in loading response and critical in knee stabilizing during the knee mid stance, however, the rectus femoris muscles activity has a short period of action between late pre swing and early initial swing (35).

The maximum force during isometric knee extension occurred when the knee was in approximately 60 degrees of flexion and that the force or torque decreased with extension (35). Therefore, inner range of knee extension is weaker than other ranges (7). So, children with spastic diplegia are difficult to do activities of daily living (ADL) such as walking, dressing of trousers, and both walking up and down stairs (29).

Normal gait has several attributes included a stability in stance, a clearance in swing, a pre-position of the foot in terminal swing, an adequate step length, and a conservation of energy. Children with CP have lost in varying degrees for all of these attributes (6). In addition, the primary gait abnormalities are loss of selective motor control, difficulties with balance and abnormal muscle tone.

The loss of selective motor control is more severe in the distal portion of the limb than the proximal (32). Children with CP can produce appropriate muscle contractions only at very low frequencies and are unable to produce the controlled temporal sequences of muscle activity required for everyday movements (26). Children with spastic diplegia usually demonstrate fairly good selective motor control

at hip, very poor control of the knee, and poor control of the ankle and foot. Consequences of disequilibrium of balance in children with spastic diplegia have fairly minimal involvement in deficient equilibrium reactions to some degrees. Children with spastic diplegia can walk without aides whose children often fail in lateral and posterior equilibrium reactions, and so may be fall backward. Furthermore, shortened muscle tendon length also exacerbates the effect of muscle spasticity, the velocity dependent increased sensitivity to stretch, further contributing to joint contracture and constraints on mobility. This imposes abnormal movement during gait is dominated by synergist muscle activation (32). There are several ways in which spasticity interferes with function in CP. For example, the spasticity acts like a brake on the system or inhibits voluntary control of movement and drags on a movement caused high energy consumption (6).

1.3 Motor function classification

There is the evidence that severity of CP is related to motor outcome, development of gross motor function (36). The Gross motor function classification system (GMFCS) provides a standardized method of classifying gross motor function and also it is valid for classifying the gross motor abilities and limitations of children with CP as shown in Appendix A (36). Emphasis is on usual performance in home, school, and community settings (i.e., what they do), rather than what they are known to be able to do at their best (capability). Bodkin et al (37) reported the good inter-rater reliability and good construct validity of GMFCS in children with CP. Moreover, the GMFCS can be used to predict the motor development (37). The GMFCS level remained relatively stable in children with CP and is supported its use

in clinical practice and research. At present, there has been documented of development of gross motor function in CP. Furthermore, GMFCS is based on self initiated movement with particular emphasis on sitting and walking (35). Classification is based on observation of their self-initiated movement and need for assistive technology and wheeled mobility. The descriptions for the 6 to 18 years old reflect the potential impact of environments factors (e.g., distances in school and community) and personal factors (e.g., energy demands and social preferences) (35).

1.3.1 General headings for each level

Level I Walks without limitations

Level II Walks with limitations

Level III Walks using a hand-held mobility device

Level IV Self-mobility with limitations; may use powered mobility

Level V Transported in a manual wheelchair

Distinctions between levels

- Distinctions between levels I and II

Compared with children in level I, children in level II have limitations of walking long distances and balancing; may need a hand-held mobility device when first learning to walk; may use wheeled mobility when traveling long distances outdoors and in the community; require the use of a railing to walk up and down stairs; and are not as capable of running and jumping.

- Distinctions between levels II and III

Children in level II are capable of walking without a hand-held mobility

device after age 4. Children in level III need a hand-held mobility device to walk indoors and use wheeled mobility outdoors and in the community.

- Distinctions between levels III and IV

Children in level III sit on their or require at most limited external support to sit, are more independent in standing transfers, and walk with a hand-held mobility device. Children in level IV function in sitting (usually supported) but self mobility is limited. Children in level IV are more likely to be transported in a manual wheelchair or use powered mobility.

- Distinctions between levels IV and V

Children in level V have severe limitations in head and trunk control and require extensive assisted technology and physical assistance. Self-mobility is achieved only if the child can learn how to operate a powered wheelchair.

2. Outcome measurement

2.1 Maximal voluntary isometric contraction (MVIC)

The maximum voluntary isometric contraction (MVIC) is a standardized, objective, quantitative and sensitive for the measurement of muscle strength. The MVIC can be accurate quantified with hand held dynamometer (HHD). The HHD is easier to use, light weight, portable and, less expensive. The force transducer of the instrument is placed on the examined limb and the patient is asked to perform a maximum contraction. The caveats of the instrument are the inability to stabilize the patient, the possibility of the examiner pressing on the instrument and adding to the measured force. It has a good reliability (38) and suitable for clinical use in children

with CP. For example, Taylor et al (38) evaluated the test-retest reliability of measuring lower-limb isometric strength with a HHD in young people with spastic diplegia. They suggested that a HHD can reliably measure changes in lower-limb strength for groups of young people with CP. In similarly, Berry et al (40) determined the intrasession and intersession reliability of isometric force measurements using HHD of the knee extensors in children with CP. The children with CP demonstrated well to high reliability coefficients for isometric force measurements of knee extensors muscle using handheld dynamometry. Moreover, Damiano and Abel (13) determined clinical effectiveness of in 6-weeks isometric strength training on lower extremities with a hand-held dynamometer in children with spastic CP. They found the significant increase in muscle strength, gross motor function, cadence and walking velocity. However, they did not find significant change in energy expenditure.

2.2 Modified Ashworth Scale (MAS)

The Modified Ashworth Scale (MAS) is the most widely used and accepted clinical scale of spasticity. It measures resistance during passive soft-tissue stretching. The inter- and intra-rater reliability of MAS varied from moderate to good (40-41). Therefore, the repetition of measurements by the same physiotherapist, and experience may not affect reliability. These are the general rules of measurement the MAS (41):

- Because spasticity is "velocity dependent" when the faster the limb is moved the more spasticity is encountered. The MAS is done by moving the limb at

the "speed of gravity." This is defined as the same speed a non-spastic limb would naturally drop.

- The test is done a maximum of three times for each joint. If it is done more than three times, a stretch impacts the score.
- The MAS is done prior to goniometric testing. Goniometric testing provides a stretch and a stretch impacts the score.

2.3 Quadriceps lag

Muscle lag is a clinical sign with profound functional relevance for patients (42). Muscle lag is an inability to actively move a joint to its passive limit. In more detail, the passive limit can be achieved without producing significant discomfort, and without exerting more than mild force 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 resisted by gravity but no other external load. The magnitude of quadriceps lag is defined as a condition in which the active range of the knee extension is less than the passive range of knee extension. It believes that the lag is always abnormal (42). Quadriceps lag is commonly used in healthy adult. The quadriceps weakness will be most evident in this final extension range (42). Therefore, the terminal extension exercises at the end of the functional range against gravity or in the gravity-reduced position are recommended (42).

2.4 Angles of hip, knee and ankle joints during standing

In stance, the reduction of extension joint moments produced by the lever arm dysfunction plus the elongated soleus, vasti and gluteus maximus muscles, allow the ankle to dorsiflex excessively and the hip and knee to drop into flexion. The ground

reaction force, which is now posterior to the knee, plus the abnormal action of the biarticular hamstrings, rectus femoris and gastronemius, all act to increase the flexion moments at the hip, knee and ankle (6).

Moreover, the crouch gait is characterized by excessive flexion throughout the stance phase (6-7). The knee is excessively flexed at initial contact (6) and remains excessively flexed while in contact with the ground. Additional hamstrings EMG tracing for children with spastic diplegia indicates that the hamstrings are active throughout stance phase. The excessive hip flexion in late stance could either be associated with excessive knee flexion in stance or inappropriate anterior pelvic tilt (7). Finally, the absence of excessive ankle dorsiflexion in stance indicates that plantar flexor weakness is not contributing to his crouch gait pattern (7).

3. Strength training intervention

Each movement of the body depends on muscle strength. It is essential for daily activities and learning new functions. Muscle strength is even more crucial for individuals with a disability than for those without, especially children with cerebral palsy (22). The weakness appears to have a stronger relationship with the level of motor functioning than other impairments such as spasticity or muscle tightness (22).

There is an evidence of a relationship between lower limb strength values and motor abilities. Whether a child is able to walk independently or not, 50% can be predicted from muscle strength (43). Strengthening was an integral part of early treatment for CP (19). It has been suggested that an emphasis on strengthening weak muscle combined with motor learning principles are of greater benefit than focusing on hypertonicity and its reduction (6). Several studies have focused on management

of muscle weakness and suggested that if children had a good strength of knee extensors, they could perform better on gross motor skill, walk faster and have less knee crouch (8, 34, 44). In addition, Damiano et al (7) found that children with spastic diplegia could improve quadriceps muscle strength through resistance exercise.

During the first several weeks of resistive training, there are more neuro-functional adaptive changes than structural changes within the muscle (22, 45). The body learns to recruit the correct muscles in the proper sequence and inhibits unnecessary muscle recruitments. Thus, strength gain is not solely a property of the muscle but rather it is a property of the motor system. In addition, during the first week of strengthening exercises, there is a reduction in the co-activation of other muscles which results in a decrease in energy expenditure, an improvement in movement control, and a decrease in muscle spasticity (22). After 6 weeks of resistive training, gains in strength are gradually achieved by increased size or hypertrophy (22, 45). Therefore, strength training can improve both neural function and gains in muscle mass in individuals with neuro-motor disorders (20, 45). This information provides an effective way to improve everyday physical function for children with CP, especially the duration time and intensity of the program as shown in table 1 (17).

At present, strengthening exercises improve muscle strength in children and young adults with CP without an increase in spasticity (12). Consistently, Fowler et al (46) reported that no changes in spasticity following quadriceps femoris muscle strengthening exercise with maximum efforts in children with the spastic diplegia

(46). Children with CP can increase the strength in the agonist at the same rate as children with weaknesses that do not have a central nervous system disorder (47).

Table 1 Guidelines for strengthening

Parameters	Value
Repetition	8-12
Frequency	3 times/wk
Load	65-100% of maximum voluntary contraction
Sets	3-5
Rest	1 minute
Velocity	slow
Duration	up to 6 wk

Most importantly, strength training programs can produce positive changes in walking ability and gross motor function (13, 48). Hence, children who have the greater strength may lead to higher level of function such as increasing stride length and decreasing crouch gait, and having higher scores on the Gross Motor Function Measure (GMFM) (13). If children with CP have not enough knee extensors strength, they will not be able to stand, walk, run and jump.

3.1 Conventional strength training in children with CP

There are several ways to strengthen the lower limb in children with CP such as a free weights, isokinetic and isometric training, concentric and eccentric program, weight machined, and electrical stimulations (ES) (14). These programs are

advantage in inducing the muscle strength, improving function, increasing range of motion and reducing spasticity in children with CP (14). For example, Unger et al (47) examined the effect of a strength training program targeting multiple muscle groups using basic inexpensive free weights and resistance devices in a school, 1-3 times per week, for 8 weeks. Program was individually designed in independently ambulant adolescents with spastic CP. They used 3-D motion analysis to evaluate 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. Eek et al (33) examined the effect of muscle strengthening in 16 children with spastic diplegia. Training consisted of exercises for lower extremity muscles with free weights, rubber bands and body weights for resistance during a period of 8 weeks. Three-dimensional motion analysis, GMFM, muscle strength evaluation using a hand-held dynamometer, and other clinical evaluations were performed. There was a significant increase in the strength in several muscles and a significant improvement in the GMFM. Most spatio-temporal parameters were within normal before the intervention, and there was an increase in hip extensor moment and plantar flexor power generation at push off. In addition, MacPhail and Kramer (49) examined the effects of isokinetic resistance training after 8 weeks on functional mobility and walking efficiency in adolescents with CP and found significant improvement of knee extensor and flexor strengths with no spasticity increasing. Recently, Morton et al (50) investigated the effects of progressive resistance training of the quadriceps and hamstrings muscles on children with hypertonic CP and showed very promising results that this progressive strength training is very beneficial to children with CP by

showing that quadriceps and hamstrings strengths could be increased significantly and children with CP could walk faster after strength training, however, it decreased after the follow up examination.

Isometric exercise is used for strengthening exercise in the rehabilitation (35). It is static contraction training with involve muscular actions in which the length of the muscle does not change and there is invisible movement at the joint therefore, stretch responses should not be evoked (51). The isometric exercise requires no expensive equipment, and can be added to any exercise (35). Although maximum muscular contraction can be achieved, it will only strengthen very isolated areas of the muscle group.

The isometric exercise is required muscles to hold a position for a certain length of time in which the muscles begin to recruit and activate motor units to help maintain this contraction. Therefore, the motor units that are rarely exercised within a particular muscle are now brought into use and they are forced to contract continuously, time after time; with no appreciable decrease in force output which allows muscles to achieve a state of maximum force very safely and effectively (52). However, isometric exercise causes blood pressure to rise higher than the other methods of strength training. This exercise could lead to a ruptured blood vessel or irregular heartbeat. If there are weak blood vessels or heart trouble and can rupture a blood vessel or develop an irregular heartbeat. In addition, the isometric exercise has some differences in training effect as compared to dynamic exercises. While isometric training increases muscle strength at the specific joint range of motion (ROM), the dynamic exercises increase muscle strength throughout the full ROM which enhancing the twitch force of a muscle (53).

Isometric exercise performed against stationary position without moving the joint. Therefore, children with CP have specific of muscles weakness at inner range. Importantly, isometric exercise is very safe in children with CP because the intensity can be adjusted quickly and precisely (52). Moreover, children with CP may have muscle tightness so who have difficulty performing an exercise through a full range of motion.

3.2 Electrical stimulation (ES)

If children have at least some voluntary control in a muscle group, the capacity for strengthening will exist. In the absence of voluntary control, strength training is more problematic, but may be facilitated by the use of ES (54) because it may be helpful for strengthening muscles that cannot be sufficient recruited with voluntary effort. Directly loading the muscle through specific exercises or sufficiently intense ES is the only direct way to increase muscle strength in CP and may be particularly useful in augmenting the functional outcome of other intervention that address different components of the motor disorder (51).

General considerations in the clinical application of ES has been used on subject both healthy and pathology (18). The best documented effects of ES on muscle are increasing strength and endurance, improving range of motion, neuromuscular reeducation, reducing edema, healing bone fracture and pressure sore. Furthermore, a comprehensive review of the literature in physical therapy has been appeared effectively in neuromuscular electrical stimulation (NMES). The NMES may delay muscle wasting during denervation or immobilization and optimizes recovery of muscle strength during rehabilitation (18).

The NMES is the most common form of ES used for patients with neuromuscular disorders. The NMES is adjunct therapy for children with CP which has supported since 1970s (55). It is applied of electrical current to innervated muscle to augment muscle contraction (55). The NMES has the advantages of being non-invasive, easily applied, painless, and causing minimal side effects (55). However, there are disadvantages with surface stimulation including accurate placement of surface electrodes in the appropriate position can be difficult, stimulating deep muscles may be impossible with surface electrodes, and children may not tolerate the surface stimulation at the intensity needed (55).

When the NMES was compared with conventional training, there were reportedly similar outcomes (56). A case report showed that the NMES application as a convenient home program seems to be an effective intervention for increasing muscle strength, improving functional motor skills and reducing spasticity in a child with spastic diplegia CP (33). Another preliminary study of the NMES compared to the volitional isometric strengthening showed that the quadriceps femoris and triceps surae muscles could be trained to have a greater force production in the NMES than that in the volitional isometric strength training after 12 weeks in children with spastic diplegia (53). However, there was a study showed that the NMES had no effectiveness on muscle strength (16). This recent study found that no difference was demonstrated between NMES or TES and placebo for either muscle strength or motor function after 16 weeks training (14). Therefore, the effectiveness of NMES on the quadriceps muscle strength in the ambulatory children with CP remains unclear.

The unique effects of ES have been attributed to several mechanism, most notably a reversal of the recruitment pattern typically associated with voluntary

muscle activation (57). However, the preferentially recruit fast versus slow motor units has been suggested for many years (24, 57). The present literature has concluded that the ES recruits motor units in a non-selective, spatially fixed, and temporally synchronous pattern and that muscle fibers are recruited without obvious sequencing related to fiber types (57).

3.3 The combination of NMES and voluntary exercise

The NMES may be operated to produce muscle strength augmentation including via a mechanism similar to that involved in voluntary exercise by presenting the muscle with an increased functional load. It specifically induces the activity of large motor units (type II) more effectively than volitional exercise and which are more difficult to activate during voluntary exercise (24). Moreover, it improves co-ordination between different agonist muscles and reduces co-activation of the antagonist muscles (58). Therefore, a combination technique of the NMES and isometric exercise may be the best for strengthening quadriceps in children with CP because the NMES recruits larger muscle fibers earlier and facilitates to isolate the required muscles, whereas the isometric exercise recruits smaller fatigue resistant muscle fibers first and assists the child with CP to learn voluntary muscle exercise.

Pawielski et al (23) failed to find significant effect in increasing of the right quadriceps strength in isometric strengthening with the NMES at home in a child with spastic hemiplegia. This strength training program performed in every other day for six weeks. The results demonstrated that the strength of the quadriceps did not change at the after end of training, which might be due to the limitation of single subject (23). Conversely, previous studies obtained a significant effect to increase of

muscle strength on voluntary exercise plus electromyostimulation (EMS) in healthy subjects, athletics, and patients with knee surgery (58-60). They found that the combined between voluntary exercise and EMS induced a neurogenic facilitatory effects enabling greater strength development to the recruitment of supplementary motor unit than voluntary exercise or EMS alone (59). Therefore, the combination technique of the NMES and isometric exercise in children with CP, especially spastic diplegia required more evidence to support.