THESIS

RHYTHMIC CUEING EFFECTS ON GAIT PARAMETERS OF CHILDREN WITH SPASTIC DIPLEGIC CEREBRAL PALSY: AN IMMEDIATE ENTRAINMENT STUDY

Submitted by
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In partial fulfillment of the requirements
For the Degree of Master of Music
Colorado State University
Fort Collins, Colorado
Fall 2009

COLORADO STATE UNIVERSITY

July 22, 2009

WE HEREBY RECOMMEND THAT THE THESIS PREPARED UNDER OUR SUPERVISION BY LAURA CLARICE JENSEN ENTITLED RHYTHMIC CUEING EFFECTS ON GAIT PARAMETERS OF CHILDREN WITH SPASTIC DIPLEGIC CEREBRAL PALSY: AN IMMEDIATE ENTRAINMENT STUDY BE ACCEPTED AS FULFILLING IN PART REQUIREMENTS FOR THE DEGREE OF MASTER OF MUSIC.

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ABSTRACT OF THESIS

RHYTHMIC CUEING EFFECTS ON GAIT PARAMETERS OF CHILDREN WITH SPASTIC DIPLEGIC CEREBRAL PALSY: AN IMMEDIATE ENTRAINMENT STUDY

This study was designed to examine whether children with spastic diplegic cerebral palsy can synchronize (entrain) their gait patterns with an external auditory rhythmic stimulus, and whether this rhythmic stimulus would encourage improvement in stride symmetry and knee extension at foot contact during the gait cycle. Five participants completed an immediate entrainment protocol that included a self-selected normal speed walk (SS), a self-selected normal speed walk matched with music of the same cadence (SSM), a fast walk (F), and a fast walk matched with music of the same cadence (FM). Results indicated that the participants synchronized their gait patterns with the rhythmic music stimulus, but no significant results were observed for stride symmetry or knee extension measures between no-music and music conditions. The ability to entrain to a rhythmic stimulus suggests that gait training facilitated by rhythmic cueing may be an appropriate gait habilitation technique for this population, but that research with a larger number of participants is necessary before definite conclusions in this area can be drawn.

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ACKNOWLEDGEMENTS

I have difficulty finding the words to express the gratitude I feel to so many for helping to make my graduate experience rich, rewarding, and successful. The following people have provided unending support throughout this education process:

Advisors Dr. William Davis and Dr. Michael Thaut – Thank you for providing this music therapy program, which has truly changed my life, and has provided me with a stimulating, challenging, and rewarding profession that I am excited to pursue for many years.

Dr. Blythe LaGasse – advisor, colleague, and friend. You suggested this fascinating thesis topic, and then (thank goodness) you lapped me. Finishing this project without your tireless assistance, support, and feedback would have been exceedingly difficult. Thank you for sharing your love of this field with me, for a tremendous academic and clinical education, and for your support in all things. I'll miss working and not working with you here in FC.

My outside committee member, Dr. Brian Tracy – Your excitement about Neuromuscular Aspects of Human Movement is infectious, even at 8am. Thank you for your sincere support, for your interest in learning about the field of neurologic music therapy, and for bringing your expertise to your conscientious critique of and suggestions for this research thesis.

To the research team at the Center for Gait and Movement Analysis, particularly Dr. James Carollo, Sarah Recktenwall-Work, and Timothy Nicklas. Thank you for making this thesis project a reality, and for the many hours spent collecting and processing data. Sarah, this would never have happened without your tireless assistance and your willingness to explain, again, at any odd hour. Best of luck on your medical school journey - you will make a wonderful physician. Jim and Tim, I look forward to hearing about the results of the entire study, and to other research that comes out of your remarkable lab.

Dr. Ruth Rice – I think of our trips to Denver and beyond with great fondness, and thank you for your friendship, constant encouragement, and sharing of your knowledge during this thesis project and throughout my time at CSU.

Gary Kenyon – Where would this project be without the Teddy Bear's Picnic? Thank you for creating music with me and for the project, for your willingness to explain computations at length, and for your great patience and kindness.

Thank you to the participants and families, who kindly gave their time and energy so that the research proposal could become an actuality. I hope that this and other projects will positively inform treatment possibilities for children with cerebral palsy.

James zumBrunnen – I could not have completed the analysis of the data without your statistics expertise and your patient explanations. Thank you for your kindness.

Sarah Johnson – Autoharp and NMT guru, internship supervisor, unfailing friend. Your incredible energy, enthusiasm, willingness to share your vast knowledge and skill, and belief in me I will carry with me always. My experiences with you on the LSR and elsewhere during my internship remain invaluable to my professional and personal development, and I thank you from the bottom of my heart.

To the strong, talented, lovely ladies of IMPACT dance – thank you for your love and encouragement for the last four years. Your safe haven of friendship, support, and creativity helped me hold my head high and strive for the grace of a dancer in all things.

Brian Jensen and Lisa Rahangdale Jensen – You are remarkable people, and I admire and love you both. Thank you for your constant love, support, and willingness to help me through the hard parts.

Penny and Eric Jensen – What gifts you have given me, with your music, science, interests in health and healing, and abilities to care so deeply for others. Words will never be enough to thank you for the constancy of your love, support, assistance, belief, and guidance as parents and friends. I am exceptionally fortunate.

Nicholas Norberg – Thank you for your inexhaustible patience, generosity, love, humor, and friendship. Your understanding of and respect for science has helped me appreciate the research process, and your unswerving belief in me has kept me going when I wanted to stop. I am excited to walk forward together, side by side.

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CHAPTER 1: INTRODUCTION

Purpose

The purpose of this study was to determine whether children with spastic diplegic cerebral palsy synchronize with, or entrain to an external, auditory rhythmic stimulus, and if so, how this immediate entrainment affects various temporal-spatial and kinematic measures of gait in these children. The researchers used Rhythmic Auditory Stimulation (RAS) to provide the rhythmic stimulus in this study. Previous research on rhythmic cueing techniques such as RAS has indicated that improvements in gait occur with immediate rhythmic entrainment in other populations, including stroke (Mauritz 2002; Prassas, Thaut, McIntosh, & Rice, 1997; Thaut, McIntosh, Prassas, & Rice, 1992a, 1993), Parkinson's disease (Freedland, Festa, Sealy, McBean, Elghazaly, Capan et al., 2002; McIntosh, Brown, Rice, & Thaut, 1997; Suteerawattananon, Morris, Etnyre, Jankovic, & Protas, 2004), and traumatic brain injury (Hurt, Rice, McIntosh, & Thaut, 1998). Additionally, entrainment has been shown to occur in two RAS studies with children with cerebral palsy (Kwak, 2007; Thaut, Hurt, Dragon, & McIntosh, 1998).

Need/Problem

Background on cerebral palsy

Cerebral palsy affects an estimated two of every 1,000 children born each year (Sankar & Mundkur, 2005). An umbrella term, 'cerebral palsy' refers to "a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of its development" (Shevell &

Bodensteiner, 2004, p. 2). While children with cerebral palsy may manifest varying levels of cognitive, visual, auditory, emotional, speech and/or seizure difficulties, traditional classifications of the disorder refer to motor impairments of both movement and posture.

Incidence of cerebral palsy has increased in conjunction with the decline in infant mortality rate that has resulted from advances in Western medical practices. More preterm babies live; however, premature birth is one of the main risk factors for development of cerebral palsy. These infants are born with underdeveloped and vulnerable central nervous systems (Sankar & Mundkur, 2005). The small organs associated with such underdeveloped systems and with low birth weight may lead to difficulties with adequate oxygen supply to the brain ("Cerebral Palsy," 2009). Alternatively, premature birth may result from infection in the mother during pregnancy, or an infection in the infant shortly after birth, in which case the fetal or infant central nervous system injury may be caused by the infection rather than by a lack of oxygen (Stoll et al., 2004).

There are several other prenatal, perinatal, and postnatal risk factors for cerebral palsy. Prenatal risk factors are maternal infection, seizures or hyperthyroidism as well as placental difficulties or exposure to teratogens. Perinatal problems include infections, head trauma, low blood sugar levels, anoxia, and extreme jaundice. Postnatal complications may be head trauma or infections such as meningitis or encephalitis (Sankar & Mundkur, 2005).

Early diagnosis and comprehensive intervention is recommended for the most effective treatment and habilitation of cerebral palsy. Appropriate treatment and management can help reduce deficiencies and prevent development of contractures and severe bony deformities. Additionally, early diagnosis gives the infant and the team of caregivers the greatest opportunity to habilitate associated deficits (Scherzer, 2001).

Therapeutic needs / Habilitation background on cerebral palsy

The three predominant causes of gait deviation in cerebral palsy are: "(1) loss of selective motor control, (2) impaired balance, and (3) abnormal tone" (Gage, 2004, p. 182). Selective motor control is defined as "[the] ability to isolate the activation of muscles in a selected pattern in response to demands of a voluntary posture or movement" (Sanger et al., 2006, p. 2159). Because the basal ganglia house memories of previous movement patterns, children with dyskinesia secondary to basal ganglia injury experience extreme loss of selective motor control (Gage, 2004, p. 182). Therapeutic techniques that help these patients gain some control are therefore of primary importance.

Because cerebral palsy manifests in a myriad of ways, no one therapy treats all types or aspects of the disorder. A variety of physicians and therapists have developed or implemented several different treatment approaches, including muscle education and braces, progressive pattern movements, proprioceptive neuromuscular facilitation (PNF), neuromotor development, neurodevelopmental treatment with reflex inhibition and facilitation, reflex creeping and other reflex reactions, and Conductive Education (Levitt, 2004).

In addition to these different therapeutic ideologies, many children with cerebral palsy participate in treadmill walking or strength training, and some undergo single or multilevel surgeries. Surgeries may include a selective dorsal rhizotomy in an attempt to sever nerves that produce irregular motor responses, or muscle and tendon procedures to help relieve contractures (Scherzer, 2001). Often, children with spasticity receive local

Botulinum toxin A (Botox) injections, take prescription oral medication, or are administered intrathecal baclofen via an implanted abdominal pump to allow hypertonic muscles a period of some relaxation.

Rhythmic Auditory Stimulation and the Therapeutic Needs of Cerebral Palsy

As is stated previously, loss of selective motor control is a salient problem for children with cerebral palsy. This lack of control results in the inability to direct individual joints and associated muscles, preventing streamlined, healthy gait patterning. Instability and abnormal tone can affect stride symmetry as well as stride length, cadence, velocity, and efficient muscle recruitment.

Research has shown that Rhythmic Auditory Stimulation (RAS) facilitates improvements in cadence, stride length, velocity, stride symmetry, joint range of motion, and muscle recruitment patterns for stroke (Mauritz 2002; Prassas et al., 1997; Thaut, McIntosh et al., 1992a, 1993), Parkinson's disease (Freedland et al., 2002; McIntosh et al., 1997; Suteerawattananon et al., 2004), and traumatic brain injury (Hurt et al., 1998). While the etiology of the lesions for these patients may be different than those arising from cerebral palsy, some of the effects of this central nervous system damage on gait and movement are similar to those experienced by children with cerebral palsy. It follows that RAS would be a plausible gait habilitation method for individuals with cerebral palsy.

Rationale

Researchers have demonstrated repeatedly that rhythm is an integral part of the human experience (De Montigny & Lamarre, 1973; Molinari, Leggio, Filippini, Gioia, Cerasa, & Thaut, 2005; Nettl, 1983; Rider & Eagle, 1986; Thaut, Kenyon, Schauer, &

McIntosh, 1999). Anthropologists and ethnomusicologists have long recognized rhythm in both music and dance to be a fundamental aspect of all cultures. Nettl (1983) suggests that "the close association of dance with music everywhere makes the idea of rhythm and physical movement as generative forces of music tentatively credible" (p. 165), indicating that the rhythmicity of human movement was a creating force for the rhythmicity of music. He goes on to say that "[musicologists interested in comparative complexity] do not know whether a strong, repetitive metric structure is to be regarded as primitive or as the crowning achievement of a long evolution…" (p. 170), demonstrating that rhythm is inherent in all human existence. Epstein (1985) discovered that people engaging in music across many different cultures modulated tempo based in proportions of small integers, revealing that tempo may be a human phenomenon as well. Rider and Eagle (1986) suggest that auditory rhythmic entrainment may be an element in learning.

Various studies have examined the connection between the auditory and the motor systems. De Montigny and Lamarre (1973) showed that rhythm is processed in several different places in the central nervous system of a cat, including the inferior olivary nucleus of the medulla, the cerebellum, and the reticular and vestibular nuclei of the brainstem. Pal'tsev and El'ner (1967) found that a sound stimulus increases the intensity of the patellar reflex, revealing that sound does not have to be processed by the cerebral cortex in order to affect body movement. Rossignol and Melvill Jones (1976) had the same results with the H-reflex of the knee flexor muscles. They discovered that the amplitude of the reflex increased in response to a sound stimulus after a latency period of approximately 80 msec.

Cohen (1999) explains that central pattern generators (CPGs), are neuronal circuits in the spinal cord that allow us to perform rhythmic activities such as walking, breathing, and chewing without cortical input. The existence of such CPGs, in combination with the connections between the auditory and the motor pathways discussed previously, suggests that the rhythmic activity of walking could be enhanced by an auditory stimulus, even when damage to cortical structures exists.

Thaut, et al. (1999) found that during a rhythmic finger-tapping task to an auditory stimulus, healthy participants entrained to the period between beats rather than to each phase of the sound. The participants self-corrected in response to period shifts that were below the conscious level of perception, once again entraining to the rhythmic stimulus. Molinari, et al. (2005) discovered that during the same type of finger-tapping task, patients with cerebellar lesions entrained as successfully as healthy participants to such consciously imperceptible period shifts, corroborating the evidence that rhythm is processed by more than one location in the central nervous system (De Montigny & Lamarre, 1973). Thaut (2003) reported further substantiation that rhythm is processed in many different areas in the central nervous system. Brain imaging studies showed the following:

> activated regions include[d] primary sensorimotor and cingulate areas, bilateral opercular premotor areas, bilateral SII, ventral prefrontal cortex, and, subcortically, anterior insula, putamen, and thalamus. Within the cerebellum, vermal regions and anterior hemispheres ipsilateral to the movement became significantly activated. Tracking temporal modulations additionally activated predominantly right prefrontal, anterior cingulate, and intraparietal regions as well as posterior cerebellar hemispheres (p.364).

This research demonstrates that the human brain innately responds to rhythm and creates rhythm through movement. Evidence of connections between the motor and the auditory pathways of the central nervous system indicates that auditory cueing can help drive movement. Such evidence suggests that RAS could facilitate the rehabilitation of gait patterns in children with cerebral palsy.

Hypotheses

If this study produces statistically significant results, it will indicate that children with spastic diplegic cerebral palsy can entrain to an auditory rhythmic stimulus, and that the immediate entrainment effects of RAS can produce improved gait patterns in these children, both in temporal-distance parameters and in kinematic measures. In order to determine the statistical outcome of this research, the following null hypotheses have been established:

Null Hypothesis 1: Synchronization error and absolute period error will indicate that children with spastic diplegic cerebral palsy do not synchronize their steps or step periods with an external auditory rhythmic stimulus.

Null Hypothesis 2: On the temporal-distance measure of stride symmetry, there will be no difference between uncued walking and RAS-cued walking.

Null Hypothesis 3: On the kinematic measure of knee extension at foot contact, there will be no difference between uncued walking and RAS-cued walking.

CHAPTER 2: REVIEW OF RELATED LITERATURE

Cerebral Palsy

Diagnoses and Characteristics

Spastic cerebral palsy is the most common neuromuscular type of cerebral palsy, presenting in approximately 70-75% of cases (Sankar & Mundkur, 2005). Children with spastic cerebral palsy exhibit symptoms associated with upper motor neuron impairment, including increased tone, heightened stretch reflexes, and persistent primitive reflexes. Spastic cerebral palsy results from a lesion in the vestibular or reticular brainstem nuclei, the cortical tracts running to these nuclei, or the tracts running away from these nuclei (Gage, 2004). This type of lesion results in a lack of neural inhibition that in turns causes muscular hypertonia.

The second most common type of cerebral palsy is dyskinetic, accounting for 10-15% of cases (Sankar & Mundkur, 2005). Dyskinetic cerebral palsy includes chorea, athetosis, and other involuntary movement of the limbs and face. These symptoms may intensify under emotional stress or with attempts at intentional movement, indicating a variation in tone that may alternate between too low and too high (Sankar & Mundkur). Dyskinetic cerebral palsy results from lesions in the basal ganglia (Rudenberg, 1985) secondary to birth asphyxia (Sankar & Mundkur). Many children who eventually develop this type of cerebral palsy exhibit hypotonia during the first one to three years of age.

Ataxic cerebral palsy constitutes less than five per cent of cases (Sankar & Mundkur, 2005), and is described by decreased muscular coordination and difficulties with balance and equilibrium (Rudenberg, 1985). People with ataxic cerebral palsy may also experience visual and auditory processing difficulties, fine motor limitations, and tremors (“Cerebral Palsy,” 2009). This type of cerebral palsy usually results from a cerebellar lesion.

Pure spasticity, dyskinesia, or ataxia is rarely seen. Most children with cerebral palsy have a mixed neuromuscular presentation because of lesions in two or more areas in the central nervous system; however, diagnosis is determined by the predominant presentation. Approximately 86% of cases are congenital, occurring in response to pre- or perinatal disease or injury, while the remaining 14% of cases are acquired, resulting from injury or infection during early childhood (Rudenberg, 1985).

In addition to the neuromuscular description, cerebral palsy is classified according to how much of the body is involved. Most common is diplegia, accounting for 30%-40% of cases (Sankar & Mundkur, 2005), and usually involving the legs more severely than the arms. Hemiplegia, impacting one side of the body, occurs in 20%-30% of cases, while quadriplegia, affecting all four limbs and usually associated with greater secondary impairments, is present in 10%-15% of cases (Sankar & Mundkur).

Researchers have studied various defining characteristics of cerebral palsy, including neuromuscular activation, energy expenditure, and biomechanical aspects. In 2005, Rose and McGill published a study on neuromuscular activation and motor-unit firing characteristics of cerebral palsy. They measured muscular strength during both maximal and submaximal voluntary contractions, M-wave amplitudes in response to electrical stimulation, neuromuscular activation (calculated: electromyographic (EMG) signal/possible number of motor units that could be activated), and firing rate, recruitment, and short-term synchronization of motor units in participants with cerebral palsy and in typically developing controls. These researchers discovered that submaximal voluntary contractions were similar between the two groups, as were M-wave amplitudes. However, neuromuscular activation and motor unit firing rate, recruitment, and short-term synchronization of motor units during maximal voluntary contractions were significantly lower in participants with cerebral palsy. These results indicate that the weakness that accompanies spastic cerebral palsy could result from the decreased ability of the neuromotor commands to reach the skeletal muscles responsible for contraction.

Piccinini, Cimolin, Galli, Berti, Crivellini, and Turconi (2006) examined energy expenditure of ambulation in children with cerebral palsy. They found that both energy expenditure and oxygen cost were greater in participants with cerebral palsy than in their normal controls. These authors noted, as did Gage (2004), that their clients with cerebral palsy exhibited decreased selective motor control, abnormal muscle tone, difficulties with balance, and disorganized use of agonist and antagonist muscles. They suggested that oxygen cost increases during gait with CP because of out-of-phase co-contractions of agonist and antagonist muscles. While some co-contraction is necessary to stabilize joints, too much can cause instability and weakness, and can use energy inefficiently.

Children with cerebral palsy often exhibit biomechanical impairments such as tibial torsion. Hicks, Arnold, Anderson, Schwartz, and Delp (2007) examined the effect of this rotational abnormality on the ability of the participants' muscles to extend the hip and knee during gait. As the researchers state, in normal gait, the soleus muscle accelerates the ankle, which produces acceleration in the hip and knee as well. The authors suggest that excessive tibial torsion can interfere with this "plantar flexion-knee extension coupling" (p.2), decreasing the ability of the soleus to begin the acceleration chain of events that results in full extension of the hip and knee during single-leg stance.

Recognizing that not all types of cerebral palsy manifest in the same type or degree of impairment, Lebiedowska, Gaebler-Spira, Burns, and Fisk (2004) compared the biomechanical characteristics of spastic and dystonic hypertonia. They discovered that participants with spasticity had increased tendon reflexes, but less co-contraction than did participants with dystonic hypertonia. The latter group also exhibited slower gait, increased resistance to passive movement, more muscle weakness, and less range of motion at the knee; however, tendon reflexes in this group were almost normal.

This sampling of research illustrates the fact that "the neuromuscular mechanisms underlying motor deficits and gait disorders in CP are multifaceted" (Rose & McGill, 2005, p. 333). While many features of cerebral palsy are seen commonly, they are rarely combined exactly the same way in two different people. Because of this broad range of characteristics, patients with cerebral palsy have a variety of needs, and may require a synthesis of several different types of therapy.

Therapeutic Needs

Three of the major difficulties experienced by children with cerebral palsy are loss of selective motor control, impaired balance, and abnormal tone (Gage, 2004). In children who exhibit spasticity, the degree of selective motor control difficulty is determined by the amount of damage sustained by the central nervous system (Gage). Children with spastic cerebral palsy generally have greater selective motor control over their proximal joints and muscles than over the distal aspects of their extremities (Gage). Additionally, muscles that cross two joints (biarticular muscles) seem to be more affected by spasticity than those that cross one joint. As Gage (2004) indicated, this situation has implications for therapy. For example, the traditional Achilles tendon lengthening surgery to correct for gastrocnemius contracture actually lengthens both the soleus (a monoarticular muscle) and the gastrocnemius. However, the gastrocnemius, a biarticular muscle, is more affected than the soleus; therefore, the surgery can create rather than lessen gait difficulties (Gage).

Almost all children with cerebral palsy exhibit poor equilibrium. Causes for this difficulty include cerebellar lesion (in the case of ataxia), insufficient balance reactions because of high muscle tone or contracture, or impaired base of support due to decreased or fluctuating muscle tone, abnormal skeletal forces, or bony deformities (Gage, 2004). Falls may occur anteriorly, posteriorly, or laterally, and the most common method of prevention is to provide the child with an assistive device such as a walker, crutches, or orthotics (Gage). Surgery can help re-align joints and limbs, providing limited improvement by re-aligning the base of support; however, little else has been implemented to ameliorate this problem (Gage). In his *Orthopaedic Management in Cerebral Palsy* (1987), Bleck states, "Of all the motor problems in cerebral palsy, deficient equilibrium reactions interfere the most with functional walking" (quoted in Gage, p. 184).

Types of abnormal tone include hypertonia, dystonia, and hypotonia, and they result in spasticity, or rigidity of movement, chorea, and athetosis. All types of abnormal tone create gait deviations in children with cerebral palsy, but spasticity is the most common. Increased angular velocity around a joint intensifies rigidity, and thus resistance to movement (Gage, 2004). Gage (2004) states that spasticity:

> (1) acts like a brake on the system and this drag on movement increases energy consumption, (2) inhibits voluntary control of movement, (3) interferes with the stretch on muscles that normally occurs during activity and so inhibits growth, and (4) contributes to bony deformity of the growing skeleton by inducing excessive torques on long bones during gait (p. 185).

Use of external auditory rhythmic stimuli to facilitate movement has been shown to increase coordination of muscle fiber recruitment, thereby decreasing energy expenditure (Thaut, 2005). The use of rhythmic cueing in gait habilitation with children with cerebral palsy could therefore assist both with motor efficiency during ambulation and with delaying fatigue.

Cerebral palsy can produce many different gait deviations. These include internally rotated gait, excessive knee flexion that is often referred to as 'crouch gait,' excessive knee extension, internal foot progression, asymmetrical hip and pelvic rotation, and various increases or decreases of motion in all planes of all lower extremity joints (Ounpuu, in Gage, 2004). While a variety of techniques, including medication and surgery, can be helpful in the treatment of the gait deviations associated with cerebral palsy, these therapies are not always successful, and can be invasive. One of the limitations that is often seen but that has no successful therapeutic intervention is the inability to fully extend the knee at terminal swing (Center for Gait and Movement Analysis, 2008). The current proposed research project is intended to examine whether RAS can be used to improve this particular gait impairment, as well as other gait parameters and kinematic measures.

Treatment

W.M. Phelps developed the first modern cerebral palsy treatment ideology in the mid 1900's. He suggested that occupational, speech, and physiotherapists work together in groups, in order to more completely address the needs of children with cerebral palsy. He felt that proper treatment included a specific diagnosis for each child, appropriate use of a set of fifteen therapeutic modalities, braces or calipers, and muscle education (Levitt, 2004).

Phelps identified five different diagnoses as well as some sub-classifications. His fifteen treatment types included:

> [(1) massage for hypotonic muscles, (2) passive movement throughout a joint range, (3-4) active motion both assisted and without assistance, (5) resisted motion, (6) conditioned motion, (7) synergistic motion, (8) motion training at more than one joint, (9) relaxation techniques, (10) movement out of relaxation, (11) rest periods, (12) bi-lateral reciprocal motion, (13) balance, (14) reach, grasp, and release, and (15) activities of daily living] (Levitt, 2004, p. 14-15).

He designed braces and calipers to help with deformities, upright posture, and athetoid movements. Muscle education includes stretching, strengthening, and range of motion exercises (Levitt, 2004, p. 15).

Phelps advocated braces or calipers for all patients with cerebral palsy. While these sorts of orthoses are shown to help in circumstances, they may be of little use in others. Lucareli, Lima, Lucarelli, & Lima (2007) found that floor reaction foot orthoses (FRAFOs) assisted participants with moderate to severe crouch gait, but did not significantly help those with mild crouch gait.

Temple Fay advocated Progressive Pattern Movements developed from the evolution of motion in all species. He determined five stages, including prone lying, the homolateral stage, the contralateral stage, on hands and knees, and the walking pattern. Doman and Delacato added to Fay's ideas by suggesting the patients could benefit from CO_2 inhalations, limiting drinking of fluids, developing dominance of one cerebral hemisphere, spinning upside-down to stimulate the vestibular system, and hand walking while hanging from a set of parallel, horizontal bars (Levitt, 2004, p. 15-16).

Herman Kabat, Margaret Knott, and Dorothy Voss implemented the use of proprioceptive neuromuscular facilitation (PNF) in the treatment of cerebral palsy. This method employs flexion and extension, abduction and adduction, and internal and external rotational movement patterns, use of sensory stimuli, resistance to motion, functional mat work, and several more specialized techniques. These special techniques include irradiation, rhythmic stabilizations, stimulation of reflexes, repetitive contractions, reversal of movement patterns, and relaxation techniques (Levitt, 2004, p. 17).

Eirene Collis believed in neuromotor development, adhering to a strict protocol, and the use of the term 'cerebral palsy therapist,' instead of specifying physio-, speech, or occupational therapist. She felt that mental capability would dictate the therapeutic outcome, advocated early treatment, believed in 'management' training for activities of daily living, and thought that a strict developmental sequence should be followed. She would not allow a child to employ any motor skill that was above his or her level, all the while presenting a picture of normal tone, posture, and movement for the child to emulate (Levitt, 2004 p. 17-18).

Karl and Berta Bobath developed the Bobath technique, defined as Neuro-Developmental Treatment with reflex inhibition and facilitation, and based on the assertion that "cerebral palsy is lack of inhibition of reflex patterns of posture and movement" (Levitt, 2004, p. 18). This technique is built on patterning reflex inhibition, sensory motor experience, facilitation of postural reflexes, key body parts for control, developmental sequences, and all-day management (Levitt, 2004 p. 18).

Vaclav Vojta developed his technique using reflex creeping and other reflex reactions from Fay's and Kabat's ideas. Reflex creeping and rolling are triggered in specific reflex zones on the child's body. Vojta also uses sensory stimulation to promote creeping, and resistance to motion to help encourage proper muscle response and use (Levitt, 2004, p. 19).

Andras Petö established Conductive Education. This method involves an extensive therapist training period during which time the therapist earns the title, 'conductor.' The conductor then leads groups of 15-20 children in a strictly planned all-day program, including activities of daily living and conscious movements facilitated by rhythmic intention. When using rhythmic intention, conductors and group members will say the intended movement, and then count rhythmically while performing the movement (Levitt, 2004, p. 20).

More recent studies have been completed regarding a range of other current therapeutic techniques. Some of these techniques include body weight-supported treadmill training, strength training, botulinum toxin A injections, and surgery (Desloovere et al., 2007; Kramer & MacPhail, 1994; MacPhail & Kramer, 1995; Patikas et al., 2006; Provost et al., 2007). Each of these techniques has both benefits and drawbacks, and each method appears to help some, though not all, children with cerebral palsy.

Provost, et al. (2007) examined the effects of body-weight supported treadmill training on endurance and gait in children with cerebral palsy. They found that this technique might help some children with cerebral palsy to increase walking velocity and decrease energy expenditure; however, the results regarding endurance and single-leg balance were inconclusive. While body weight-supported treadmill training seems to help habilitate some aspects of gait in some children with cerebral palsy, the equipment necessary for the training is difficult and expensive to access (Provost et al.).

Some studies have shown a strength-training program to be beneficial to gait habilitation in cerebral palsy (Kramer & MacPhail, 1994; MacPhail & Kramer, 1995). However, in a randomized controlled trial, Patikas, et al. (2006) found no significant improvement from a strength-training program they implemented with their experimental group. The authors suggest that their study differs from former studies in that their participants trained at home rather than in a physical therapy clinic, where they could access special equipment such as an isokinetic dynamometer. Patikas, et al. (2006) chose to execute this study because special equipment and professional therapists can be difficult and/or expensive to access, and not all families possess the time and money required for frequent clinic treatments.

Botulinum toxin A (BTX-A) injections are one treatment method used to help relieve spasticity (Gage, 2004). This treatment works by blocking the release of the neurotransmitter acetylcholine at the presynaptic nerve terminal, thereby preventing muscle contraction through the inhibition of nervous signal transmission (El, Peker, Kosay, Iyilikci, Bozan, & Berk, 2006). El, et al. (2006) found that participants with cerebral palsy displayed significant improvement both in selective motor control at the ankle and in ankle dorsiflexion following injections in the gastrocnemius and hamstring muscles. After six months, these improvements decreased, but did not return to baseline.

Desloovere, et al. (2007) also found positive results with multilevel BTX-A injections in children with cerebral palsy. They noted that these treatments seemed to help improve walking patterns of the experimental group by allowing proximal muscles to grow longer, or closer to the size appropriate to the participants' bone length. This muscle length resulted in a reduction of anterior pelvic tilt, and improved hip and knee extension during mid-stance. However, both the control group (that did not receive BTX-A) and the experimental group showed similar knee flexion at initial contact, indicating that the injection did not help participants maintain this extension at the knee throughout the gait cycle.

When less invasive therapies are ineffective, orthopedic surgeries are employed to lengthen muscle contractures, and thereby improve gait and posture in children with cerebral palsy (Gage, 2004). Patikas, Wolf, Schuster, Armburst, Dreher, and Döderlein, (2007) recorded EMG patterns in children following multilevel surgery. The participants of this study showed statistically significant decreases in stance duration and in stride length; however, their EMG patterns changed after surgery in lower leg muscles only. The patterns of the knee flexor and extensor muscles of the thigh remained the same following surgery, indicating that these muscles may require a different therapeutic technique in order to work more functionally.

One surgery employed for children with cerebral palsy is Achilles tendon lengthening, in an effort to treat ankle equinus (Dietz, Albright, & Dolan, 2006). Often, this type of surgery over-lengthens the Achilles, causing weakness in the triceps surae (the gastrocnemius and soleus muscles of the posterior lower leg), which results in crouch gait (Gage, 2004; Dietz et al., 2006). In a follow-up study of this type of surgery, Dietz et al. (2006) state that the best treatment for ankle equinus is not known, and that therapists have yet to establish a reliable therapy for crouch gait. These authors suggest that the best candidates for this Achilles lengthening surgery are people who require only one surgery on one leg. Dietz et al. believe that alternative, non-surgical treatments are preferable for diplegic or quadriplegic patients with cerebral palsy who exhibit ankle equinus.

The various treatments that have been developed for gait habilitation in cerebral palsy have been shown to help some children in some circumstances. Because the 'umbrella term' of cerebral palsy refers to a vast number of diagnoses and symptoms, most children with cerebral palsy will need a composite of several different therapies. While the available treatment techniques help to a certain extent, they leave some symptoms either untreated, or poorly treated. RAS is a non-invasive gait rehabilitation technique that is motivational, more affordable and accessible than treatment with elaborate equipment, less invasive than botulinum toxin A injections, oral medications, or surgery, and has been shown to improve select kinematic measures and motor unit firing patterns in some muscles associated with gait (Fernandez del Olmo & Cudeiro, 2003; Thaut & McIntosh, 2006; Thaut et al., 1993; Thaut et al., 1997). Research such as the present study is vital in order to advance gait habilitation in cerebral palsy.

Rhythmic Auditory Stimulation and Gait Rehabilitation

Two published studies have been completed that examined the use of RAS with children with cerebral palsy (Kwak, 2007; Thaut, Hurt et al., 1998). Kwak (2007) included a control group, a therapist-guided training group (TGT), and a self-guided training group (SGT). The control group participated in traditional gait training with a physical therapist, while the TGT and the SGT participated in RAS. All groups were asked to train thirty minutes per day for three weeks. Results showed a statistically significant difference from pre- to posttests in the stride length (p=0.014), velocity (p=0.016), and gait symmetry (p=0.048) of the TGT.

Thaut, Hurt, et al. (1998) performed a within subjects repeated measures investigation that employed both an immediate entrainment and a training design. Thaut, Hurt, et al. (1998) reported significant results for improvement of the participants' gait velocity and swing symmetry for the entrainment portions of the study. During the normal walking speed, velocity increased from 28.3 ± 4.6m/min to 36.4 ± 7.6m/min, and during the fast walking speed, velocity increased from 40.7 ± 7.4m/min to 43.9 ± 7.8m/min. Swing symmetry ratios improved from 0.87 to 0.91 for the normal walk and from 0.88 to 0.91 for the fast walk. They also reported a velocity increase of 21.2%, as well as improved knee and hip range of motion following the training procedure.

The feed-forward mechanism of an auditory rhythmic stimulus has been documented for several other types of neurologic disorders. In 1983, Staum investigated whether a rhythmic stimulus could help improve gait symmetry and reduce fluctuations in velocity in participants with a variety of gait disorders. She found that after training with a rhythmic stimulus, all of her participants improved their stride symmetry and/or reduced inconsistencies in velocity. Other researchers have shown repeatedly that external, auditory, rhythmic cueing is effective in increasing cadence, stride length and velocity in stroke patients, both during immediate entrainment exercises and following a several-week training period (Mandel, Nymark, Balmer, Grinnell, & O'Riain, 1990; Thaut et al., 1992a; Thaut et al., 1993; Prassas et al., 1997; Thaut et al., 1997; Mauritz, 2002). Similar results have been documented about gait training for patients with

Parkinson's disease (Thaut et al., 1996; Freedland et al., 2002) and traumatic brain injury (Hurt et al., 1998).

Electromyography (EMG) recordings indicate that muscles recruit fibers more efficiently during auditory rhythmic stimulation than without this stimulus. Thaut, Schleiffers, and Davis (1992) reported a decrease in variation of muscle fiber firing in the biceps and triceps brachii muscles of healthy college students. Thaut, McIntosh, Prassas, and Rice (1992b) found similar results in the gastrocnemius muscles of healthy adults walking to a rhythmic auditory stimulus. Thaut et al. (1993) showed that the gastrocnemius muscles of hemiparetic stroke patients followed more efficient fiber recruitment patterns on both the affected and unaffected sides, as well as decreased variability on the affected side.

Hurt, et al. (1998) investigated whether RAS could improve gait characteristics of participants with traumatic brain injury. Following a training study, the researchers noted significant increases in velocity, cadence, and stride length of the participants between normal speed no-RAS and RAS conditions. They also observed trends toward improved swing symmetry in both normal and fast walks, and noted increases in cadence, velocity, and stride length from fast no-RAS to fast RAS conditions.

Music and Cerebral Palsy

The use of music with children with cerebral palsy is variously documented. Many of the older articles report on the use of music to assist people with cerebral palsy with development of speech, motor skills, and social interaction. Weigl (1954) describes the development of a 'rhythm band' with children with cerebral palsy at a New York public school. The author suggests that learning to play an instrument as part of this band provided the children with a sense of satisfaction, and that improvements in motor skills often occurred because a child was motivated to play a particular instrument. Weigl goes on to say that the children played musical phrases as 'conversations' with each other, and that playing in the 'rhythm band' provided physical exercise, developed the imagination, and provided opportunities for leadership when the children were asked to direct to group.

Boswell and Vidret (1993) also used rhythmic movement and music to help children with cerebral palsy to increase joint range of motion and improve muscle coordination, increase speech utterances, and develop a greater sense of self-worth. Lasseter, Privette, Brown, & Duer (1989) learned that rhythmic dance therapy to music helped children with cerebral palsy improve motor skills, acquire a sense of accomplishment, and develop increased social skills. Monti (1985) employed music therapy to assist children with cerebral palsy with active play, emotional expression and well-being, organizing, increasing attention, exploratory behavior, and interacting appropriately with others. Perry (2003) observed music to be useful for improving communication between children with cerebral palsy in a group setting. Snow (1950) utilized rhythm and instrument playing to help children with cerebral palsy learn functional movements.

May (1956) found that group music sessions encouraged greater social interaction, that singing assisted speech development, and that rhythmic playing inspired dance movements and cued marching. Rudenberg (1985) observes, "the desire to dance and to move rhythmically seems to be innate in all human beings" (p. 53). She presents case studies of the use of music with children with cerebral palsy. One child learned to play the chord organ and sing, which provided an outlet for self-expression and improved respiration. A second child listened to music for relaxation and played instruments to improve upper body motor function, increase lung capacity, express emotions, and heighten self-confidence. For the third child, music provided an orienting stimulus, encouraged vocalizations, and assisted with maintenance of postural control.

Ball, McCrady, & Hart (1975) tested two participants with cerebral palsy to determine whether music listening contingent upon upright head posture encouraged longer periods of head control. The researchers observed both participants under non-contingent music conditions and contingent music conditions, and found that both participants maintained upright head posture for reliably longer periods of time under the contingent music condition. Walmsley, Crichton, and Droog (1981) also noticed improvement in head control while using music as a biofeedback tool, while Neilson and McCaughey (1982) helped young adults with cerebral palsy self-regulate muscle contraction and reflex response levels by using preferred music as a reward for accomplishing a task.

Other researchers report that music can help reduce athetotic tremors and spasticity. Palmer and Zerbe (1945) observed that certain types of music helped a participant with athetoid cerebral palsy to stabilize his tremor rate, and then experience relief from tremor for increasing periods following music termination. Scartelli (1982) used 'sedative' background music to help adult participants with spastic cerebral palsy to reduce spasticity in their finger extensors. The researcher found that participants in the control group, receiving only EMG biofeedback, decreased finger extensor tension by 32.5%, while the experimental group, receiving EMG biofeedback in conjunction with background music, decreased muscle tension by 65%. Schneider (1956) found that background music seemed to increase the performance of people with cerebral palsy during various psychomotor tasks, that the same music affected the participants' behaviors, and that participants with spasticity were more productive during "stimulative" music, while participants exhibiting athetosis increased productivity during "sedative" music.

Berel, Diller, & Orgel (1971) learned that children with cerebral palsy could accomplish increasingly complex visual motor sequencing tasks when they used music as an auditory cue. The participants in the study were less successful with these sequencing tasks when the auditory cueing was removed, leaving the children to perform using their visual motor systems exclusively.

Holser & Krantz (1960) taught group singing to help their adult clients with cerebral palsy to improve lung capacity and speech, and employed rhythm to help the participants raise and lower their arms while inhaling and exhaling in a rhythmic manner. They found that instruction on musical instruments increased motor abilities of their clients. Holser and Krantz suggest that cooperative sessions between music therapists and physical therapists would be of great benefit to their clients. Alvin (1961) also encouraged co-facilitation between a music therapist and a physical therapist, noting, "music possesses rhythm and duration which can be linked with motor control in time and space" (p. 257).

Flodmark (1986) investigated the use of auditory biofeedback to help children with cerebral palsy improve gait by enhancing body perception and proprioception abilities. Flodmark reported that participants with diplegia or hemiplegia caused by damage to the motor cortex, and without associated disabilities, improved their gait while maintaining their speed. Two participants with athetosis showed trends toward improvement, but had inconsistent results because they had difficulty with turns during ambulation. While auditory biofeedback is a different training technique than RAS, this study suggests that auditory input can have a powerful influence on gait habilitation in patients with cerebral palsy.

Investigations that employed RAS to facilitate gait training in children with cerebral palsy produced positive results. Kwak (2007) showed a statistically significant difference in the stride length, velocity, and gait symmetry of the participants in the music therapist-guided training group, while Thaut, Hurt, et al. (1998) reported significant results for improvement of the participants' gait velocity and swing symmetry, as well as improvements in cadence, stride length, and range of motion in the knee and hip. Master's theses by Latzke (2004) and Santistevan (1999) showed trends toward increased gait velocity in children with cerebral palsy due to RAS cueing, while Santistevan noticed an additional trend of increased stride length.

Although several studies have been published on the use of music with children with cerebral palsy, and some of them mention the use of various rhythmic exercises to assist with motor function habilitation, only two published studies used quantitative measures and statistical analysis to examine the use of RAS for gait habilitation. The current study employed quantitative measures to examine the following: (1) whether children with spastic diplegic cerebral palsy can entrain to an external, auditory, rhythmic stimulus; (2) whether stride symmetry changed between uncued and RAS-cued walking conditions; (3) whether knee extension at foot contact changed between uncued and RAS-cued walking conditions.

CHAPTER 3: METHODOLOGY

Participants

This pilot study included 5 participants, 1 male and 4 female, all of whom had been diagnosed with spastic diplegic cerebral palsy, ages 7-13 (mean age 8.2 ± 1.3 years). During the study, all participants ambulated without assistive devices, and were classified as level I or level II in the Gross Motor Function Classification System (GMFCS). A classification of level I indicates that the child walks “without limitations” (Palisano, Rosenbaum, Bartlett, & Livingston, 2007, p.2), but “speed, balance, and coordination are limited” (p.3) for more demanding gross motor skills. Children classified as level II walk “with limitations” (p.2), has more difficulty coordinating advanced gait activities, and may require hand-held assist or an assistive device to ambulate longer distances (Palisano, Rosenbaum, Bartlett, & Livingston).

All participants exhibited limitations of cadence, stride length, velocity, and stride symmetry, and were cognitively, behaviorally, and physically able to complete all parts of this study. Children were not included in this study if they had a medical condition that would interfere with their safety while completing the study, if they were unable to ambulate without assistive devices, if they displayed lower limb contractures of greater than 15 degrees, if they had received a surgical intervention within 12 months or neurotoxin injections within 6 months prior that affected their typical gait pattern, or if they had a hearing impairment. The children joined the study after seeing an advertisement at a cerebral palsy or hippotherapy clinic, or were referred to the study by orthopedic surgeons, physiatrists and physical therapists at The Children's Hospital in Denver, Colorado. The researchers obtained informed, written consent from each participant as well as from his or her legal guardian. Participant descriptors are outlined in Table 1.

Table 1

Participant Descriptors

Participant	Age	Sex	GMFCS level
1	10	F	II
2	7	M	I
3	8	F	I
4	9	F	I
5	7	F	I

Note. All participants had diagnoses of spastic, diplegic cerebral palsy. GMFCS = Gross Motor Function Classification System.

Instrumentation

All data were collected in the Center for Gait and Movement Analysis (CGMA) at The Children's Hospital in Denver, Colorado. Standard VCM markers were placed on bony landmarks of the pelvis, hip, knee, ankle, heel, and forefoot of each participant, and a 12-camera Vicon MX motion capture system recorded the trajectory data at those locations. The Vicon CCD Oxford Metrics Pulnix Progressive Scan visible red light strobe cameras were set at 120 Hz. Cadence, stride length, and velocity were measured using 4 Kistler 6-degree of freedom force plates. Observational video for kinematic analysis was taken with 2 Panasonic high-resolution digital signal processing color video cameras mounted on pan and tilt units. Data were processed with a Vicon Workstation plug-in gait module, and using Vicon software, the Polygon model synchronized the video in a multimedia format (CGMA, 2008).

Procedure/Design

This immediate entrainment study is part of a larger project designed to examine the effects of a 3-week ambulation-training period with and without RAS on gait patterns of children with spastic, diplegic cerebral palsy. The study was approved by the Institutional Review Board (IRB) at the University of Colorado at Denver and Health Sciences Center, and funded by The Children's Hospital Research Institute. All participants participated in all four conditions of this entrainment study. When they arrived at the CGMA, the participants and their caregivers received an explanation of the study and the caregivers signed Study Consent and HIPAA B forms. Participants signed assent forms if they were at least 7 years old, cognitively able to understand the form, and physically able to sign the form. Trained technicians then placed reflective markers on the participants at bony landmarks of the pelvis, hip, knee, ankle, heel, and forefoot.

The data collection protocol included four stages: (1) an un-cued, participant-selected normal walk, (2) an RAS-cued walk matched to this normal walking cadence, (3) an un-cued, participant-selected fast walk, and (4) an RAS-cued walk matched to the participant's fast walk. The participants walked, barefoot, 10 meters across the CGMA flooring, crossing the four force plates in the middle of the walk. One researcher stood at each end of the flooring for all stages of the data collection. For the first stage, each participant was instructed to "walk how you normally walk." After a normal cadence had been established, the participants were given wireless headphones to wear with music of the established normal cadence, and were instructed to tap along with the beat of the music for a few moments. For the second stage of data collection, the participants were instructed to "walk to the beat of the music." Before the third stage of data collection, each participant was instructed to walk "with your fast walk" across the floor. After the fast cadence was established, the participant once again received headphones, tapped to the beat of the music and were told to "walk to the beat of the music" for the fourth stage of data collection. During each stage of data collection, the participants were asked to walk down the floor and back 3-5 times. All music used during RAS was in duple meter, and had been arranged or composed and electronically produced by Colorado State University (CSU) research staff.

For this study, the following data were collected for both the no-RAS and the RAS conditions: timing of foot contact, left and right stride lengths, left and right stride times, and knee angles in the sagittal plane (flexion/extension) at foot contact. Timing of foot contact is measured in milliseconds from the starting point, with foot contact being the point in the gait cycle that terminates the swing phase. Stride length is measured in meters, and is defined as the distance traveled between heel strike (often referred to in this study as foot contact) of one foot to heel strike of the same foot. Stride time is measured in milliseconds, and is the time taken to complete one stride length. Sagittal plane knee angles are measured in degrees of flexion. These data were used to examine synchronization error (SE) between the beats of the music and participant foot contact, absolute period error (APE) of time between beats and time between foot contacts, stride time symmetry, stride length symmetry, knee extension at foot contact, and variability of degree of knee extension at foot contact. SE and APE are measured in milliseconds (ms).

Stride symmetry is a ratio produced by dividing the shorter stride time/length by the longer stride time/length for successive left and right strides, and stride symmetry increases as the ratio approaches 1, with 1 being 100% symmetrical. Knee extension at foot contact in normal gait typically ranges from 5-10 degrees of flexion (Gage, 2004), and varies within this range from one stride to the next.

Analysis

Statistical analysis of data from this study includes means, standard deviations, percent change scores from uncued to RAS-cued conditions, and randomized block analysis of variance (ANOVA) for determining significant differences between conditions. The significance level was set to $p < 0.05$.

In order to compute grand means and standard deviations for synchronization error and absolute period error, the data were normalized using a scale factor. This was necessary due to the variety of gait speeds the participants selected, which ranged from 120 beats per minute (bpm) – 150 bpm for self-selected normal speeds and 130 bpm – 180 bpm for fast speeds.

CHAPTER 4: RESULTS

The four conditions in this study were self-selected speed normal walk (SS), self-selected matched with music at the same cadence (SSM), fast walk (F), and fast matched with music at the same cadence (FM). The independent variables were speed (self-selected and fast), and music cueing (no RAS and matched with RAS). The dependent variables were synchronization error (SE), absolute period error (APE), stride time symmetry, stride length symmetry, knee extension at foot contact, and variability of knee extension at foot contact. To determine differences of SE and APE between SSM and FM conditions, a randomized block ANOVA was used, blocking on participant with speed as the one fixed effect. Examinations of stride symmetry ratios and knee extension angles employed a randomized block ANOVA, blocking on participant with speed and music as the two fixed effects. Tukey's adjustment for multiple comparisons was used to perform pairwise comparisons of speed, music, and their interaction means.

Table 2 provides individual means and standard deviations of SE and APE by participant, while Table 3 delineates overall mean and standard deviation of SE and APE for music trials. Average SE between participants for SSM trials was 13ms before the corresponding beat of music, ± 100ms. The average APE for SSM trials was 25 ± 22ms. Mean SE during FM trials was after the music beat 28 ± 145ms, while mean APE during FM trials was 45 ± 61ms. Participants maintained a low average period error in both SSM and FM trials, suggesting gait synchronization with the auditory stimulus. No significant difference was found between SSM and FM trials, indicating that speed did not affect the ability of the participants to synchronize with the rhythmic stimulus.

Table 2

Means and Standard Deviations of Synchronization Error and Absolute Period Error by Participant

		Synchronization Error (ms)		Absolute Period Error (ms)	
Participant	Trial	Mean	SD	Mean	SD
1	SSM	34.25	31.54	13.41	10.98
	FM	38.28	150.43	60.14	74.44
2	SSM	-17.69	103.10	41.82	20.49
	FM	38.03	141.29	70.47	100.43
3	SSM	-19.72	64.73	50.11	18.56
	FM	125.35	71.87	52.46	22.49
4	SSM	-60.90	44.84	9.56	7.39
	FM	-66.12	177.00	15.93	11.78
5	SSM	-0.20	172.71	11.55	7.98
	FM	2.60	97.12	23.01	14.65

Note. SSM = Self-selected matched; FM = Fast matched.

Table 3

Mean and Standard Deviation of Synchronization Error and Absolute Period Error for Music Trials

	Synchronization Error (ms)		Absolute Period Error (ms)	
Trial	Mean	SD	Mean	SD
SSM	-12.93	100.22	25.31	22.14
FM	27.72	145.45	44.94	61.49

Note. SSM = Self-selected matched; FM = Fast matched

Figures 1-4 illustrate SE and APE for selected trials for two different participants. The graphs visually demonstrate that the APE for these trials remained small, despite generally larger values for SE.

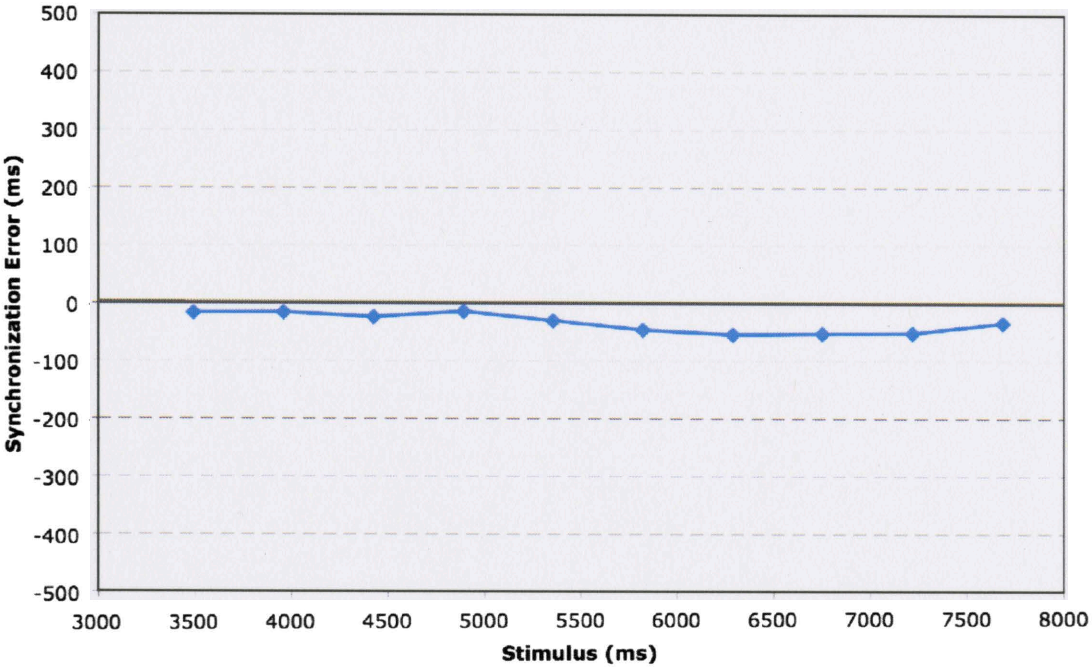

Figure 1. Synchronization error (response deviation from stimulus) for participant 4, SSM condition, trial 1. Negative values indicate that the response (foot strike) anticipated the stimulus (beat of music).

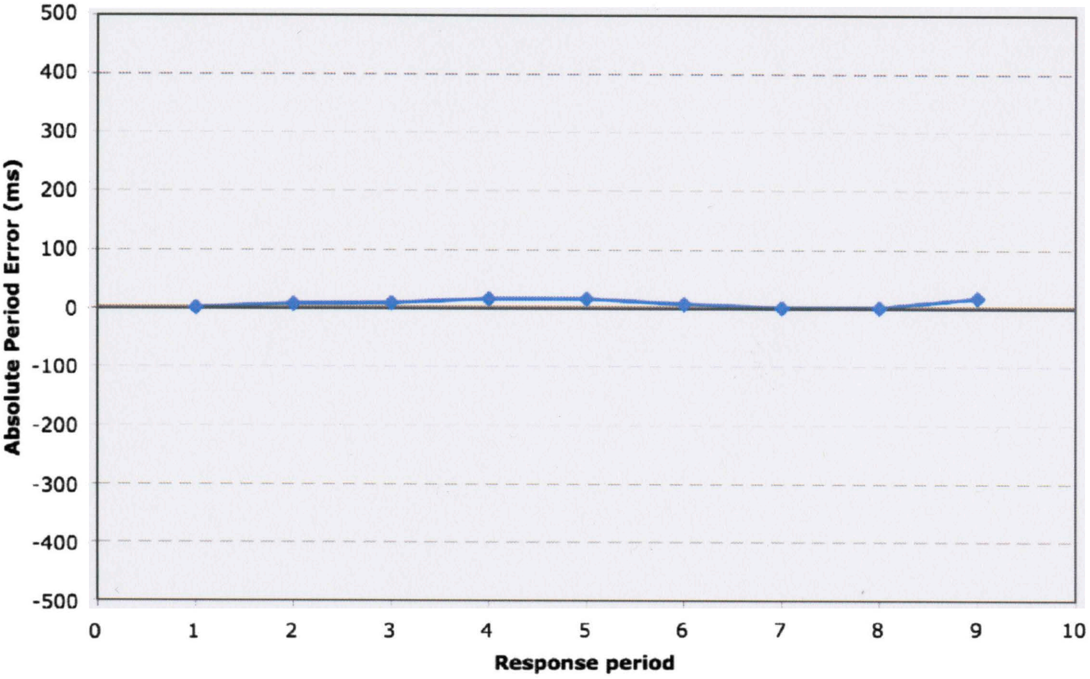

Figure 2. Absolute period error for participant 4, SSM condition, trial 1.

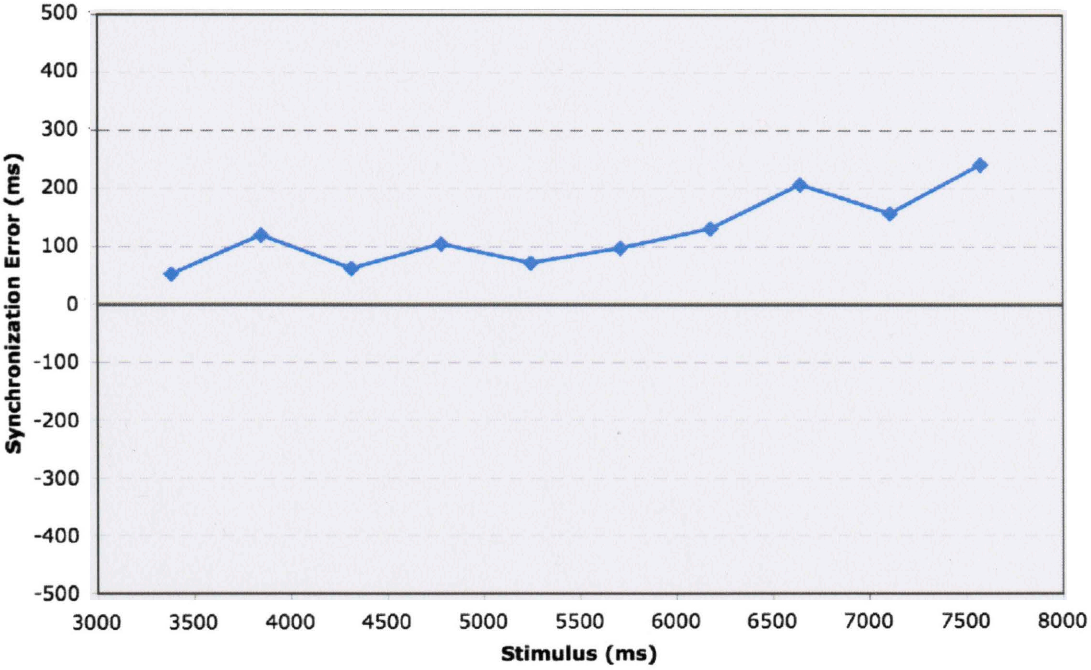

Figure 3. Synchronization error for participant 2, SSM condition, trial 3. Positive values indicate that the response followed the stimulus.

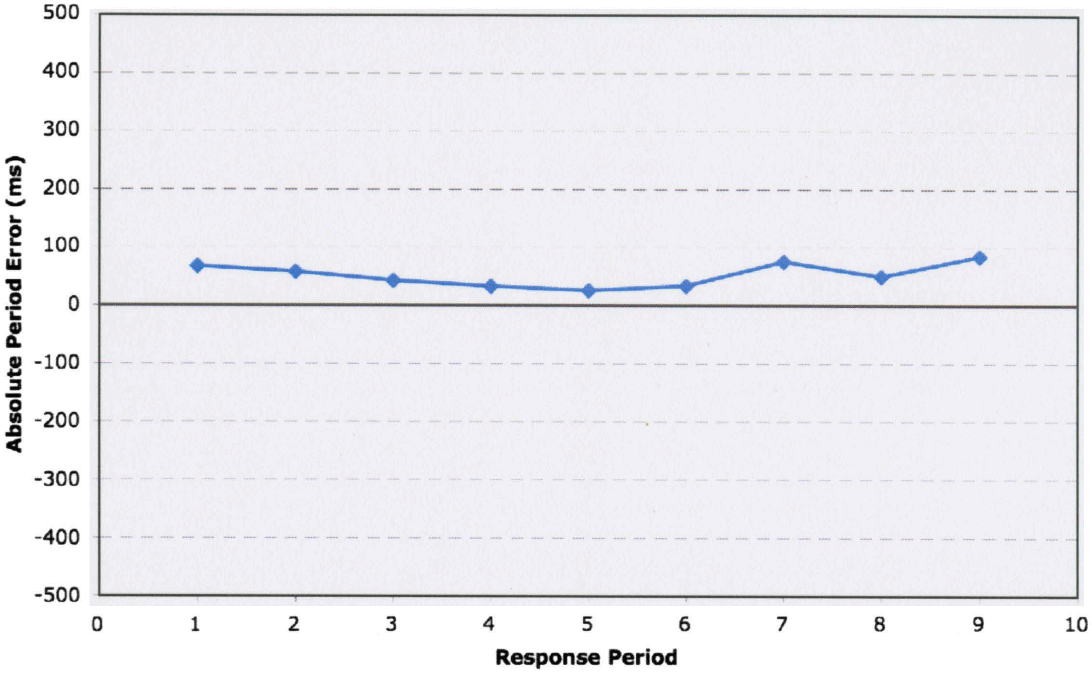

Figure 4. Absolute period error for participant 2, SSM condition, trial 3.

Stride symmetry ratios are one measure of gait uniformity between right and left sides of the body. These ratios were produced by dividing the shorter stride time by the longer stride time, and the shorter stride length by the longer stride length of the same gait cycle for each of the 4 conditions for each participant. No significant differences were found between music, speed, or music by speed effects for stride symmetry, and all ratios were within 0.1% of 1.

Table 4 displays the mean, standard deviation, and percent of change for self-selected speed trials (SS and SSM) of stride symmetry and knee extension at foot contact. Raw data show that the average stride time symmetry increased by 0.29% during the music trials (p = 0.07). From F to FM trials, stride length symmetry increased by 0.16% (p = 0.44) (see Table 5).

Table 4

Mean, Standard Deviation, and Percent of Change for Self-Selected Speed Trials

Parameter	SS		SSM		% Change
	Mean	SD	Mean	SD	
Stride Time Symmetry	0.9946	0.0020	0.9975	0.0027	0.29%
Stride Length Symmetry	0.9970	0.0031	0.9963	0.0044	-0.07%
Knee extension at foot contact	18.7118	1.4992	19.4388	1.1600	3.89%

Note. Knee extension at foot contact is presented as degrees of flexion.
SS = Self-selected; SSM = Self-selected matched.

Table 5

Mean, Standard Deviation, and Percent of Change for Fast Speed Trials

Parameter	F		FM		% Change
	Mean	SD	Mean	SD	
Stride Time Symmetry	0.9963	0.0031	0.9949	0.0010	-0.14%
Stride Length Symmetry	0.9961	0.0034	0.9977	0.0014	0.16%
Knee extension at foot contact	18.7170	1.2801	20.3790	1.6596	8.88%

Note. Knee extension at foot contact is presented as degrees of flexion.
F = Fast; FM = Fast matched.

Mean overall degrees of knee flexion increased from no-RAS to RAS trials; however, when examining the raw data for participant 1, it appears that the average degrees of knee flexion decreased during music conditions, with the following results: between SS and SSM conditions, mean left knee flexion decreased from 29.08 ± 1.07° to 28.25 ± 0.06°, while right knee flexion decreased from 26.86 ± 1.92° to 24.62 ± 0.80°;

between F and FM conditions, average left knee flexion decreased from 27.86 ± 0.25° to 25.39 ± 0.50°. No significant differences were found for music, speed, or music by speed conditions for this measure. Additionally, no significant differences were found between conditions for variability of knee extension at foot contact.

CHAPTER 5: DISCUSSION

This immediate entrainment study was completed to examine whether children with spastic diplegic cerebral palsy synchronize their walking cadence with auditory rhythmic cueing, and whether rhythmic cueing techniques such as RAS influence stride symmetry and knee extension at foot contact in this population. Null hypothesis 1 was rejected; null hypotheses 2 and 3 were not rejected. Any inferences made from the analysis are limited in scope, due to study design and small sample size (N=5); however, some valuable information may be obtained from the results of this study.

In the SSM condition, 4 out of 5 participants exhibited negative synchronization errors, indicating that they anticipated the beat during these trials. During FM trials, 4 of 5 participants stepped following the stimulus, as demonstrated by positive synchronization errors; however, mean and SD of APE remained relatively low (45 ms ± 61), demonstrating that they continued to entrain even during fast speed conditions. It is likely that the positive mean SE during FM conditions is due to the participants' difficulties maintaining the faster speed. The number of contractures associated with spastic diplegia may have prevented the participants from ambulating at a consistently fast speed, or fatigue may have contributed to the step delay. Because this entrainment study is the first part of a longer training study, it was decided not to randomize the order of the walking trials, resulting in the FM condition being the last of four in any given testing session.

In finger-tapping entrainment studies (Molinari et al., 2005; Thaut, Miller et al., 1998), healthy individuals synchronized motor movements with an auditory rhythmic stimulus, demonstrating a slight anticipation of the beat, and maintaining a small period error. This research indicates that entrainment occurs across the stimulus period, allowing the brain to plan and implement motor patterns to be completed in a designated amount of time. In this case, phase synchronization, or tapping at precisely the same time as the auditory cue, becomes a secondary synchronization strategy. Because period entrainment appears to be the primary synchronization strategy, APE may be a more accurate measure of entrainment than SE.

In the current study, the SD of the APE in both SSM and FM conditions remained low (22.14ms and 61.49ms respectively) when compared to the SD of SE (SSM = 100.22ms; FM = 145.45ms). This comparison of SD indicates that the participants demonstrated less variability between response period and stimulus period than between response time and stimulus time. They therefore employed the same synchronization strategy as the participants in the previously discussed finger tapping studies (Molinari et al., 2005; Thaut, Miller et al., 1998). The participants in this study were more consistent with timing between steps than with stepping precisely at the same time as the auditory stimulus. Clinical implications of this entrainment strategy will be discussed in a later section.

No significant differences were found for stride time symmetry or stride length symmetry between no-music and music conditions. Although average stride time symmetry increased (0.29%) from SS to SSM trials and average stride length symmetry increased (0.16%) from F to FM trials, ratios for all conditions were within 0.1% of 1. It is possible that for many participants with diplegic cerebral palsy, stride symmetry may not be the best indicator of improvement in gait, as both sides of the body are affected by upper motor neuron damage. However, both sides are not always affected in the same manner or to the same degree. With this in mind, tests of symmetry using kinematic measures such as hip or knee range of motion may be more appropriate than stride time symmetry and stride length symmetry. It is more likely, however, that the small number of participants did not allow adequate investigation into this area of gait analysis, due to a lack of statistical power. The small sample in this study may not be generally representative of children with spastic diplegic cerebral palsy, as others of this population would display less stride symmetry during their normal walk, possibly allowing for greater improvement with rhythmic cues.

Previous studies have examined stride time symmetry, or swing symmetry. The population sample that participated in the immediate entrainment study completed by Thaut, Hurt, et al. (1998) demonstrated significant improvement from no-cueing to cueing conditions for both normal and fast speeds; however, stride symmetry ratios in that study were measured at 0.87 for the normal uncued walk and 0.88 for the fast uncued walk. Both mean ratios improved to 0.91 during the rhythmic cueing conditions, but even these improved numbers are lower than the mean ratios for the current study, in both uncued and cued conditions. The difference in findings between these two studies can be accounted for by the fact that the stride symmetry ratios for the current study started within the normal range, leaving very little room for improvement.

Although Thaut, Hurt, et al. (1998) did not report improvement in gait symmetry following the 3-week training study, Kwak (2007) reported significant improvements (p=0.048) in swing symmetry for the therapist-guided training (TGT) group when comparing pre- and posttest means in a paired samples t-test. However, Kwak (2007) did not find significant differences between control, self-guided training (SGT), and TGT groups, possibly because the differences found within groups were not large enough to create significance between groups. Direct comparison of stride symmetry results between the current study and that completed by Kwak (2007) is not possible, both because Kwak (2007) does not report symmetry ratios, and because the protocols for the studies were different. The current study completed only an immediate entrainment protocol, while Kwak (2007) completed only a 3-week training study, which allows for repetition, learning, and some development of motor patterns. Additionally, the TGT group completed rhythmically cued warm-up exercises prior to gait training, which may have contributed to the significant results in this group from pre- to posttests.

Santistevan (1999) reported a 0.267% increase in stride symmetry from the normal uncued walk to the normal cued walk of an entrainment study using RAS with children with cerebral palsy. This increase was not significant, but is comparable to the 0.29% increase in stride symmetry found in this study between the SS and the SSM conditions. Unlike the current study, in which the ratios were computed by dividing the shorter stride time by the longer stride time, Santistevan (1999) reported calculating ratios by dividing the longer stride time by the shorter stride time; therefore, ratio comparisons between these two studies is not appropriate.

Examination of stride symmetry in RAS studies with children with cerebral palsy highlights the substantial variation of involvement experienced by children diagnosed with cerebral palsy. Kwak (2007) included participants at varying levels of cognitive function, with and without need of assistive devices, and with different orthopedic surgical histories. Santistevan (1999) included participants with hypotonia, spastic hemiplegia, and spastic diplegia as diagnoses, some of who required assistive devices for ambulation. This study and Thaut, Hurt, et al. (1998) included a more specific diagnosis of spastic diplegia, but the sample sizes for both studies were small. Research that includes larger participant pools, but that limits the diagnoses, GMFCS level, and other varying characteristics of participants is needed in this field in order to understand further the habilitation potential of RAS for children with cerebral palsy.

Children with spastic diplegic cerebral palsy often have difficulty accomplishing full knee extension at terminal swing and foot contact (Gage, 2004). Treatments for this problem are limited in number and effectiveness (CGMA, 2008). In this immediate entrainment study, no significant differences were found between no-music and music trials for knee extension at foot contact. Likewise, no significant differences were found in knee extension variability between no-music and music conditions. While this lack of significance may be an effect of the small sample size, it also may be that a training period is necessary for improvement to occur in this area. The training study with which this entrainment study is associated may provide more insight into this area of gait analysis. Thaut, Hurt, et al. (1998) reported improved knee range of motion with rhythmic cueing following the 3-week training study, but no quantitative data is provided for further analysis of this gait parameter. Neither Kwak (2007) nor Santistevan (1999) reported investigation of this kinematic measure.

This study had several limitations. The small sample size (N=5) decreased the power and increased the likelihood of both type I and type II errors. In this case, two of the three null hypotheses were not rejected, due to a lack of significant results. As previous RAS research on children with cerebral palsy and other populations with neurological impairments has suggested, utilizing an auditory rhythmic stimulus to facilitate gait habilitation can produce significant improvements in stride symmetry as well as in other gait parameters. This suggests that the sample size in this study may not have been large enough to measure probability of change accurately in this population.

Another limitation to this study was the lack of randomization of the order of conditions. While it was necessary to have the participants walk without music first, in order to establish the appropriate music cadence, it may have been more beneficial to randomize by speed, having some participants complete F and FM conditions prior to completing SS and SSM conditions. This randomization would help control for any learning and order effects that occurred during the testing session.

Relating to order of conditions, fatigue and boredom could have been factors in performance as well. Ambulating across 10 meters and back for all trials of each of 4 conditions was likely tiring for the participants both physically and mentally. Additionally, the entire process of explaining the study, gaining informed consent, placing the markers on the participants' bony landmarks, standing and ambulating to ensure proper functioning of the computers and instruments, waiting for appropriate music cadence to be established, and completing the study trials often took up to two hours per participant. Although the researchers engaged the children in conversation during much of the preparation and non-testing time, and the participants were given a seated rest between conditions, it is likely that the participants grew bored as well as tired, which could have influenced their performance during the data collection periods.

Future research could take these considerations into account, and make an attempt to streamline the data collection process as much as possible. The larger training study of which this investigation is a part will include 20 participants, all of who will undergo the immediate entrainment portion, and half of who (the experimental group) will complete three weeks of ambulating with RAS for 30 minutes per day. A control group will walk for 30 minutes per day for three weeks without a rhythmic stimulus, and posttest data will be collected at the end of the training as well as two weeks following the end of the training period. Data collected may indicate that a training period is preferable in order to accomplish noticeable, significant results.

It is recommended that other studies examine gait cadence, stride length, and velocity measures, as well as step symmetry and single and double support times, in order to reference these data with similar studies in this and other populations. Examining such parameters may also give further insight into how the use of a rhythmic stimulus may or may not assist with gait rehabilitation in children with spastic diplegic cerebral palsy. Researchers may also wish to examine whether there appear to be differences between the functional use of RAS with children with diplegic cerebral palsy and those with hemiplegic cerebral palsy. EMG studies, which have indicated increases in muscle recruitment efficiency and more appropriate contraction timing of antagonist muscles in stroke populations (Thaut, McIntosh, Prassas, & Rice, 1993), have not been implemented with this population. Such studies could determine whether children with cerebral palsy also demonstrate increased organization and recruitment of agonist and antagonist muscles when ambulating with an auditory rhythmic stimulus. EMG studies could also investigate whether auditory rhythmic stimuli could assist with the "plantar flexion-knee extension coupling" (Hicks et al., 2007, p. 2) that assists with achieving full hip and knee extension during normal gait.

Some studies have shown that strength training can improve gait in participants with cerebral palsy (Kramer & MacPhail, 1994; MacPhail & Kramer, 1995). Because rhythmic cueing has been shown to improve motor recruitment patterns, it would be interesting to investigate whether the addition of a rhythmic stimulus to a strength-training program would further gains, resulting in greater gait improvements for these children. Kwak (2007) implemented warm-up exercises with rhythmic cueing for the TGT group prior to gait training. Although the significant changes in several gait parameters found in the TGT group in this study may have been due to a variety of factors, these exercises could have contributed to the gains made by this group.

Clinical Implications for Music Therapy

The participants in this study demonstrated less variability of APE than of SE, indicating that their synchronization strategy is period rather than phase entrainment. The result of this synchronization strategy is that the participants may not have appeared to entrain because they may not have stepped exactly on the auditory cue. However, this study shows that assuming lack of entrainment in this situation would be a misperception of the neuromotor events taking place. In a clinical setting, maintaining a steady tempo with which the participants (or clients) could continue to synchronize their step period would be more appropriate than altering stimulus tempo in an attempt to reduce SE.

The stride symmetry ratios found in this study indicate that the participants were in the normal range for stride time and stride length symmetry. In the absence of other types of stride symmetry measures that may or may not indicate a habilitative need, the measures obtained in this study suggest that increased stride symmetry is not necessarily an appropriate goal for children with spastic diplegic cerebral palsy. Because Kwak (2007) and Thaut, Hurt, et al. (1998) measured stride symmetry ratios that were lower at pre-test than the current study, and because they found statistically significant improvements in stride symmetry, it is likely that this need is present in some, but not all, children with spastic diplegic cerebral palsy. However, despite stride symmetry ratios that measured in the normal range for healthy gait, it was evident during data collection that the participants exhibited decreased efficiency of movement, and would benefit from habilitation work to address this difficulty.

Immediate entrainment did not significantly change knee extension at foot contact in this study; however, Thaut, Hurt, et al. (1998) reported some improvements in knee ROM following a 3-week training study. Although more evidence is needed, it is possible that training with RAS over time would help participants to increase knee extension at foot contact, decreasing degrees of flexion from the means measured in this study to closer to the 5°-10° range of normal gait.

Another consideration for the clinical setting is the manner in which the rhythmic stimulus is delivered. In this study, the participants listened to pre-recorded music played through adult-sized wireless headphones as they ambulated across the walkway. While some participants stated that they liked some of the songs they heard, it is likely that most would have preferred other types of music. Additionally, in a clinical setting, a music therapist would have the ability to play live music, and to interact with the children in an age-appropriate manner. Use of client-preferred live music and personal interaction could maintain attention, improve mood, and decrease boredom, which may result in larger habilitative gains. Kwak (2007) interacted with the participants in the TGT group, both with verbal encouragement, and by adding a live drumbeat to the recorded music played during the training sessions. In that study, the TGT group was the only group that showed significant improvements from pre-to posttests, and this improvement may have been due in part to the personal interaction and live music presentation.

Conclusion

This immediate entrainment study examined the use of Rhythmic Auditory Stimulation as a gait habilitation technique for children with spastic diplegic cerebral palsy. The results support the idea that children of this diagnosis have the ability to synchronize their gait patterns with an external auditory rhythmic stimulus. While this information alone does not indicate that RAS is necessarily an appropriate therapeutic technique for this population, it does suggest the possibility that rhythmic cueing may be helpful, and that research in this area with a larger participant pool is pertinent. The ability to entrain to a rhythmic stimulus may provide children with cerebral palsy an added therapeutic tool for gait habilitation.

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