Integrating Electrical Stimulation into Pediatric Cerebral Palsy Rehabilitation

An Evidence Based Rationale for Application and Practice for the (Reluctant) Clinician

Electrical Stimulation (ES) involves the therapeutic application of electrical impulses of sufficient intensity to induce skeletal muscle contractions in patients with neurological or musculoskeletal injuries. It is one of the most studied interventions with measurable clinical outcomes, and is applicable to all fields of rehabilitation. The earliest documented case of using ES with children was in 1976 (Gracanin et al., 1976). Though it is widely used in children with peripheral nerve lesions, it has yet to become a popular modality of choice for children with central nervous system injuries (Alon, 2016).

This article will describe how ES acts on the neuromuscular system, will elaborate on its utility with the pediatric population, and will overall describe a paradigm shift with respect to the applications techniques, parameters and approach in children with Cerebral Palsy (CP). We will focus on two non-invasive cutaneous applications, Neuromuscular Electrical Stimulation (NMES) and Functional Electrical Stimulation (FES).

Physiology of ES:

In order to design treatment protocols with ES it is beneficial to understand the neuromuscular response to ES, and how these muscular contractions differ from physiological contractions. **In physiological contractions**, motor units (MU) are recruited by two mechanisms, spatial and temporal. Spatial recruitment relates to the **number** of motor units recruited, while temporal recruitment relates to the **firing rate or the frequency** of the MUs recruited, the summation of which creates muscle tension. Both mechanisms work concurrently in proportion to the force applied during a contraction. The order of recruitment based on the **size of the MUs** is described by the 'Henneman size principle'. This principle states that small and typically slow MUs are recruited before large and typically faster MUs during a contraction (Gregory & Bickel, 2005). Of the three types of MUs (Type I, Type II a and II b), Type I is recruited first, and when additional force is required, Type II units are recruited next. To summarize, the motor unit activation in physiological contraction is varied and asynchronous. To maintain muscle activity and prevent fatigue, there is a switching between active and inactive motor units by modulation of firing rates (Bosques et al., 2016) or by activation of additional MUs at lower firing frequencies (Gregory & Bickel, 2005).

As compared to physiological contractions, **electrically generated muscle contractions** may be more likely to cause muscle fatigue.

nerves), spatially fixed (number of MU) and temporally(firing frequency) synchronous pattern with no relation to the muscle fiber selectivity (Gregory & Bickel, 2005).

The physiological benefits of electrical stimulation have been well documented. ES depolarizes the peripheral sensory and motor nerves causing physiological responses including muscle contraction, joint motion, increased peripheral blood flow, reduction of edema, and connective tissue mobilization, while simultaneously causing alterations in spinal cord and brain activation (Alon, 2013). Peripherally, it has many positive effects including increased strength, improved motor control, improved range of motion, reduced pain, and prevention or reversal of disuse atrophy (Alon, 2013; Bosques et al., 2016). Centrally, it is known to drive neuroplastic changes, including neural recovery following injury. Advances in science have demonstrated that the CNS has the potential to regenerate (Hamid & Hayek, 2008), and the neuroplasticity related to ES occurs due to motor region reorganization and motor relearning with repeated peripheral electric input (Schuhfried et al., 2012), assisted by the sensory input via the afferent nerves to the spinal cord and brain.

There are a number of proposed mechanisms to explain the therapeutic benefits of ES in CNS lesions. The bidirectional conduction (anterograde and retrograde) of action potentials due to stimulation of the sensory and motor fibers of the muscle are responsible for the concurrent effects. The direct peripheral sensory nerve stimulation via the spinothalamic tracts stimulate the sensory cortex while the efferent responses from muscle contractions stimulate the Golgi tendon organ, proprioceptors and other receptors causing multiple area activation in the brain via the multi synapse connections (Alon, 2013). Functional magnetic resonance imaging (FMRI) studies have shown that prolonged, consistent afferent inputs from ES additionally activate the somatosensory cortex, the supplemental motor cortex and the corticospinal connections including the cerebellum (Schuhfried et al., 2012). In addition to reorganization at the brain level, there is a local effect that leads to a reduction in spasticity. This may occur via stimulation of the opposing non-spastic muscle which activates the spinal interneurons and causes reciprocal inhibition (Schuhfried et al., 2012). Alternatively, the sub-motoric sensory stimulation may cause an inhibitory effect on spasticity due to alpha motor neuron excitement and sensorimotor reorganization (Schuhfried et al., 2012).

NMES and FES:

While the literature describes multiple nomenclatures for varied ES applications, the emphasis of this article will be on NMES and FES. As described by Kerr & McDowell (2007), NMES is the application of an electrical stimulation of sufficient intensity to elicit a muscle contraction. When this stimulation is used in a task specific manner by eliciting muscle contractions during a functional activity, it is referred to as FES. During a cutaneous application, the electrodes are placed along the length of the targeted muscles to cover the maximum bulk. Muscle contraction occurs via stimulation of the intramuscular branches of the motor nerve supplying the muscle. With NMES, the muscle contraction and relaxation is cyclical within the determined parameters and duration. With FES, the muscle contraction occurs only when triggered during the functional task.

Research for Implementation of ES Modality in Cerebral Palsy Rehabilitation:

The physiological and functional benefits of ES for individuals diagnosed with CP include increased muscle mass and strength, reduction in hypertonicity (spasticity), improvement of passive and active range of motion, reduction of co-contraction, and improvement in walking speed and gait mechanics (Doucet et al., 2012; Kerr & McDowell, 2007).

ES for Hypertonic (Spastic) Muscles: Muscle spasticity and muscle weakness co-exist in children with central lesions, and this is commonly seen in patients diagnosed with CP. The weakness is due to a lack of excitatory input from the damaged pathways causing fewer motor units to activate during muscle contractions. This limited activation is seen in both spastic and opposing non-spastic muscles leading to deficits in movements and gait (Rose & McGill, 2005). Studies have shown that spastic muscles are weak muscles and will benefit from strengthening (Stackhouse et al., 2007). The literature describes application of ES to both the opposing non-spastic muscles (Arya et al., 2012) and on spastic muscles (Carmick, J., 1997; Seifart et al., 2009). The short term reduction in spasticity creates a window of opportunity to implement other forms of therapy. Many studies have found that applying ES to specific muscles in children with CP reduces spasticity leading to functional improvements. For example, reduction of hip adductor tone and scissoring pattern was noted with stimulation of bilateral hip adductor and abductor muscle groups during gait training (AlAbdulwahab, 2011).

It is important to note that ES can also be applied to muscles that have received BOTOX (P. A. Wright & Granat, 2000). The use of BTX-A as an adjunct treatment to NMES may enhance the treatment effects by temporarily reducing muscle tone (Bosques et al., 2016; Schuhfried et al., 2012), however, the effects of Botox are known to spread beyond the target area. ES can also help limit the number of orthopedic procedures in CP. In a comparison study of children who received traditional orthopedic procedures versus minimal surgeries combined with FES during gait training found that the FES group underwent an average of 4.5 fewer ablative procedures (Johnston et al., 2004).

ES for Strengthening: Initial gains in strength in progressive resisted exercises (PRE) is due to an improved ability to recruit motor units, and later gains are due to an increase in the cross sectional area of the muscle (Reed, 1997). Children with cerebral palsy have difficulty generating muscle force due to reduced CNS motor unit recruitment and discharge rates, increased antagonist co-activation during agonist contractions, and changes in muscle morphology, including atrophy (Stackhouse et al., 2007). The longstanding belief that strengthening muscles impaired by CNS dysfunction would increase spasticity has been disproved by decades of research. An early study showed that children with spastic CP provided with a strengthening program, demonstrated improved strength without increased tone (Damiano & Abel, 1998). In a review of 10 empirical studies, Dodd and colleagues (2002) found enough evidence to establish strength-training programs (with free weights, isometric, isokinetic, concentric and eccentric exercises) improve muscle strength in spastic and non-spastic muscle groups in children and young adults with CP, with no increase in spasticity.

13-year-old child diagnosed with CP based on the overload principle of strength training. Post intervention, this child demonstrated increased isometric strength in the quadriceps, as well as decreased hamstring spasticity, improved spatiotemporal parameters of gait and increased functional abilities. Pool et al., (2016) applied NMES to the ankle dorsiflexors of children with CP during ambulation to assess changes in strength in the spastic muscles. Following 8 weeks of intervention increased muscle strength and volume were noted in the tibialis anterior as well in the medial/lateral gastrocnemius.

ES for Gait Training: The use of ES in children with CP for gait has undergone significant modifications over the years, and has progressed from eliciting reliable muscular contractions using NMES to triggering multiple muscles throughout the gait cycle using FES (Mooney & Rose, 2019). Improvements have been noted in walking speed, cadence, physiological cost index of walking (Arya et al., 2012; Mukhopadhyay et al., 2017), ankle propulsive force linked to an improvement in GMFM scores (J. Carmick, 1993a), and improved temporal-spatial parameters of gait measured with computerized dynamic posturography (AlAbdulwahab, 2011). Reports of the benefits of FES as an intervention to improve gait in children with CP have been upheld in two systematic reviews (Cauraugh et al., 2010; Mooney & Rose, 2019). ES of the triceps surae and medial/lateral hamstrings, with or without stimulating the opposing non-spastic muscle, has been shown to improve gait parameters and gross motor function (Carmick, J., 1993a). Stimulating the gastrocnemius also leads to improved joint mechanics and alignment in gait (Carmick, J., 1993a). When FES was applied to the post-tibial muscles in children with spastic unilateral CP, they demonstrated improved propulsive capacity during push off (Gonçalves et al., 2019), and symmetry of gait with improved foot contact (Durham et al., 2004). Additionally, stimulating the post-tibial muscles leads to improved swing phase kinematics with improved foot clearance (Philip A Wright et al., 2012). It has also been noted that FES training during gait may lead to other functional gains. In a study by Khamis et al., (2015) an intervention of walking with FES on the quadriceps in a 18 year old boy with a crouch gait led to an increase in the patient's knee extension during stance phase, and carry over to a stair negotiation activity with an improved step through pattern.

ES for Posture and Balance Training: Two case-control studies were conducted in which children diagnosed with CP either received conventional therapy, or conventional therapy combined with FES to their abdomen and lumbar regions. In the FES group there was a greater improvement in posture and dynamic sitting balance. Additionally, an increase in GMFM score and changes in radiological measurements of the kyphotic and Cobb angles were noted in the FES group when compared to the control group (Karabay et al., 2012; Park et al., 2001).

ES for Upper Extremity Function: Case studies demonstrating the feasibility of applying NMES to the upper limbs of children with CP have been reported since the 1990s. Studies have found that NMES improves upper extremity function in children with cerebral palsy, including the thumb, hand, and wrist (Carmick, J., 1993b), due to improved muscle length, reduction in tone and improved muscle strength (Bosques et al., 2016). For example, FES applied to the wrist extensor muscles in 8 children with CP improved hand function as measured by the Jebsen Test, with effects lasting up to 6 weeks (P. A. Wright & Granat, 2000). In another study, NMES alternatingly applied to the long flexors and extensors of the wrist of 8 children with CP over the course of 3 months resulted in improved active wrist extension (Kamper et al., 2006).

Limitations in research studies and its implication:

Although there is a tremendous literature base supporting the use of ES, there are a number of limitations. At this time there continues to be a lack of standardization of the nomenclature to describe different types of ES, which can lead to confusion about how to clinically translate reported protocols. Additionally, there is no established consensus about optimal parameters that are tolerable and have a therapeutic impact. Lack of consensus on optimal dosage (the frequency, pulse width, intensity/amplitude of the electric input, session treatment duration and the duration of the entire study trial) creates conflict in clinical decision making. Another limitation relates to outcome measures and long term follow up. In most studies the outcome measures assess the impairment level as opposed to function or participation levels of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) model (Franki et al., 2012). Studies that assess the long term benefits of ES are also lacking. Overall, most studies are rated as low quality due to the limited number of subjects, heterogeneity of the subjects and poor methodologies. Stanger & Oresic (2003) found that the studies on NMES for children with CP mostly report evidence levels of III (Cohort and case-control studies) and V (case series, studies with no control) based on the 'Levels of Evidence from Sackett' (Burns et al., 2011).

Despite these limitations, the existing literature do support ES as a safe and feasible treatment option for children diagnosed with CP, with impact seen at both impairment and functional levels. In fact, a recent study by Novak et al.,(2020) using the Evidence Alert Traffic Light Grading System tool has given ES treatment modality as green signal or "go" due to high quality evidence supporting this intervention. Jewell, (2014) states "Evidence-based physical therapy practice is an open and thoughtful clinical decision making process integrating the best available evidence with clinical judgement, which includes the literature, expert opinion and personal experiences"(p. 35). Therefore, it is incumbent upon each therapist to assess and interpret the literature and appropriately apply findings to patient management.

Foundational Theoretical Constructs for ES as a Rehabilitation Modality:

Principles of Motor Learning: In order to understand how ES drives long term functional changes, we should consider the theoretical principles of motor learning. Motor learning theory explains our system's ability to improve motor actions through practice leading to acquisition of skills for functional recovery after brain lesions. It states that for functional recovery to occur, practice must be repetitive, goal-oriented, and at the limit of performance (Merrill, 2009). Motor learning reflects a relatively permanent change in the motor skill, conventional therapy due to limited active participation (Chiu & Ada, 2014). Integrating ES into a plan of care over an extended period of time can lead to the permanent changes that we strive to help our patients gain.

ICF model: Another theoretical construct to consider when integrating ES into a plan of care is the ICF model. This model can be used as a conceptual framework to set patient goals and to measure practical and meaningful outcomes. Stanger & Oresic (2003) noted that most studies reporting on the efficacy of NMES have measured improvements on the impairment level but there has not been a focus in literature on functional changes in children with CP. This in fact should be the emphasis of integrating ES into rehab programs, as functional gains lead to greater independence, social participation, and integration into society on the community level (Abbaskhanian et al., 2015).

Entire Life Participation: An entire life perspective in terms of safety during mobility, prevention of pain, and maintenance of function should be the cornerstone of rehabilitation goals (Bottos et al., 2007). Adults with developmental disabilities, specifically with CP, have a high incidence of musculoskeletal pain, and most of these individuals stop walking by the age of 25 (Andersson & Mattsson, 2007). One study reported that children and youth with CP GMFCS Levels III-V are at the greatest risk of losing motor function, with the greatest decline seen in GMFCS level IV, due to a combination of physical growth, increased energy costs and persisting impairments (Hanna et al., 2009). Additional factors like biomechanical forces, immobility leading to excessive physical stress and strain, overuse syndromes, early joint degeneration (Bottos et al., 2007), and failed orthopedic surgeries (Dan, 2007) also lead to deterioration of walking in this population. In an effort to help individuals diagnosed with CP maintain their function throughout the lifespan, ES should be considered as the conservative treatment modality of choice, as it can potentially offer varied long term impact in a cost effective manner, before automatically defaulting to more invasive treatments.

Clinical Considerations When Integrating ES into Pediatric Rehabilitation:

Treatment Protocols for Children: The ES parameters used for adults were established based on the principle of electrical contractions mimicking voluntary tetanic contractions which are of frequency 30-50pps and a pulse duration of 200-300us (Gregory & Bickel, 2005; Reed, 1997). When these protocols are applied to pediatric patients, there is poor tolerance and compliance with the ES treatment program. Over the past few decades other parameters have been explored by clinicians and researchers, including Alon (Alon, 2019; Motavalli et al., 2019) & Hastings (Hastings, S., 2018). In children, a tetanic contraction can be achieved with frequencies and pulse durations as low as 5-7Hz and 48-52 μs respectively. The intensity or amplitude is increased until a visible or palpable contraction is obtained. These child-friendly protocols lead to improved tolerance, compliance, function, and impairment reduction. Every protocol should be individually tailored as per realistic goals set by the parents, patient, and therapist.

Safety of ES Treatment Modality for Children:

Studies have found ES to be safe and well tolerated in children with various disabilities with no adverse effects (Bosques et al., 2016). ES has been reportedly used in children as young as 24 months of age (J. Carmick, 1993a; Karabay et al., 2012; Park et al., 2001). Medical clearance should be obtained for children with complex medical diagnoses including seizure disorders, shunts or cardiac issues.

Recommended Dosage:

Based on clinical experience, ES protocols are most successful when implemented 4-5 times a week. The specific dosage, and the number of muscles to be targeted will vary and be determined by the therapist based on the child's level of function and therapeutic goals. Changes including reduction in tone, improved posture, and improved components of balance can be seen within weeks. However, motor learning research has demonstrated that it takes months, and sometimes years of practice for a child to achieve functional changes (Alon, 2013). It is important to consider that factors including growth spurts and illness can impact the rate of change as well. Additionally, to optimize impact of intervention, carryover throughout the day at home, school, and work is important, and is often implemented with recommendations for compressive garments and orthoses for correct biomechanical alignment. Consistency with the treatment protocol and integrating ES as part of a home exercise program are essential and can hasten progress.

Recommended ES Unit:

It is advantageous to use an ES unit that offers wide range of parameters (i.e. pulse frequency, amplitude, etc) to design individual treatment protocol. Units with remote/hand-held triggers can be used for FES protocols, and small, portable units are easiest to use for stimulating multiple muscle groups. When using FES for gait training, a feature that automatically switches to the opposite channel when the remote switch is released is highly beneficial, as stepping occurs rapidly in gait. Safety features like an automatic lock of parameters that prevents the child from making adjustments, and an automatic shut down if any components get loose are optimal as well.

Specific Application Considerations for Cerebral Palsy:

The goals of integrating ES into the rehab program for children with CP include (1) spasticity reduction (2) reduction of co-contraction of agonist/antagonistic muscles; (3) improving active and passive muscle length (4) strengthening of muscles (spastic, opposing non-spastic and hypotonic muscle) (5) improve motor control and (6) improve coordination (Merrill, 2009). Clinical decisions about which muscle groups to target should be based upon the biomechanics of an activity, the developmental age of the patient, and whether or not the consider any muscle that is a candidate for a tone reducing intervention, such as BOTOX, or surgery, to be a candidate for ES.

A recent systematic review with a meta-analysis of 6 randomized controlled trials in which NMES was combined with other conventional therapies reported medium to large effect sizes for improved gross motor function, including sitting and standing, in children with CP ES is potentially appropriate for use with all levels of functioning across the GMFCS levels (Salazar et al., 2019). For example, for patients presenting at GMFCS levels IV-V, ES can be provided to reduce hypertonicity to help families perform caregiving and hygiene tasks (i.e. diaper change, bathing), to improve posture (in wheelchairs, standers, and gait trainers) and to assist with breathing. For patients functioning within level I-III, ES can be implemented to assist in skill development including floor mobility skills, transitional mobility skills from the floor to stance, stance control, and coordinated, energy efficient walking. Children at GMFCS level II can benefit from integration of ES protocols for specific and intense training to improve higher level balance skills including single limb stance, controlled and varied jumping skills, and independent stair negotiation.

Conclusion:

Electrical stimulation is a well-studied, safe, and effective modality for use in pediatric cerebral palsy rehabilitation programs, yet it is not largely implemented as a standard intervention. Considering the evidence that electrical stimulation facilitates improved function when used in conjunction with conventional therapy, the strong physiological and theoretical foundations of this modality, as well as its cost effective nature, pediatric therapists should feel confident to integrate this intervention into their repertoires when working with individuals diagnosed with cerebral palsy.

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Jovial Lewis PT, MPT is a licensed physical therapist working exclusively in pediatric rehabilitation. She works for Early Intervention Services and with St Mary's Children's Hospital Home Care program, NYC. She did her Bachelor's and Master's program with specialty in Neurological Sciences at Manipal Academy of Health Sciences (MAHE) , India. She has worked as an Assistant Lecturer/Clinician in India for 5 year later transitioning as full time clinician for the past 15 years in NYC. She started using E-stim in 2017 in children with cerebral palsy and developmental delay and can say that it has transformed her practice. Yocheved Bensinger-Brody PT, PhD, PCS is a board certified pediatric physical therapist. She is an Assistant Professor in Touro College's Doctor of Physical Therapy program in NYC, and she is the clinician owner of a pediatric practice in northern NJ, Boutique Pediatric PT. Yocheved graduated with a BS in Physical Therapy from Florida International University, an MA in Movement Sciences from Teachers College, Columbia University, and a PhD in Psychology from the Graduate Center at CUNY. Yocheved started integrating E-stim into her pediatric practice a few years ago and has found it to be a game changer!

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