

NeuroGame Therapy for the Improvement of Ankle Control in Ambulatory Children with  
Cerebral Palsy

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**Abstract**

NeuroGame Therapy for the Improvement of Ankle Control in Ambulatory Children with  
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**Background**

Cerebral palsy (CP) describes a group of permanent movement and posture disorders occurring from non-progressive injury to the fetal or developing brain. CP affects 3-4 per 1,000 children in the United States and the majority of those children have spasticity as a primary presentation. Though most children with CP are able to walk, with or without the help of an assistive device, many of them have trouble with toe clearance, which can lead to instability and increased fall risk. Learning new motor patterns is possible due to neuroplasticity, but requires a high number of repetitions with the proper challenge, and appropriate feedback. A NeuroGame Therapy (NGT) system was developed that utilizes surface electromyography (sEMG) as a biofeedback device that allows the participant to practice activation of any targeted muscle. The sEMG activation controls a video game to make the practice enjoyable.

**Objectives**

The first objective was to determine valid and reliable outcome measures to assess walking in children with CP. The second objective was to determine if a different method of

assessing range of motion was valid and reliable. The final objective was to determine if using NGT to train the tibialis anterior muscles and pairing game play with immediate walking practice would affect balance, walking, and selective ankle movement in children with CP.

## **Methods**

First, a scoping review was conducted to determine the most appropriate measures of gait in children with CP, based on the measures' validity and reliability when used with children with CP. Second, a study was conducted on children with CP and those who were typically developing to determine if using video recording was a valid and reliable method of measuring ankle range of motion. Finally, an intervention study was conducted where nine children with CP used NGT at home for 6 weeks to train their tibialis anterior muscles bilaterally.

## **Results**

The results of the review indicated that the 6-minute walk test possessed the strongest evidence for use on children with CP when directly measuring walking capacity. Though the review showed that the Edinburg Gait Scale had the greatest evidence of appropriate psychometric properties in children with CP for measuring movement quality, the gold standard of 3-dimensional kinematic analysis was chosen for the intervention study. The study to determine the validity and reliability of using video recording to measure ankle range of motion revealed high validity and reliability when comparing measurements from video recording to real-time goniometry. Children who participated in the NGT study all showed improvement in at least one outcome measure assessing body structure and function (e.g. sEMG outcomes, muscle contraction force, selective motor control, etc.). Most, but not all, participants showed at least slight improvement in activity level measures (i.e. 6-minute walk test or reported falls).

NGT to train the tibialis anterior muscles was found to be feasible in children with CP. More NGT play appeared to yield better results, especially with sEMG outcomes. Overall, NGT did not appear to have an effect on dorsiflexion during gait, but did have a positive influence on dorsiflexion muscle contraction force. The majority of results seen in the post-test were observed again at the follow-up testing.

## **Limitations**

The suggested measures from the scoping review were based on psychometric properties rather than the intended or desired application of the outcome. The range of motion study was based on standardized marking of the leg and ankle and camera position, so is not generalizable to video recording of any other joint, or with recording without using the standardized set-up. The NGT study was subject to measurement error due to the technical nature of the data collection and the fact that children were assessed five times over 12 weeks, so natural variability was inevitable. There were also some technological limitations of the NGT system. Finally, the small sample size and mixed results make it difficult to generalize the results broadly to children with CP.

## **Conclusions**

Of the direct measurements, the 6-minute walk test possessed the strongest evidence of psychometric properties when measuring walking capacity in children with CP. The Edinburg Gait Scale had the strongest evidence for use in assessing kinematic properties during gait. Other measures, however, may have clinical utility based on the goals for that child. The video recording assessment of ankle range of motion in children used within this study was supported as a standardized method with high validity and reliability.

There is some promise for the NGT system to assist children with CP to improve muscle contraction force, however carry-over to changes in the children's gait pattern was limited. Given that the children who showed the most change were those that played the longest, more practice may be needed to effect greater change.

## Plain Language Summary

Cerebral palsy (CP) affects 3-4 per 1,000 children and is an umbrella term to describe a group of disorders that affect movement and posture. CP is the result of a non-progressive injury to brain prior to birth, or within the first two years of life. Though the brain injury itself is non-progressive, the secondary movement disorders affect mobility as children grow and become adults. Most children with CP are able to walk, but most have some difficulty doing so and may require an assistive device such as crutches or a walker. A major problem with walking is the difficulty in clearing the toes and dragging the foot during walking. This leads to decreased stability and a potential increase in falls.

Due to the decrease in toe clearance and balance, we thought that training the shin muscles responsible for picking up the toes would possibly lead to improvements in balance and walking. A previously developed system, called ‘NeuroGame Therapy’ (NGT) was used in children with CP and adults who had a stroke or traumatic brain injury to train muscles in the wrist. NGT uses surface electromyography (sEMG) to detect the electrical signals generated by the muscles as they contract and these signals can then be used to play a video game. We expected children to like to do NGT and therefore would work hard and longer at contracting their muscles than if we just asked them to practice this motion in isolation. We used the sEMG electrodes on each shin muscle, so depending on which side the child contracted, the game cursor would move to the same side (i.e. when the child tried lifting his right foot, the game would move to the right, and vice versa).

Prior to conducting the NGT experiment though, we needed to discover the best measures to use to evaluate change in walking and so conducted a review of walking tests previously used in children with CP. From this we learned that the 6-minute walk test (how far a child can walk

in 6 minutes) would be the most appropriate measure with the greatest amount of evidence to support use with children who have CP. We also wanted to measure changes in the ankle range of motion (ROM) carefully but without taking a lot of time, so we conducted a study to determine if measuring ankle ROM using a video recording of the testing, with measurement later would be acceptable. The ROM study concluded that this method was an acceptable alternative.

For the NGT study itself, each of nine children was instructed to play NGT using their shin muscles. The children were asked to do this 25-40 minutes and to take a five-minute walk 3-5 times per week for six weeks. We tested the children every three weeks for a total of five assessments, with two before NGT, one in the middle, and two after NGT. Measurements included various components of balance, walking, ankle movement, muscle isolation, strength, and foot control during movement and walking tasks.

Not all children played the game the full number of times recommended, and those who played it more seemed to show more improvement on the measures. All children showed at least one positive change on one of the measures used with most children showing change in shin muscle contraction force. Only a few children showed any changes in their walking activity. The biggest struggles with this study were difficulty recruiting a large number of children and some technological problems with the game system. It does appear that using NGT to train the shin muscles is feasible. We would like to use the NGT system in younger children and for different amounts of time to determine how much NGT play is needed to have some carry-over into walking.

Human Subjects approval number: 41590

Institutional review board: University of Washington

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## **Chapter 1:**

### **Outcomes and Intervention for Improved Ankle Control in Children with Cerebral Palsy**

Cerebral palsy (CP) is a group of permanent movement disorders attributed to non-progressive damage to the fetal or infant brain (Rosenbaum et al., 2007) and affects 3-4 out of every 1000 school-aged children in the United States (Aisen et al., 2011). CP is categorized by the dominant diagnosed muscle tone disorder (e.g., spasticity, ataxia, dystonia or athetosis), and the topography of impairment (e.g., one arm and leg, both legs, etc.) (Rosenbaum et al., 2007). The Gross Motor Functional Classification System (GMFCS) has been used to categorize levels of function in children with CP up to 18 years of age (Palisano et al., 1997; Palisano, Rosenbaum, Bartlett, & Livingston, 2008). This system incorporates five different levels of functioning where children at level I have the highest functional ability (i.e., walk independently but have difficulty with running, jumping, etc.) and level V depicting children with the lowest functional ability (i.e., require full assistance with movements). Children at GMFCS levels I-III are considered to be ambulatory children with CP as they are able to walk with or without the use of an assistive device (e.g., forearm crutches or a walker). Children with spasticity and a distribution primarily within the lower extremities (e.g., spastic diplegia) often have difficulty with foot/ankle control affecting foot placement during walking. This can cause a propensity for falls and problems with long term walking and joint health.

The primary impairment of CP is motor incoordination consisting of decreased selective voluntary muscle activity, especially in reciprocal patterns between the right and left side, and increased co-contraction of agonist/antagonist muscle groups (Fowler, Staudt, Greenberg, & Oppenheim, 2009; Kwon et al., 2012). These impairments affect gait and balance (Bolek, 2003).

Although CP is considered a non-progressive disorder, failure to appropriately address individuals' needs puts people with CP at risk for developing multiple long-term secondary impairments which cause loss of function, including decreased ambulation and problems with participation within family, school, and community-based activities later in life (Chiarello, Palisano, Bartlett, & McCoy, 2011; Damiano, Alter, & Chambers, 2009). Among the secondary impairments related to CP are joint contractures, bony deformities, pain, and decreased endurance and strength (Fowler et al., 2007; Ostensjø, Carlberg, & Vøllestad, 2004).

Improving walking ability is then a primary focus of physical therapy intervention for children with CP who are GMFCS levels I-III and have spastic diplegia. Balance and stability while walking can be affected by multiple primary and secondary impairments. Discovering better ways to prevent secondary impairments such as reduced range of motion and muscle strength is the focus of much research. Thus, the primary study of this project was to evaluate one method of intervention for children with CP designed to improve the use of muscles that control the ankle and foot position during walking. This is important because many interventions have been shown to be successful with children with CP, but few have shown retention and carry-over to function or improved safety while walking. Prior to discussing intervention for improving walking, however, we must first understand how to evaluate and assess change in walking. Evaluation of change is dependent on many aspects of the outcome measure used to assess the construct of interest. Aspects of an outcome measure that relate to its utility include psychometric properties such as validity and reliability.

Validity refers to an instrument's ability to measure what it is intended to measure and has multiple facets. There are many types of validity and different ways to determine each type. When developing a new measure that may replace a known measure, it is important to determine

concurrent validity to provide confidence in the psychometric properties of the new measure. Concurrent validity is based on a comparison between the results of two measures taken at relatively the same time (Portney & Watkins, 2009). Ideally, criterion validity would be established for supporting the use of the new measure. Criterion validity is not directly measureable and is inferred through comparison against a gold standard (Finch, Brooks, Stratford, & Mayo, 2002). For example, within gait analysis, three dimensional computerized gait analysis (3DGA) is considered the gold standard (Narayanan, 2007) and is frequently compared to observational gait analysis to suggest validity.

Reliability refers to the consistency that an instrument or a rater measures a given variable (Portney & Watkins, 2009). Test-retest reliability refers to the consistency of performance on a measure across a specified time frame and is vitally important in longitudinal measures. A measure with high test-retest reliability when re-administered to a child with CP soon after the initial administration should return similar, if not identical, scores as there should not be a change in the child's actual functioning in a short amount of time. An example would be measuring the same values for range of motion a few minutes apart, as true range of motion would not change in this time period. Rater reliability is also important to consider with outcome measures. Intra-rater reliability refers to the consistency that a single rater has when administering an outcome measure to the same subject on multiple occasions. Inter-rater reliability refers to the consistency that multiple raters have to each other when administering the same outcome measure to the same individual.

Beyond a measurement tool having evidence of validity and reliability, these psychometric properties also need to be applied to the population of interest to be considered appropriate measurement tools (Portney & Watkins, 2009). As walking is a major activity and

construct of interest within children with CP, and a focus of this dissertation project, a review of gait measures for children with CP was conducted. There are different types of measurements of gait, and different applications for these measurements, so no consensus was reached about any single “best” measurement of gait in children with CP and an expanded discussion of these measures is presented in Chapter 2.

Individuals who have poor selective voluntary dorsiflexion muscle control and drag their toes while walking are at increased risk of falls (Sung & Bang, 2000). Children who walk on their toes due to spasticity can have increased knee hyperextension during stance phase and reduced ability to utilize ankle movements for standing balance (Burtner, Woollacott, Craft, & Roncesvalles, 2007). Weakness of the primary ankle dorsiflexor (tibialis anterior) muscle combined with spasticity in the plantarflexor triceps surae muscle group (gastrocnemius, soleus, and plantaris) affects not only toe clearance, but also energy expenditure during gait (Ballaz, Plamondon, & Lemay, 2010; Dallmeijer, Baker, Dodd, & Taylor, 2011). As literature suggests that ankle dorsiflexion range of motion may be a key limiting variable in walking ability, assessment of ankle range of motion is an important aspect of therapy.

Goniometry is the most commonly used method for assessment of range of motion (ROM) in children with CP (Darrah, Wiart, Gorter, & Law, 2014) and is considered the current gold standard method of assessing ROM. It is not always practical for a single person to assess dorsiflexion range of motion in children with CP as tone or spasticity may create the need for using two hands to stabilize the foot and ankle with a second person to actually perform the measurement. By adding a second person, the possibility of error is increased. Therefore, there is a need for a single rater method for measurement of ankle dorsiflexion range of motion with good reliability and validity. Beyond difficulty of a single person assessing dorsiflexion range of

motion, there is a need for unbiased assessment of ankle range of motion when attempting to report true change. Bias can be reduced by blinding the rater, or creating a condition where the rater is unaware of any intervention that may have taken place prior to the assessment. One method for blinding is to use video recording during the range of motion assessment with post hoc measurement. There was no literature found to support this method and therefore a study was conducted to determine criterion validity, and test-retest, intra-, and inter-rater reliability of this video assessment method. That study and results are described in Chapter 3.

Though selection of appropriate measures to determine walking ability and establishment of a valid, reliable and unbiased way to assess ankle range of motion were important for this project, the main focus of this project was to evaluate change in walking through an intervention study. The intervention was designed to improve the use of muscles that control the ankle and foot position during walking. Specifically, a unique ‘NeuroGame Therapy’ (NGT) system, which utilizes surface electromyography (sEMG) biofeedback to improve motor control of muscles, was studied. This system was based on neural plasticity studies suggesting that permanent improvements with motor control can occur with sustained specific and relevant practice (Johnston, 2009; Kleim & Jones, 2008). Clinical studies have shown substantial improvements with motor control as a result of physical therapy that includes repetitive practice at specific tasks (Oujamaa, Relave, Froger, Mottet, & Pelissier, 2009; Urton, Kohia, Davis, & Neill, 2007). Success has also been seen with use of sEMG biofeedback to improve ankle range of motion and gait in children with hemiplegic CP (Binder, Moll, & Wolf, 1981). Dursun, Dursun, & Alican (2004) reported improvements in active ankle range of motion with the use of biofeedback and showed improvements in gait.

Given that previous literature reports success in the recruitment of the tibialis anterior muscle via training with sEMG, and that improved ankle dorsiflexion has had an impact on gait, we utilized NGT to train the bilateral tibialis anterior muscles. This system promotes salient and motivating practice in individuals with neuromotor dysfunction utilizing sEMG to control popular video games. In addition to simple training of the muscles, we chose to pair NGT play with immediate walking practice in order to promote carry-over of the sEMG training to the specific functional activity of primary interest in the study. The details of this study are presented in Chapter 4.

## Chapter 2:

### Clinical Gait Measures for Ambulatory Children with Cerebral Palsy: a Scoping Review

Torey Gilbertson, Kristie Bjornson, Brian Hafner, Sarah Westcott McCoy

#### Abstract

**Purpose:** Valid and reliable measures of walking ability are needed to document intervention effectiveness for ambulatory children with cerebral palsy (CP). Selection of measures suited to evaluating children with CP can be informed by evidence of a measure's clinical utility and psychometric performance in the population of interest. The purpose of this scoping review was therefore to identify clinical measures that have been used to evaluate gait of children with CP, to review measures' evidence of psychometric testing, and to determine which measures are most appropriate for use with children with CP in a clinical setting. **Methods:** PubMed, Web of Science, and PsycINFO databases were searched for measures with evidence of psychometric testing in children with CP. **Results:** Eleven measures suitable for clinical administration were identified across 22 articles. Only two measures had sufficient psychometric evidence to support clinical use in children with CP. **Conclusion:** The 6-Minute Walk Test and the Edinburgh Gait Scale are recommended for directly measuring walking capacity and rating walking by observation, respectively. Results of this scoping review suggest that, although a range of measures exist for evaluating pediatric gait, additional work is needed to assess and document measures' psychometric properties in children with CP.

## Introduction

Cerebral palsy (CP) is a group of permanent movement and posture disorders attributed to non-progressive damage to the fetal or infant brain that affects 3-4 of every 1000 school-aged children in the United States (Rosenbaum *et al.*, 2007; Aisen *et al.*, 2011). Although persons with CP may present with a variety of motor impairments, it is typically categorized by the dominant movement disorder (e.g., spasticity, ataxia, dystonia or athetosis), distribution of affected body parts (e.g., one, two, or four extremities), and/or topography of impairment (e.g., bilateral lower extremities, arm and leg on one side, etc.) (Rosenbaum *et al.*, 2007). Spasticity is common in children with CP and often impairs basic movements, such as walking (Flamand, Massé-Alarie, & Schneider, 2013; Sorsdahl, Moe-Nilssen, & Strand, 2008).

Walking is a fundamental motor skill that enables participation in numerous daily activities such as play and interaction with siblings or peers at home, school, and within the community (Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011). As such, gait training is a key therapy modality for physical therapists (Sorsdahl, Moe-Nilssen, & Strand, 2008). Therapists' use of standardized outcome instruments is critical for accurate assessment and documentation of a variety of treatment outcomes, including gait (Oeffinger *et al.*, 2008). Reliable and valid clinical tests or measurement tools are necessary for documenting change in walking ability, both within and across treatment sessions (Finch *et al.*, 2002). Standardized instruments selected to assess patient outcomes need to have sufficient evidence of psychometric testing (e.g., reliability, validity, and responsiveness) and evidence of measures' performance in specific populations or diagnostic groups is essential (Portney & Watkins, 2009). Further, information gained from measures is hypothesized to generalize to environments a child may encounter (e.g., home, school, and community) outside of the clinic.

Tests and measures that are often used to assess gait in children with CP include measures of body segment motions (e.g., joint kinematics) or other spatiotemporal outcomes (e.g., stride length, velocity, and cadence). Many measures are intended for administration in a laboratory environment. Fewer are suited to clinical administration where space, equipment, and personnel are generally limited. For most physical therapists, selection of measures will be restricted to those that can be administered in clinical settings (e.g., exam rooms, hallways, or training areas). In order to optimally evaluate longitudinal changes in gait function, measures are desired that are both suited to clinical settings and possess sound psychometric properties in the population of interest. Therefore, this scoping review was conducted to determine the measures most appropriate to use with children with CP, based on evidence of reliability and validity with this specific patient population. The primary aims of this paper are to 1) inform clinicians about measures suited to evaluating walking capacity and/or joint movement characteristics in clinical practice, and 2) discuss situations where use of these measures may (or may not) be considered best practice for evaluating the effect of interventions to enhance walking in children with CP.

## **Methods**

PubMed, Web of Science, and PsycINFO medical databases were searched to identify publications that described outcome measures suitable for assessing walking ability and specifically examined at least one aspect of reliability or validity in children with CP. These databases were chosen because they index journals that have historically published research evidence related to children with CP. Search terms related to the population and activity of interest, as well as evidence of psychometric testing, were used to locate candidate publications. The population of interest was identified using the phrase “cerebral palsy” and the acronym “CP”. To identify publications that described the activity of interest, the root terms “gait”,

“walk\*” and “ambulat\*” were selected. Lastly, to identify publications that presented new evidence of psychometric testing, the root terms “psychometric,” “valid\*,” and “reliab\*” were chosen. Phrases and keywords were combined to yield a single search strategy that was used across the selected databases: ("Cerebral Palsy" OR CP) AND (gait OR walk\* OR ambulat\*) AND (valid\* OR reliab\* OR psychometric\*). The authors elected to confine the search to literature published between January 1, 2003 and December 31, 2013 so as to identify outcome measures used in the most recent decade. *A priori* inclusion and exclusion criteria (Table 1) were defined to identify relevant publications.

Table 1. Inclusion and Exclusion Criteria for review publication selection.

Inclusion Criteria	Exclusion Criteria
<p>Publication criteria</p> <ul style="list-style-type: none"> <li>• Article published in English</li> <li>• Article describes an original research study</li> <li>• Article describes a psychometric evaluation of a clinical instrument designed to measure gait speed, distance, or quality via observation or standard video recording</li> </ul> <p>Participant criteria</p> <ul style="list-style-type: none"> <li>• Participants include ambulatory children with CP (mean age 3-18)</li> <li>• Participants are capable of independent, or modified independent walking (e.g., Gross Motor Functional Classification System [GMFCS] levels I-III)</li> </ul> <p>Instrument criteria</p> <ul style="list-style-type: none"> <li>• The described instrument can be administered or scored by a physical therapist</li> </ul>	<p>Instrument criteria</p> <ul style="list-style-type: none"> <li>• The described instrument requires costly investments in equipment, infrastructure, or other resources that would not be readily available in a clinical environment (e.g. three-dimensional gait analysis, surface electromyography, etc.)</li> <li>• The described instrument is an indirect measure of walking ability (e.g., strength)</li> <li>• The described instrument measures walking ability as part of overall assessment (e.g., Gross Motor Function Measure or Pediatric Evaluation of Disability Inventory)</li> <li>• The described instrument is a self- or proxy-reported measure</li> </ul>

Titles and abstracts were screened to identify candidate publications and eliminate those that clearly did not meet the described inclusion and exclusion criteria. The full text of each remaining publication was then reviewed, using the specified selection criteria to identify articles that were ultimately included in this review. As part of the evaluation, pearling (inspection) of the references was conducted and appropriate articles were included. A separate search of each measure identified was also conducted to facilitate discussion of the measure’s clinical utility.

Identified measures were grouped into categories (i.e., ‘Direct measurement’ or ‘observed rating’), based on the type of instrument. ‘Direct’ measurements included instruments designed to assess distance walked over a set amount of time (e.g., 6 minutes) or time required to walk a set distance (e.g., 10 meters). ‘Observed rating’ measures included clinical tools developed to describe joint or body segment position at various phases of gait via visual observation.

Elements extracted from each publication included name of the described outcome measure(s), number of participants and/or raters, participants’ ages (range and mean), percentage of males, Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997; Palisano et al., 2008) level of participants, distribution of involvement, other participant diagnoses, test-retest reliability, inter-rater reliability, intra-rater reliability, and evidence of validity. Relative strength of each measure’s psychometric properties was determined using rating scales defined by Portney & Watkins (2009). The research design (e.g., randomized controlled trial, case study) and statistical method (e.g., intraclass correlation coefficient and percentage of agreement) were also identified during appraisal. Recommendations for clinical use of measures in each category were made by the review authors based on the number of children with CP involved in the reviewed studies, the described research designs, the quality of the reported validity and reliability testing, and the discussion of the measures by the study investigators.

## **Results**

The search yielded a total of 22 articles and 11 different measures (Figure 1). Identified ‘direct measurements’ included the 1 minute walk test (1MWT), the 10 meter walk test (10mWT) or 10 meter fast walk test (10mFWT), the 6 minute walk test (MWT), the 10 minute walk test (10MWT), and the timed up and go (TUG). ‘Observed rating’ measures identified included the Edinburgh Gait Scale (EGS), Observational Gait Analysis (OGA), Observational

Gait Scale (OGS), Physicians Rating Scale (PRS), Salford Gait Tool (SF-GT), and Visual Gait Assessment Scale (VGAS). Brief descriptions of each measure, including scoring instructions, were created (Table 2). A summary of published evidence, including populations studied, reported psychometric properties, and notes on clinical applications was similarly developed to facilitate critical review of the measures included in this review (Table 3).

Figure 1. Search strategy and selection process.

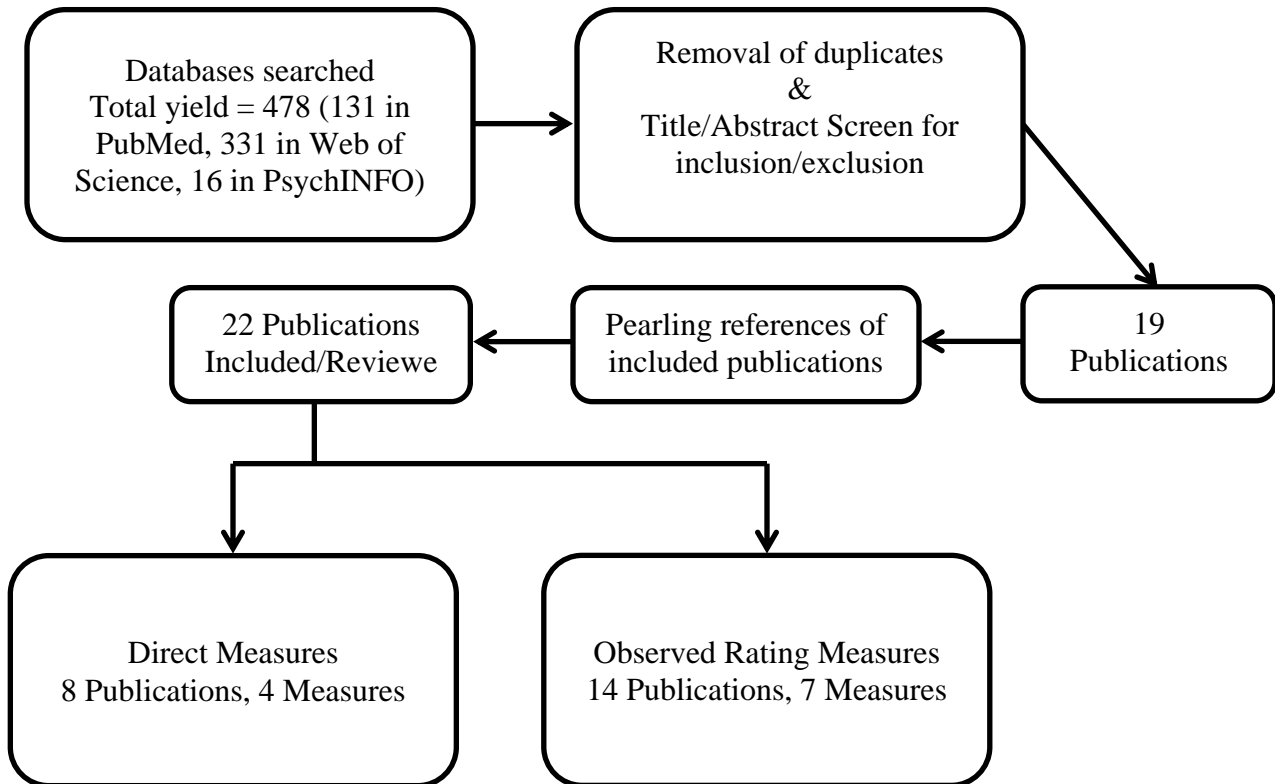


Table 2. Descriptions of Gait Measures

Measure	Scoring	Description
10-meter fast walk test (10mFWT)	Time (ratio)	Test representing the minimum distance for functional ambulation which measures the time required to walk 10 meters (from initiation of first step to lead foot crossing finish line).
1-Minute Walk Test (1MWT) or 1-Minute Fast Walk Test (1MFWT)	Distance (ratio)	Test of walking ability. Participants instructed to walk as fast as they can for 1 minute without running. Track set up to allow participants to walk in loop rather than abrupt change in direction. Total distance traveled in 1 minute is measured and reported.
6-Minute Walk Test (6MWT)	Distance (ratio)	Self-paced test of walking ability/endurance. Participants are instructed to walk as far as they can in 6 minutes. Walk either in circle or between 2 lines. Status and standardized encouragement provided every 30 seconds. Total distance traveled in 6 minutes is measured and reported.
10-Minute Walk Test (10MWT)	Distance (ratio)	Self-paced test of walking ability/endurance. Participants are instructed to walk as far as they can in 10 minutes. Walk either in circle or between 2 lines. Total distance traveled in 10 minutes is measured and reported.
Edinburgh Gait Score (EGS)	Rating total (ordinal)	Visual gait analysis using video recordings in all planes developed specifically for children with CP. 17 variables of gait for only one leg scored 0=typical, 1=moderate deviation, or 2= severe deviation. Summed to provide a total score.
Gillette Functional Assessment Questionnaire (FAQ)	Rating scale (ordinal)	Self- or proxy report measure of functional capability. 10-point scale from 0="Cannot take any steps at all" to 10="Walks, runs, and climbs on level and uneven terrain without difficulty or assistance".
Observational Gait Analysis (OGA)	Rating (nominal )	Triplanar visual gait analysis looking at 10 dimensions of walking and rating each leg independently. Observations are recorded as normal or deviated to one direction or the other.
Observational Gait Scale (Boyd and Graham version) (OGS)	Rating total (ordinal)	Visual gait analysis from sagittal and coronal plane video. Consists of 8 sections and a maximum of score of 22 per leg. 6 sections on body motion, 1 on assistive device, and one rating change. Mainly 0-3 scoring with higher scores representing more "normal" gait.
Physician Rating Scale (PRS)	Rating (ordinal)	Visual gait analysis from sagittal video. Several versions with dimensions including: amount of crouch, hip flexion in stance, knee flexion in stance, ankle dorsiflexion in stance, knee recurvatum, foot contact, and change.
Salford Gait Tool (SF-GT)	Rating total by joint (ordinal)	Visual gait analysis from sagittal plane video. Evaluates ankle, knee and hip position at 6 events during gait (Initial contact, end double support, mid stance, start double support, toe-off, mid swing). Ratings for each position: -2, -1, 0, 1, 2.
Timed Up & Go Test (TUG)	Time (ratio)	Test where child is timed from time that child leaves seat with backrest but no arms, walks to touch a target placed on the wall 3m away and returns to sitting in seat.
Visual gait assessment scale (VGAS)	Rating by item (ordinal)	Visual gait analysis using video recordings (sagittal plane). 7 parameters of joint position during various phases of gait are rated on scores from 1-3, 1-4, or 1-5 based on parameters. 1's usually represent restrictions or weakness. 4's or 5's may indicate hypermobility or other abnormalities (based on each parameter).

Table 3. Evidence table of psychometric properties of clinical gait measures.

Measure	Reference	N	Age	Topography	GMFCS	Validity Reporting	Reliability Reporting
10mWT	Thompson <i>et al.</i> (2008)	31	4:3-18:2, 9:5 (3:7)	D, H, Q, T, O	I – III	GMFCS level: paired <i>t</i> , <i>p</i> <0.015	Test-retest: 10.6 days; ICC(2,1), 0.81 (MDC-12.2)
1MWT	Chong <i>et al.</i> (2011)	60	5-18, 11.2	D, H, Q, O	I – IV	GMFCS, <i>r</i> = -0.75; 6MWT, <i>r</i> = 0.81, ABILOCO kids, <i>r</i> = 0.70	Test-retest: 1 week; ICC, 0.94 (MCD=13.1m)
	McDowell <i>et al.</i> (2009)	17	3 – 18, 12:8	D	I – III		
	Kerr <i>et al.</i> (2007)	46	11.68 (3.47)	D	I – IV	O <sub>2</sub> consumption: Quadratic <i>r</i> <sup>2</sup> = 0.477, <i>p</i> <0.001	
6MWT	McDowell <i>et al.</i> (2005)	34	4-16 years	D, Q	I – IV	GMFMS-88: <i>r</i> = 0.92, <i>p</i> <0.001	Test-retest: 10.6 days; ICC (2,1), 0.98 (MDC-54.9)
	Chong <i>et al.</i> (2011)	60	5-18, 11.2	D, H, Q, O	I – IV	GMFCS, <i>r</i> = -0.75; 1MWT, <i>r</i> = 0.81, ABILOCO kids, <i>r</i> = 0.70	
	Thompson <i>et al.</i> (2008)	31	4-18, 9:5 (3:7)	D, H, Q, T, O	I – III	GMFCS level: paired <i>t</i> , <i>p</i> <0.015	
	Maher <i>et al.</i> (2008)	41	11-16	D, H, Q, O	I – III		
10MWT	Leunkeu <i>et al.</i> (2012)	24	10-16, 14.2 (2.0)	D, H	I – II	VO <sub>2</sub> peak: ICC = 0.948, <i>p</i> <0.001	Test-retest: 1 week; ICC, 0.87
	Pirpiris <i>et al.</i> (2003)	29 (18 CP)	6-16, 11.5 (3.5)	D, H	N/R	3DGA speed: <i>r</i> = 0.65 (0.56 – 0.75)	Test-retest: 1 week; ICC, 0.91 (0.77-0.99)
EGS	Bella <i>et al.</i> (2012)	8 P, 3 R	10:6 (3.8)	D	I & II		Intrarater: <i>k<sub>w</sub></i> = 0.20 - 0.81 Interrater: <i>k<sub>w</sub></i> = 0.47 – 1.00
	Viehweger <i>et al.</i> (2010)	10 P, 8 R	9-16	D	N/R		Intrarater: ICC< 0.59 – 0.96 Interrater: <i>k<sub>w</sub></i> agree, 21.4 – 67.9%
	Hillman <i>et al.</i> (2007)	58	4-39, 12	D, H, T, Q	N/R	FAQ: <i>r<sub>s</sub></i> = -0.52; GGI: <i>r<sub>s</sub></i> = 0.83-0.89	
	Read <i>et al.</i> (2003)	5 (4 CP)	9-15	N/R	N/R	3DGA: agree = 47-83%, mean 64%	Intrarater: LSD; 2.63 – 4.01, mean 3.20 Interrater: agree; 55-96%, mean 70%
OGA	Kawamura <i>et al.</i> (2007)	50 P, 4 R	>8	D	I & II	3DGA: <i>k</i> = 0.01-0.65; M <i>X</i> <sup>2</sup> = 0-10.67	Interrater: <i>k</i> = 0.25 -0.88 (all <i>p</i> ≤ 0.01)
OGS	Borel <i>et al.</i> (2011)	12 P, 20 V	5-14, 8.9 (3.0)	D, H	N/R		Interrater: <i>k<sub>w</sub></i> = 0.16 - 0.80
	Mackey <i>et al.</i> (2003)	20	6-21, 12	D	I – III	3DGA: <i>k<sub>w</sub></i> = 0.38 – 0.94	Intrarater: <i>k<sub>w</sub></i> = 0.30 – 1.00 Interrater: <i>k<sub>w</sub></i> = 0.29 – 1.00
PRS	Wren <i>et al.</i> (2005)	30 P, 4 R	5-20	N/R	N/R	Kinematics: ANOVA <i>p</i> <0.0001 for all joint	Intrarater: <i>k<sub>w</sub></i> = 0.26 - 0.81 Interrater: <i>k<sub>w</sub></i> = 0.40–0.71 (live) 0.36-0.74 (video)

Table 3. Evidence table of psychometric properties of clinical gait measures. continued

SF-GT	Toro <i>et al.</i> (2007a)	13 V (11 CP), 23 R	6-16, 9:6	D, H, Q	N/R		Intrarater: agree means (%); h 72, k 78, a 73* Interrater: agree means (%); h 77, k 81, a 75*
	Toro <i>et al.</i> (2007b)	13 V (11 CP), 23 R	6-16, 9:5	D, H, Q	N/R	Kinematics: LSD, 1.41-27.14	
TUG	Williams <i>et al.</i> (2005)	41 (33 CP)	3-17:6, 8:7 (4:1)	D, H, Q	I – III	GMFCS levels: K-W ANOVA, p = 0.001	Test-retest: 1 min; ICC (1,1) 0.99 Test-retest: 20 min; ICC (1,3) 0.98
	Dhote <i>et al.</i> (2012)	30	2-12: 8.16 (2.76)	N/R	I - III		Test-retest: 30 min; ICC, 0.99; 1 week; ICC, 0.99
VGAS	Dickens <i>et al.</i> (2006)	31 P, 2 R	5-17, 10.6	H	I – IV	3DGA: $k = -0.11 - 0.51$	Intrarater: $k = -0.04 - 0.86$ Interrater: $k = 0.44-0.89$
	Bella <i>et al.</i> (2012)	8 P, 3 R	10:6 (3.8)	D	I & II		Intrarater: $k_w = 0.54 - 1.00$
	Brown <i>et al.</i> (2008)	4 P, 10 R	N/R	N/R	N/R		Intrarater: $k_w = 0.54 - 1.00$ , COR, 4.06 – 5.94 Interrater: agree, 33.3 -100%

Table Legend:

N: (P = total participants, R = Raters, V = videos of gait, CP = participants with CP); **Age** (years: months): reported in range, mean (SD); **Topography**: D=Diplegia, H=Hemiplegia, Q=Quadriplegia, T=Triplegia, O=Other; **GMFCS**: reported in levels represented by the population.

**Measure Abbreviations**: **10mWT** – 10 meter Walk Test, **1MWT** – 1 Minute Walk Test, **3DGA** – 3dimensional computerized gait analysis, **6MWT** – 6 Minute Walk Test, **10MWT** – 10 Minute Walk Test, **EGS** – Edinburgh Gait Scale, Questionnaire, **FAQ** – Gillette Functional Assessment Questionnaire, **GMFM-88** Gross Motor Function Measure (88 item), **MCD** – minimal detectable difference, **OGA** – Observational Gait Analysis, **OGS** – Observational Gait Scale, **PRS** – Physician Rating Scale, **SF-GT** – Salford Gait Tool, **TUG** – Timed Up and Go, **VGAS** – Visual gait assessment scale, **VO<sub>2</sub> peak** – peak oxygen consumption

**Validity**: comparison test: statistical test, values;

**Reliability**: Test-Retest Reliability: time frame between tests; statistical test, values (95% CI); other reliability: statistical test, value, mean (significance),

Statistical test abbreviations: **r** – Pearson r, **r<sub>s</sub>** – Spearman r, **k** – Kappa, **k<sub>w</sub>** – Weighted Kappa, **M X<sup>2</sup>** – McNemur's Chi Square, **ANOVA** – Analysis of Variance, **LSD** – Least Significant Difference, **K-W** – Kruskal Wallis, **ICC** – Intraclass Correlation Coefficient, **COR** – coefficient of reliability, **agree** – percentage of agreement

\* - abbreviations: h = hip, k = knee, a = ankle

## **Direct Measures**

### ***1 Minute Walk Test (1MWT)***

McDowell, Kerr, Parkes, and Cosgrove (2005) compared the 1MWT to Gross Motor Function Measure (GMFM) scores in 34 children. The GMFM is an evaluative measure with five dimensions: Lying and Rolling, Sitting, Crawling and Kneeling, Standing and Walking, and Running and Jumping (Russell et al., 1989; Russell et al., 2000). Distance on the 1MWT was significantly correlated with GMFM scores ( $r = 0.92$ ,  $p < 0.001$ ) (McDowell *et al.*, 2005). This evidence supports convergent validity. The investigators reported significant differences between 1MWT distances and GMFCS levels ( $p < 0.001$ ) with differences between those in levels II and III ( $p = 0.003$ ) and levels III and IV ( $p = 0.008$ ), but not significant between levels I and II ( $p = 0.42$ ). This indicates good evidence of discriminant validity for the 1MWT. Also, McDowell, Humphreys, Kerr, and Stevenson (2009) reported good test-retest reliability ( $ICC = 0.94$ ) with the 1MWT over a 1-week period in 17 children.

The 1MWT was reported by Chong, Mackey, Broadbent, and Stott (2011) to have good evidence of convergent validity through strong correlations between both the GMFCS ( $r = -0.75$ ) and ABILOCO-kids questionnaire of locomotion ability ( $r = 0.70$ ) using 60 children. This group also reported good concurrent validity via a strong correlations to the 6MWT ( $r = 0.81$ ).

Kerr, McDowell, and Cosgrove (2007) examined 46 children and reported a correlation between the 1MWT distances and oxygen consumption ( $r = 0.69$  [ $p < 0.001$ ]). Through evidence of strong concurrent validity, the 1MWT is not valid for assessing energy consumption.

### ***10 Meter Fast Walk Test (10mFWT) or 10 Meter Walk Test (10mWT)***

Test-retest reliability of the 10mFWT was examined by Thompson *et al.* (2008) on 31 children. They found very strong correlation of  $ICC(2,1) = 0.81$  at a mean of 10.6 days (7-31

day range). The test-retest reliability was inversely correlated with function (0.59 for GMFCS level I, 0.70 for level II, and 0.78 for level III).

### ***Six-Minute Walk Test (6MWT)***

The reliability and validity of the 6MWT was also assessed by Thompson *et al.* (2008) A 1-2 week test-retest ICC (2,1) of 0.98 was found for the same 31 children. Differences in 6MWT distances among GMFCS levels (I-III) using a paired t-test with correction for multiple comparisons ( $p < 0.015$ ) were reported. This demonstrates good discriminant validity. Maher, Williams, and Olds (2008), in a study of 41 older children, reported an ICC of 0.98 for 30 minute test-retest reliability. Similarly, Leunkeu, Shephard, and Ahmaidi (2012) studied 1-week test-retest reliability and reported an ICC of 0.87 on 12 older children. They also studied 24 older children to compare 6MWT performance to energy expenditure.  $VO_2$  peak was significantly correlated with 6MWT ( $r = 0.95$ ,  $p < 0.001$ ), supporting convergent validity. Good convergent validity between the 6MWT with GMFCS levels ( $r = -0.75$ ) and the ABILOCO-kids questionnaire ( $r = 0.70$ ) was also reported by Chong *et al.* (2011).

### ***10 Minute Walk Test (10MWT)***

In a study of 7-day test-retest reliability, in 29 children with either CP or meningomyelocele, Pirpiris *et al.* (2003) reported an ICC of 0.91 (95% CI: 0.77 - 0.99). The investigators also noted a significant difference ( $p < 0.01$ ) between 10MWT walking speed as compared to a multi-trial average speed of 10 meter walks evaluated using three-dimensional gait analysis (3DGA), suggesting poor criterion validity. Due to the different distances walked by children in these tests, the investigators hypothesized that the 10MWT may be a better indicator of community walking speed than the 10 meter walking test.

### ***Timed Up and Go (TUG)***

The TUG was designed to assess mobility and dynamic balance in various populations (Podsiadlo & Richardson, 1991). It measures the time required for an individual to rise from a seated position, walk three meters, turn around, and return to a seated position. Williams, Carroll, Reddihough, Phillips, and Galea (2005) examined the TUG in a study with 41 children, 33 of whom had CP. The investigators conducted 2 initial trials (time 1) and 2 more trials after 10-20 minutes (time 2) and documented immediate test-retest reliabilities (ICC [1,1] = 0.98 within both times 1 and 2) and same-day retest between time 1 and 2 (ICC [1,3] = 0.99) (Williams *et al.*, 2005). These investigators found significant differences ( $p = 0.01$ ) in TUG times between levels (I-III) using a Kruskal-Wallis ANOVA, which suggests evidence of discriminant validity.

Dhote, Khatri, and Ganvir (2012) also studied test-retest reliability of the TUG in 30 children. They performed both 30 min and 1 week retests and reported ICCs of 0.99 for each.

### **Observed Rating Measures**

#### ***Edinburgh Gait Scale (EGS)***

The EGS (also known as the Edinburgh Visual Gait Analysis Interval Test [GAIT]) was developed as an observational gait analysis tool. It is composed of 17 ratings of joint position (e.g., amount of flexion) for each lower limb evaluated on six anatomical sites: trunk, pelvis, hip, knee, ankle and foot (Read, Hazlewood, Hillman, Prescott, & Robb, 2003). The parameters are analyzed in both sagittal and frontal planes through videotape observation. Bella, Rodrigues, Valenciano, Silva, and Souza (2012) compared the EGS to an Observational Gait Scale (Araujo, Kirkwood, & Figueiredo, 2009) and the Visual Gait Assessment Scale (VGAS) (Dickens & Smith, 2006) in 8 children. There was high inter-rater agreement ( $k = 0.47 - 1.00$ ) among ratings made using all three scales (i.e., OGS, VGAS, and EGS). Most intra-rater combinations had

moderate to excellent weighted kappa index frequencies ( $k = 0.20 - 0.81$ ). The reliability of the EGS using 10 videos of children was studied by Viehweger *et al.* (2010) who found moderate to very strong intra-rater reliability (ICC = 0.59 – 0.96) and weak to strong inter-rater agreement (21.4 – 67.9%) among eight raters with varying degrees of clinical and gait observation experience. Better agreement was found between items in the stance phase than those in the swing phase. Greater correlations with increasing clinical and gait analysis experience suggested that those with more experience often yielded more consistent scores (Viehweger *et al.*, 2010).

Hillman, Hazlewood, Schwartz, van der Linden, and Robb (2007) compared the EGS to the Gillette Gait Index (GGI) and the Gillette Functional Assessment Questionnaire (FAQ) in 58 children and adults. The correlation between the EGS and GGI was very strong ( $r_s = 0.83 - 0.89$ ), documenting criterion validity as the GGI was developed from the ‘gold standard’ 3DGA. The correlation between the EGS and the FAQ was lower ( $r_s = -0.52$ ). The observed inverse relationship was expected as higher FAQ scores indicate higher functional walking and higher EGS scores show higher deviation from “normal”, suggestive of evidence of convergent validity.

In a comparison in five children by Read *et al.* (2003), the EGA and 3DGA had varying levels of agreement between the different joints (47 – 83% agreement, mean 64%). Intra-rater reliability showed Least Significant Difference (LSD) differences between 2.63 and 4.01 (mean of 3.20). Inter-rater agreement among five clinicians ranged from 55 to 96% (mean of 70%).

### ***Observational Gait Analysis (OGA)***

The OGA consists of 10 items evaluating general positioning of the pelvis, hip, knees, and ankle at various times during the gait cycle and in all three planes of motion (Kawamura *et al.*, 2007). Kawamura *et al.* (2007) assessed 10 periods in the gait cycle using OGA in 50 older children. Four different raters evaluated position (joint angle) of the hip, knee, and ankle as

primary interest. Very weak to strong agreement ( $k = 0.01 - 0.65$ ) was reported between OGA and 3DGA. Joint and period of gait influenced inter-rater agreement which was weak to very strong ( $k = 0.25 - 0.88$ ). Between OGA and 3DGA, the highest agreement ( $k = 0.21 - 0.65$ ) was reported for the knee joint angle at three different periods of gait (initial contact, terminal stance, and initial swing). Criterion validity for OGA appears to be dependent upon rater and joint.

### ***Physician Rating Scale (PRS)***

The PRS was initially developed for evaluation of gait in children post botulinum toxin A injection (Mackey, Lobb, Wait, & Stott, 2003). The PRS emphasizes position of the foot and knee in stance with six categorical ratings. Wren *et al.* (2005) modified the PRS to include hip rating and greater specify knee and ankle rating in a study of 30 children and young adults. Ratings of joint positions from four raters were compared to positions simultaneously-collected with 3DGA. Differences between visual rating and 3DGA were found to be significant for all joints ( $p < 0.0001$ ) indicating poor criterion validity. The investigators also assessed inter- and intra-rater reliability among four raters using live observations, full-speed and slow motion video. Inter-rater agreement varied across conditions with the highest agreement on crouch ( $k = 0.71$ ) and hip ( $k = 0.40$ ) using live observation. Slow motion video agreement was highest for foot contact ( $k = 0.74$ ) and dorsiflexion (DF) ( $k = 0.52$ ). Agreement for the knee ( $k = 0.65$ ) was highest using full speed video. Intra-rater agreement also varied ( $k_w = 0.50 - 0.78$  for foot contact, 0.71 - 0.80 for crouch, 0.26 - 0.44 for hip flexion, 0.60 - 0.86 for knee flexion, and 0.39 - 0.61 for DF).

### ***Observational Gait Scale (OGS)***

The OGS is a modification of the Physician Rating Scale (PRS) which includes eight sections evaluated in frontal and sagittal planes (22 points per leg) (Boyd & Graham, 1999) and

places a higher emphasis on the foot and knee during stance than the PRS (Mackey *et al.*, 2003). Mackey *et al.* (2003) studied the reliability of the OGS in 20 children and found moderate intra-rater agreement ( $k_w = 0.30$  to  $0.86$ ) of scores across joints was slightly higher than inter-rater agreement (mean  $k = 0.69$  versus  $0.62$ ) (Mackey *et al.*, 2003). Good criterion validity was documented by moderate to high agreement between OGS and 3DGA ( $k_w = 0.38 - 0.98$ ).

Borel, Schneider, and Newman (2011) used the OGS in 12 children, assessing 20 gait videos. Traditional visual rating of OGS was compared to ratings using Dartfish analysis software (Dartfish USA; Alpharetta, Georgia). Inter-rater reliabilities with visual rating ( $k = 0.16 - 0.80$ ) and using Dartfish ( $k = 0.35 - 0.85$ ) were reported. Thus, the analysis software was found not only to be more efficient in terms of time during assessment, but also more reliable.

### ***Visual Gait Assessment Scale (VGAS)***

The VGAS is also adapted from the PRS (Brown, Hillman, Richardson, Herman, & Robb, 2008). It consists of seven parameters (hip and knee position in terminal stance and mid swing, initial foot contact, foot contact in stance, and timing of heel rise). Two raters measured walking ability in 31 children, using the VGAS and 3DGA (Dickens & Smith, 2006). Agreement between measurement methods ranged from  $k = -0.11$  to  $0.51$ , depending on the parameter, making criterion validity very weak to moderate. Intra-rater agreement ( $k = -0.04 - 0.86$ ) and inter-rater agreement ( $k = 0.44 - 0.89$ ) were also reported. Dickens and Smith (2006) noted the ratings at the hip were the least reliable. The VGAS and EGS were compared by Bella *et al.* (2012) in 8 children. Intra-rater reliability among three raters ranged from  $k_w = 0.54$  to  $1.00$ . In another study of four children and 10 raters of differing experience, Brown *et al.* (2008) found Bland-Altman coefficients of intra-rater reliability of  $4.06$  for experienced raters and  $5.94$  for inexperienced raters suggesting the VGAS was more reliable with experienced observers. They

also determined that more experienced raters generally had higher intra-rater (50.0% - 87.6% versus 41.7 - 83.3%) and inter-rater (45.8% - 100.0% versus 38.3% - 91.7%) agreements on ratings. Agreements were higher for ratings at the ankle and foot than those at the knee or hip.

### ***Salford Gait Tool (SF-GT)***

The SF-GT is a sagittal plane observational measure used to evaluate hip, knee, and ankle positioning at six specific events during the gait cycle (i.e., initial contact, end double support, mid stance, start double support, toe-off, and mid-swing). Toro, Nester, & Farren (2007b) compared the SF-GT to 3DGA and found least significant differences between SF-GT and 3DGA varied from 1.41 to 27.14. This suggests moderate to low evidence of criterion validity. Those investigators also examined inter- and intra-rater reliability of SF-GT ratings with 13 children and 23 raters evaluating videos of walking. The inter-rater reliability was high in nearly all instances with reported agreement at or over 63% in nearly every joint and position rating with means of 77% at the hip, 81% at the knee, and 75% at the ankle (though lower in children with severe crouch [56%]). Mean intra-rater agreement was reported as 72% for the hip, 78% for the knee, and 73% for the ankle (overall agreement between 51% and 90%) (Toro, Nester, & Farren, 2007a).

## **Discussion**

Selection of measures to evaluate the effectiveness of an intervention is essential for therapists to assess and document changes in patient function. The goal of this review was to identify and discuss those measures most suited to evaluating walking performance and quality of movement in children with CP in a clinical setting. Review and analysis of measures' clinical utility and psychometric performance in children with CP revealed that the 6MWT and EGS are

well suited to measurement of children with CP. However, other measures may be appropriate for use in select situations. Recommendations for use of each measure are discussed below.

### **Direct Measurements**

Of tools classified as direct measurements, the 6MWT best documented evidence of reliability and validity in the measurement of walking capacity of children with CP compared to other measures, and is therefore recommended for most children with CP. Three studies, all with adequate sample sizes and similar levels of test-retest reliability, suggest the 6MWT as a highly reliable test for longitudinal assessments (Leunkeu *et al.*, 2012; Maher *et al.*, 2008; Thompson *et al.*, 2008). The Thompson *et al.* (2008) study allows for generalization to other therapists based on ICC model used (2,1). The high correlation with  $VO_2$  also suggest that the 6MWT may be an effective measure of energy expenditure (Leunkeu *et al.*, 2012) and could therefore also be used to measure general physical functioning. Lastly, there is also evidence of discriminant validity between the 6MWT and GMFCS level (Chong *et al.*, 2011). Though the 6MWT has better psychometric properties than the 1MWT, children who are not able to walk for 6 minutes due to physical or cognitive functioning may be better suited for evaluation using the 1MWT.

The 1MWT test, while clinically-friendly, should be used with caution as test-retest reliability was assessed in only one study and an unreported ICC model was used (McDowell *et al.*, 2009). Chong *et al.* (2011) found a high correlation between the 1MWT and the 6MWT in children with CP, suggesting that the 1MWT may be as useful for assessing walking capacity, but requires less time to administer. However, further research is needed to substantiate this claim. The correlation between the 1MWT and  $O_2$  consumption was low, so may not be a sound proxy for energy expenditure (Kerr *et al.*, 2007). Studies showed positive relationships to the 1MWT with GMFM scores (McDowell *et al.*, 2005) and GMFCS level (Chong *et al.*, 2011).

Based on this review, the 10mFWT (or 10mWT) is a psychometrically appropriate measure for evaluation of speed of ambulation over short distances (e.g., within a child's house) and had good test-retest reliability with the ICC model used (2,1) suggesting generalizability to other therapists (Thompson *et al.*, 2008). This test was also strongly related to physical functioning based on the GMFCS level, especially for those at GMFCS level III. Though the 10mFWT shows promise for use in children with CP, caution should be taken with its use until further evidence is available as psychometric evidence was derived from only one study.

The published 10MWT literature suggests evidence of good test-retest reliability, using an ICC model that supports generalizability (Pirpiris *et al.*, 2003). However, the 10MWT may not be appropriate for children functioning at a lower physical level (e.g., GMFCS III) due to the difficulties of sustaining walking for 10 minutes (Pirpiris *et al.*, 2003). Based on the current evidence for children, the 10MWT is not recommended for clinical use at this time as it does not appear to possess superior psychometric properties to the 6MWT.

The TUG has been studied frequently in children with CP, though only two studies are more recent. These studies included large sample sizes and employed ICC models (i.e., 1,1 and 1,3) suited to generalizability of the results (Dhote *et al.*, 2012). The TUG showed promising evidence of test-retest reliability with assessments occurring on the same day (Williams *et al.*, 2005) and a week apart (Dhote *et al.*, 2012). The TUG is recommended as a longitudinal assessment tool, due to test-retest reliability over both short and longer periods. Since the TUG requires a child to stand-up, sit down, and turn 180 degrees, it is not solely a measure of ambulation. However, it may be a useful and reliable tool to assess general mobility.

### ***Observed Rating Measures***

Of the observed rating measures reviewed, the EGS appears to be the most appropriate tool for visual kinematic assessment of gait in a clinical setting. Psychometric testing of the EGS showed good intra-rater reliability (Bella *et al.*, 2012; Read *et al.*, 2003; Viehweger *et al.*, 2010), the strongest inter-rater reliability of the measures reviewed (Bella *et al.*, 2012; Read *et al.*, 2003; Viehweger *et al.*, 2010), and good concurrent and criterion validity (Hillman *et al.*, 2007; Read *et al.*, 2003). All studies that examined the EGS had moderate-to-strong intra-rater reliability. However, these studies included relatively small sample sizes (i.e.,  $n = 4$  (Read *et al.*, 2003), 8 (Podsiadlo & Richardson, 1991), and 10 (Dhote *et al.*, 2012)) and thus may limit generalizability of the reliability reported. Inter-rater reliability of the EGS varied and was generally lower than intra-rater reliability. However, evidence of EGS inter-rater reliability was still stronger than the other observed rating measures reviewed. The EGS also had the highest correlations reported for concurrent and criterion validity, suggesting it may most closely measure the underlying construct of gait. Bella *et al.* (2012) reported that the EGS was the most user-friendly tool (i.e. easiest to understand and use) of the three they evaluated (EGS, VGAS, and Brazilian OGS). The EGS was also the only measure for which minimal detectable difference (MDD) was reported where Read *et al.* (2003) showed that a change in 3 units is likely indicative of true change. Lastly, the EGS has a greater number of joint position ratings than the other observational gait assessments found in this study providing more information for the clinician. This greater number of ratings extends the time required to score the test to about 25 minutes for scoring (Maathuis, van der Schans, van Iperen, Rietman, & Geertzen, 2005). Unlike some of the other measures, the EGS also used both sagittal and coronal plane video, which may be of interest to clinicians who want assess coronal plane aspects of gait (i.e. hind foot valgus, foot rotation, and

lateral trunk shift), requiring more recording and evaluation. After training, the high intra-rater agreement indicates the EGS could be useful for longitudinal assessment of children with CP.

The OGA was reviewed in only one study, though was suggested that it had slightly better inter-rater reliability than the EGS. It was less correlated to the gold standard of 3DGA and may therefore not as well reflect the measurement of gait kinematics (Kawamura *et al.*, 2007). Since the OGA was only reported in one study over the period of this review, these authors feel its use is not well justified for clinical use when compared to the EGS.

Studies examining the psychometric properties of the OGS reported mixed results (Borel *et al.*, 2011; Mackey *et al.*, 2003). Though the OGS was highly correlated to the gold standard of 3DGA, the intra-rater and inter-rater reliabilities were quite mixed, depending on the rated joint. Implications for use of this instrument include, for example, it may not reliably measure base of support or hind foot position (Mackey *et al.*, 2003), which could be important with interventions that target hip abduction or evaluating use of foot orthotics. This measure was reported to have increased efficiency and reliability when used with companion analysis software (e.g., Dartfish) (Borel *et al.*, 2011). Thus, it is recommended to use analysis software for improved reliability when using the OGS clinically. Time for administration of this measure was not reported.

The SF-GT shows promise as a measure for children with CP, but the single study and reliability statistic (i.e., percent agreement) used weakens the interpretation of repeatability. The study included a fairly small n (i.e., 13 videos), but a high number of raters (i.e., 23 therapists) (Toro, Nester, & Farren, 2007a). This number of raters may make the inter-rater reliability agreements more generalizable, despite the small sample of source videos. The wide variation between SF-GT scores and 3DGA may suggest the SF-GT is slightly less robust of an assessment than the EGS. However, the SF-GT may be more useful than the EGS if sagittal-

plane kinematics are of interest (e.g., hip or knee joint position beyond mid-stance or mid-swing phases of gait).

Psychometric testing of the VGAS across multiple studies revealed lower inter- and intra-reliability than the EGS (Bella *et al.*, 2012; Brown *et al.*, 2008; Dickens & Smith, 2006). Similarly, the evidence of validity was less convincing than that identified for the EGS. Bella *et al.* (2012) stated the VGAS was less user-friendly than the EGS. Therefore, the VGAS may not be clinically appropriate as a longitudinal measure of gait kinematics in children with CP.

Choosing the correct assessment tool is directly related to the goals established and the focus of intervention. Time- or distance-based measures are recommended for assessing walking capacity or speed, though are unable to provide information on movement quality. If gait quality is a goal and focus of intervention, then use of observed rating measures is recommended. The EGS, for example, could be used to assess heel position during initial contact and document changes associated with orthoses or intervention intended to improve ankle dorsiflexion. While choice of an appropriate measure is ultimately at the discretion of the therapist, the results of this review can be used to guide their selection.

The aforementioned suggestions for use of gait measures in children with CP were derived from careful analysis and review of published evidence within this scoping review. However, the authors acknowledge that several limitations exist. First, this review was not a systematic review and, although rigor of the included publication was analyzed, publications were not scored using an established methodological appraisal tool. Extraction of data from the publications was performed by only one author (TG), which could bias the evidence selected for review. The evidence search also included only three databases and additional evidence may

have been identified if other databases (e.g., CINAHL) were used. Lastly, sensitivity to change was rarely reported, therefore recommendations were not based on this psychometric property.

## **Conclusion**

Based on the psychometric properties assessed in this review, the 10mFWT appears to be a sound measure to assess peak gait speed in children with CP. Similarly, the 6MWT appears to be the most useful direct measurement to assess walking ability over longer distances and is recommended over the 1MWT and 10MWT due to stronger evidence of validity and reliability. At this time, the 1MWT may only be recommended for children with CP who may not be able to complete the 6MWT. Similarly, the 10mFWT and the 6MWT are recommended over the TUG when the primary goal is to measure walking ability, as the TUG requires transition activities.

The EGS appears to be the most psychometrically sound measure for clinical evaluation of gait quality in children with CP. It has been reported as the most user-friendly observed rating measure and has the highest reliability of the measures reviewed. The EGS closely correlates with the gold standard of 3DGA and integrates both sagittal and coronal plane observations. Regardless of the observed rating measure used, the use of video recording is advised over real-time measurement. Supplementary assessment with companion analysis software is also recommended for enhanced reliability of the evaluation. Evidence suggests that reliability of any observational rating measure may be higher when performed by a consistent, experienced rater.

Of the reviewed gait measures, few have substantial documented psychometric evidence among children with CP. It is vital that measures intended for longitudinal use are capable of accurately reflecting true change over time. The reliability of any measure only reflects the population studied and may only be confidently used in children closely resembling those studied in terms of age, topographical distribution, movement disorder, and GMFCS level. Thus,

further psychometric testing is needed to establish the acceptable evidence of reliability and validity of clinical gait measures for children with CP.

### Chapter 3:

#### Use of Video Recording to Determine Ankle Dorsiflexion Range of Motion in Both Children with Typical Development and Those with Cerebral Palsy

Torey Gilbertson, Sarah Westcott McCoy, Brian Hafner, Chet Moritz, Kristie Bjornson

##### Abstract

**Background:** Ankle dorsiflexion (ADF) range of motion (ROM) has been associated with walking ability and postural stability in children with cerebral palsy (CP). However, few studies have examined the intra-rater, inter-rater, and test-retest reliability of ADF measurements with children with CP. Specifically, no studies have measured ADF using post hoc measurements from video recordings.

**Method:** ADF ROM was assessed two times about one week apart with standardized real-time measurement and concurrent video recording on 26 children, 9 children [8 males] with CP and 17 children [8 males] who were typically developing (TD); all 8-18 years old. Measurements were performed real-time by one rater and post hoc via video recordings by three raters.

**Results:** High correlations were found between real-time and post hoc video measurements. Intraclass Correlation Coefficients (ICC) for both intra-rater and inter-rater reliability were above 0.95 for trial averages and above 0.88 for individual trials. Test-retest reliability was also high with ICCs generally above 0.85 for individual trials and 0.90 for average measurements. Reliability was typically lower with the sample of children with CP, but ICCs were generally within 0.05 of those for the TD children.

**Conclusion:** Using video to record passive ROM for ADF in children is feasible and reliable when a standardized protocol is followed.

## **Introduction**

Assessment of range of motion (ROM) is an important aspect of rehabilitation services (Allington, Leroy, & Doneux, 2002; Darrah et al., 2014; Gatt & Chockalingam, 2011; McDowell, Hewitt, Nurse, Weston, & Baker, 2000; Mutlu, Livanelioglu, & Gunel, 2007; Ostensjø, Carlberg, & Vøllestad, 2004). Clinicians are able to measure changes in ROM as a basis for potential improvements in many functional outcomes through standard goniometry (Allington et al., 2002; Darrah et al., 2014; Gatt & Chockalingam, 2011; Mutlu et al., 2007; Sullivan et al., 2007). The assessment of ROM in children with cerebral palsy (CP) is meaningful as reduced ankle ROM may lead to pain (Kadhim, Holmes, & Miller, 2013) and functional limitations (Darrah et al., 2014). ROM examination can also be used to guide the design of orthotics, assess effectiveness of stretching programs, and even predict an individual's ability to perform various functional activities (i.e., sitting in chairs or being able to ascend and descend stairs with optimal body mechanics) (Sidaway et al., 2012). Ankle ROM assessment is especially critical for children with CP as literature suggests that ankle dorsiflexion (ADF) has an influence on the stability, quality, and efficiency of gait (Ballaz et al., 2010; Toner, Cook, & Elder, 1998) and reduced ADF may be associated with increased fall risk and functional ability (McDowell et al., 2000; Ostensjø et al., 2004). Though ROM measurements are crucial for clinical practice, they are not always precise with individual or different clinicians.

Knowledge of measurement error is essential when evaluating, interpreting, and applying ROM measurements in clinical practice (Allington et al., 2002; Mutlu et al., 2007; Sidaway et al., 2012; van Trijffel, van de Pol, Oostendorp, & Lucas, 2010; Wrobel & Armstrong, 2008). Error in measurements acquired by a clinician, like ROM, may be characterized by estimates of intra-rater, inter-rater, and test-retest reliability. Intra-rater reliability is the degree of agreement between measurements acquired by the same individual (clinician) over repeated assessments

(Portney & Watkins, 2009). High intra-rater reliability is desirable to provide confidence that the clinician is not introducing error into the ROM measurement. When multiple people are performing measurements on the same individual, a high degree of inter-rater reliability (degree of agreement between measurements acquired by different individuals) is critical for assurance that neither individual is introducing error into the measurement. High test-retest reliability (degree of agreement between measurements taken at specified times) is necessary to reflect true change across time or intervention rather than change due to measurement or other error. Precision within and between individuals is somewhat predicated on the stability of the test, or specific item of measurement and studies have helped establish this precision.

Researchers report greater intra-rater reliability than inter-rater reliability with all ROM measurements, and there is evidence supporting both high (Allington et al., 2002; Glanzman, Swenson, & Kim, 2008; Herrero, Carrera, Garcia, Gomez-Trullen, & Oliván-Blázquez, 2011; McWhirk & Glanzman, 2006; Mutlu et al., 2007; Wrobel & Armstrong, 2008) and low (Sidaway et al., 2012; van Trijffel et al., 2010) intra- and inter-rater reliability with goniometric measurements of the ankle. It is well documented, through test-retest reliability studies, that measurement of ankle ROM in children is variable, particularly in children with CP (Allington et al., 2002; Darrach et al., 2014; Kilgour, McNair, & Stott, 2003; Ostensjø et al., 2004; Rachkidi et al., 2009). It is hypothesized that spasticity and hypertonicity have a negative impact on test-retest reliability of ROM in children with CP (Allington et al., 2002; McDowell et al., 2000; Mutlu et al., 2007). Though the reliability of ROM in these children is debated, Kilgour et al. (2003) and Mutlu et al. (2007) report similar differences in test-retest variability between children with CP and typically developing children. Overall, the mean measurement error with ADF is reported as 3-5° (Allington et al., 2002; Wrobel & Armstrong, 2008), though those numbers do

not tell us the amount of change necessary to reflect true change rather than just measurement error.

Understanding of the factors related to the reporting of true change is also necessary. The standard error of measurement (SEM) describes the stability of response using the standard deviation of measurement errors in a set of repeated scores (Portney & Watkins, 2009). The SEM provides an amount of certainty that the true value of a measurement falls within a specific range. Minimal detectable difference (MDD) is critical to define the amount of change in a measurement necessary to reflect a true change in the item being measured after accounting for measurement errors (Portney & Watkins, 2009). By development of a method that would reduce error, the precision of measurement could improve and clinicians could be more certain that changes in measurement reflect true change.

An alternative to assessing ROM through real-time goniometry measurement is through post hoc measurements of video recordings. Using video to assess ROM may allow a single clinician to assess ROM during functional tasks and to create a reviewable record that may be assessed more easily. Few studies have been conducted to directly assess the validity or reliability of ROM measurements obtained through two-dimensional (2D) video recording. One study used 2D video recording for assessment of elbow, forearm, wrist, and finger motion using one of three ratings (i.e., I – 0°-30°, II – 30°-60°, III – 60°-90° for elbow ROM) based on amount of motion in adolescents with hemiplegia. The researchers found variability with overall lower values for both intra-rater ( $k = 0.50-0.92$  depending on joint and motion) and inter-rater reliability ( $k = 0.12-0.85$  depending on joint and motion) (Waters, Zurakowski, Patterson, Bae, & Nimec, 2004). Larson, Maanum, Frøslie, & Jahnsen (2012) found large discrepancies (i.e., 15°) between examination of 2D sagittal plane video recording and three-dimensional (3D)

kinematics when measuring ADF during gait. Though some evidence of use of video recording to examine ROM exists, no estimates of intra-rater, inter-rater, or test-retest reliability of 2D video recording to assess ADF using standard ROM examination techniques were identified.

Based on the conflicting evidence on reliability of passive ROM assessment for children with CP, this study was designed to address the need for evidence of the reliability of ROM measurements acquired through post hoc measurement of 2D video recordings. The aims of this study were to examine ADF ROM measurements in children with CP, specifically to determine: 1) Concurrent validity, comparing ROM gathered in real time to that assessed via video recording 2) intra-rater reliability of goniometric assessment via video recording 3) inter-rater reliability of goniometric assessment via video recording 4) test-retest reliability of goniometric assessment via video recording and 5) if use of 2D recording to measure ADF ROM may reduce the Standard Error of Measure (SEM), or Minimal Detectable Difference (MDD).

## **Methods**

### ***Design***

An observational repeated measure study (Portney & Watkins, 2009) was carried out in which concurrent validity and intra-rater, inter-rater, and test-retest reliability were calculated using videotaped ROM assessment from two different testing sessions. This study was reviewed and approved through the University of Washington (UW) institutional review board.

### ***Sample***

A convenience sample of 26 children, 17 children with typical development (8 males) ranging from 8 years, 5 months to 14 years, 11 months (Mean 10.31, SD 2.10 years) and 9 children with CP (8 males), aged 8 years, 5 months to 18 years, 2 months (Mean 13.08, SD 3.54

years), participated in this study. Participants were recruited through announcements and flyers to physical therapist's in the area and word-of-mouth from both previous participants and therapists in the area. Further description of children can be found in Table 1.

Table 1. Demographic and descriptive information for participants with cerebral palsy and demographic information for typically developing children.

	Child	Gender	Age (Yrs:Mos)	GMFCS Level	Distribution
Cerebral Palsy	1	Male	12:1	I	Unilateral (Right arm/leg)
	2	Male	11:5	II	Bilateral (both legs)
	3	Male	9:5	I	Unilateral (Left arm/leg)
	4	Female	8:5	IV	All four limbs
	5	Male	10:6	II	Bilateral (both legs)
	6	Male	18:2	II	Both legs and right arm
	7	Male	15:10	II	Both legs and left arm
	8	Male	14:5	II	Bilateral (both legs)
	9	Male	17:6	II	Bilateral (both legs)
Typically Developing	10	Male	9:4		
	11	Male	14:0		
	12	Male	8:3		
	13	Female	10:11		
	14	Male	8:0		
	15	Female	10:2		
	16	Female	8:9		
	17	Female	14:11		
	18	Male	13:0		
	19	Male	11:0		
	20	Male	8:6		
	21	Female	8:11		
	22	Female	12:3		
	23	Female	9:1		
	24	Female	9:0		
	25	Male	10:4		
	26	Female	8:10		

### ***Instrumentation***

Data were collected in the UW Human Motion Analysis Laboratory or at participants' homes. Washable ink markings were applied to each participant to indicate the position of the head of the fibula, lateral malleolus, base and head of the 5<sup>th</sup> metatarsal in order to align the

goniometer in accordance with other studies (Glanzman et al., 2008; Kilgour et al., 2003; Mutlu et al., 2007). A plastic goniometer with 8 in arms and 2 deg markings (McCoy Health Science Supply, Maryland Heights, MO) was used for all measurements (i.e., real-time and post hoc video) (Glanzman et al., 2008). Children were positioned supine on a treatment table with their knees extended and ankles over the edge of the table to allow unrestricted movement. A video camera was placed at a height even with the top of the treatment table, perpendicular to the participant's lower leg, and approximately 4 feet away. The Toshiba Camileo X100 camera (Toshiba Corporation, Minato, Tokyo, Japan) and frame were adjusted to include 1" proximal to the fibular head mark and 1" beyond the mark on the 5<sup>th</sup> metatarsal head when the child was in full plantarflexion. This framing was chosen to capture the applied movements with the greatest amount of detail (figure 1).

Figure 1. Video capture of ankle dorsiflexion at end range.



### ***Real-time goniometry***

Children attended two test sessions, planned at one week for most participants. Some children with CP were enrolled in another study and were tested at 3 week intervals. In each test

session, children were asked to relax and allow passive stretch of their ankle by the assessor. The assessor (TG) held the child's ankle at the end range position and ensured the movement was recorded by the video camera (figure 1). The camera was then blocked from viewing by the assessor and the real-time goniometric measurement was taken (figure 2). This process was repeated 5 times with each foot.

Figure 2. Blocking of camera during real-time assessment of ankle dorsiflexion.



### ***Video goniometry***

After each participant had completed both test sessions, the test and retest pairs were randomly coded to blind the assessors. Assessor 1 (TG) was the primary assessor and is a licensed therapist with more than 10 years of clinical experience. Assessor 2 (CW) and Assessor 3 (DH) were physical therapy students trained in goniometry. Assessors 2 and 3 were trained by Assessor 1 on the use of the markings to establish the lever arms. To reduce measurement bias between trials, assessors were instructed to move the goniometer arms at least 45 degrees after each measurement. Using universal goniometers identical to the one used during the clinical

assessment, each assessor measured ROM for all trials. The assessors repeated the video measurement procedure after at least 3 days (range 3-7 days) in a second session using at least 10 participants' video recordings (i.e., six from the typically developing group and four from the group with CP). A minimum of 3 days between measurements was mandated to ensure assessors did not remember measurements. During measurement, assessors were allowed to rewind and pause the video to give them time to measure the ROM at the instant just prior to the primary assessor blocking the screen to capture the real-time measurement.

### **Data Analysis**

All data were analyzed using SPSS Version 19 (SPSS, Inc., Chicago, IL). Passive ROM data were compared using individual trials and also using averages of the first two, three, and four trials, and all five trials. Right and left legs were compared separately. A technical error created the loss of data for three trials on one leg in the second session for one participant with CP. **Concurrent Validity** was computed using interclass correlation coefficient (ICC) model 1 comparing the real-time goniometry to each assessor's measurements from the video for each test. **Intra-rater** reliability was computed using ICC model 2 comparing one assessor's measurements for the first test to same assessor's measurements for the measurements taken for the second time. **Inter-rater** reliability was computed using ICC model 2 comparing one assessor's measurements to another assessor's measurements for the same test. **Test-retest** reliability was computed using ICC model 1 comparing each participant's measurements for the first session to the measurements for the second session.

## Results

Ankle dorsiflexion varied between the two populations. The typically developing children had mean ADF of 10.47 (SD 8.83) and 11.00 (SD 9.66) whereas the children had mean ADF of 0.44 (SD 11.35) and 4.89 (SD 13.43) on the left and right, respectively.

### Concurrent Validity

The overall results for individual trials for typically developing children ranged from ICC (1,1) 0.89 – 0.98 and trial averages ranged from ICC (1,2-5) 0.95 – 0.99. Concurrent validity for the children with CP ranged from ICC (1,1) 0.89 – 0.98 for individual trials and ICC (1,2-5) 0.97 – 0.99 for trial averages (Table 2).

Table 2. Concurrent Validity. Table shows data from assessor 1 (PT) comparing real-time measurement to video rating including confidence interval (C.I.).

Test/ Leg	Comparison	Typically developing children				Children with CP			
		N	ICC	95% C.I.		N	ICC	95% C.I.	
				Low	High			Low	High
First/ Left	Average of all 5 trials	17	0.981	0.949	0.993	9	0.968	0.868	0.993
	Average of trials 1-4		0.982	0.952	0.994		0.968	0.868	0.993
	Average of trials 1-3		0.979	0.944	0.992		0.966	0.862	0.992
	Average of trials 1-2		0.977	0.937	0.992		0.969	0.873	0.993
	Individual Trials		0.935- 0.971	0.832- 0.923	0.976- 0.989		0.966- 0.969	0.862- 0.873	0.992- 0.993
Retest/ Left	Average of 5 trials	17	0.963	0.900	0.986	9	0.966	0.861	0.992
	Average of trials 1-4		0.966	0.909	0.988		0.969	0.873	0.993
	Average of trials 1-3		0.958	0.888	0.985		0.965	0.855	0.992
	Average of trials 1-2		0.969	0.915	0.989		0.970	0.875	0.993
	Individual Trials		0.945- 0.982	0.857- 0.953	0.979- 0.994		0.892- 0.951	0.620- 0.812	0.974- 0.988
First/ Right	Average of 5 trials	17	0.986	0.962	0.995	9	0.988	0.953	0.997
	Average of trials 1-4		0.989	0.969	0.996		0.987	0.948	0.997
	Average of trials 1-3		0.985	0.960	0.995		0.989	0.955	0.997
	Average of trials 1-2		0.988	0.967	0.995		0.990	0.957	0.998
	Individual Trials		0.948- 0.972	0.888- 0.939	0.976- 0.987		0.961- 0.980	0.848- 0.920	0.991- 0.995
Retest/ Right	Average of 5 trials	17	0.956	0.881	0.984	9	0.987	0.946	0.997
	Average of trials 1-4		0.953	0.872	0.983		0.984	0.936	0.996
	Average of trials 1-3		0.953	0.873	0.983		0.983	0.930	0.996
	Average of trials 1-2		0.951	0.868	0.982		0.985	0.939	0.997
	Individual Trials		0.889- 0.921	0.726- 0.889	0.958- 0.985		* 0.951- 0.976	0.796- 0.895	0.990- 0.995

Note that all ICC values were found to have a p-value  $\leq 0.001$ .

\*N for retest on right leg was 9 for trials 1 and 2 and 8 for trials 3-5.

### ***Intra-rater Reliability***

The intra-rater reliability for the video-based measurements varied from ICC (2,1) 0.92 – 0.99 for the different assessors on individual trials and ICC (2,2-5) 0.99 – 1.00 on averages for multiple trials within the group of typically developing children. The 95% confidence intervals were also very high for the typically developing children (0.60 – 1.00 for individual trials and 0.92 – 1.00 for averages). ICC (2,1) values of 0.97 – 1.00 and ICC (2,2-5) 0.99 – 1.00 for individual trials and averages, respectively, were obtained for children with CP. The confidence intervals (95%) were wider (0.34 for an individual trial to 1.00 for both individual trials and averages) for children with CP. Table 3 provides details on intra-rater reliability.

Table 3. Intra-rater Reliability. Table shows data from all three raters for intra-rater comparisons of the average of all 5 trials, the first 3 trials, and a single randomly selected trial.

			Typically developing children				Children with CP			
			N	ICC	95% Confidence Interval		N	ICC	95% Confidence Interval	
Leg	Rater	Comparison			Low	High			Low	High
Left	PT	Average of 5	6	0.999	0.996	1.000	4	1.000	0.998	1.000
		Average of 1 <sup>st</sup> 3		0.998	0.990	1.000		1.000	0.997	1.000
		Individual Trial		0.992	0.953	0.999		0.999	0.982	1.000
	SPT1	Average of 5		1.000	0.996	1.000		1.000	0.999	1.000
		Average of 1 <sup>st</sup> 3		1.000	0.995	1.000		1.000	0.998	1.000
		Individual Trial		0.999	0.990	1.000		0.999	0.992	1.000
	SPT1	Average of 5		1.000	0.996	1.000		1.000	0.997	1.000
		Average of 1 <sup>st</sup> 3		0.999	0.993	1.000		0.999	0.989	1.000
		Individual Trial		0.997	0.983	1.000		0.997	0.971	1.000
Right	PT	Average of 5	6	0.999	0.992	1.000	4	1.000	0.999	1.000
		Average of 1 <sup>st</sup> 3		0.996	0.977	0.990		1.000	0.976	1.000
		Individual Trial		0.940	0.637	0.991		0.994	0.908	1.000
	SPT1	Average of 5		0.993	0.952	0.999		0.997	0.949	1.000
		Average of 1 <sup>st</sup> 3		0.989	0.924	0.998		0.996	0.949	1.000
		Individual Trial		0.921	0.604	0.988		0.991	0.887	0.999
	SPT1	Average of 5		0.997	0.981	1.000		0.994	0.731	1.000
		Average of 1 <sup>st</sup> 3		0.996	0.970	0.999		0.991	0.293	0.999
		Individual Trial		0.987	0.923	0.998		0.972	0.337	0.998

**Inter-rater Reliability**

Inter-rater reliability for the video-based measurements for typically developing children varied from ICC (2,1) 0.98 – 0.99 for individual trials and ICC (2,2-5) 0.99 – 1.00 for trial averages. For single trials of typically developing children, 95% confidence intervals ranged from 0.73 – 0.99 with separate individual raters compared to one another and 0.98 – 1.00 when comparing all raters together. When using averages of trials, the ICC (2,2-5) ranged from 0.99 – 1.00 with 95% confidence intervals ranging from 0.97 – 1.00. For children with CP, inter-rater reliability varied for individual trials from ICC (2,1) 0.96 – 1.00 and averages of trials from ICC (2,2-5) 0.97 – 1.00. The 95% confidence intervals associated with these values were 0.67 – 1.00 and 0.88 – 1.00 for individual trials and averages of trials, respectively. Results of inter-rater reliability can be found in Table 4.

Table 4. Interrater Reliability. Table shows data from comparisons of physical therapist (PT) and students of physical therapy (SPT) interrater comparisons.

Test/ Leg	Raters	Comparison	Typically developing children				Children with CP			
			N	ICC	95% Confidence Interval		N	ICC	95% Confidence Interval	
					Low	High			Low	High
First/ Left	PT/ SPT1	Average of 5 trials	17	0.991	0.970	0.997	9	0.995	0.980	0.999
		Average of trials 1-4		0.993	0.825	0.999		0.995	0.978	0.999
		Average of trials 1-3		0.993	0.843	0.998		0.995	0.977	0.999
		Average of trials 1-2		0.993	0.856	0.998		0.996	0.984	0.999
		Individual Trials		0.983- 0.989	0.728- 0.917	0.996- 0.997		0.983- 0.994	0.927- 0.973	0.996- 0.999
	PT/ SPT2	Average of 5 trials		0.992	0.978	0.997		0.997	0.986	0.999
		Average of trials 1-4		0.995	0.983	0.998		0.996	0.984	0.999
		Average of trials 1-3		0.995	0.981	0.998		0.996	0.985	0.999
		Average of trials 1-2		0.995	0.980	0.998		0.996	0.985	0.999
		Individual Trials		0.987- 0.990	0.951- 0.969	0.996- 0.997		0.989- 0.993	0.958- 0.973	0.998- 0.999
	SPT1/ SPT2	Average of 5 trials		0.998	0.981	0.999		0.996	0.972	0.999
		Average of trials 1-4		0.997	0.984	0.999		0.996	0.982	0.999
		Average of trials 1-3		0.997	0.987	0.999		0.996	0.984	0.999
		Average of trials 1-2		0.997	0.988	0.999		0.997	0.983	0.999
		Individual Trials		0.984- 0.993	0.958- 0.978	0.994- 0.998		0.982- 0.993	0.665- 0.970	0.997- 0.998

Table 4. Interrater Reliability continued

Test/ Leg	Raters	Comparison	N	ICC	Low	High	N	ICC	Low	High
Retest/ Left	PT/ SPT1	Average of trials 1-3	17	0.995	0.975	0.998	9	0.995	0.979	0.999
		Individual Trial		0.990	0.948	0.997		0.986	0.942	0.997
	PT/ SPT2	Average of trials 1-3		0.995	0.986	0.998		0.973	0.877	0.994
		Individual Trial		0.990	0.973	0.996		0.940	0.758	0.986
	SPT1/ SPT2	Average of trials 1-3		0.997	0.985	0.999		0.985	0.933	0.997
		Individual Trial		0.993	0.969	0.998		0.970	0.875	0.993
	All	Average of 5 trials		0.998	0.994	0.999		0.990	0.968	0.997
		Average of trials 1-4		0.997	0.993	0.999		0.990	0.967	0.997
		Average of trials 1-3		0.997	0.993	0.999		0.990	0.968	0.997
		Average of trials 1-2		0.997	0.993	0.999		0.987	0.961	0.997
		Individual Trials		0.989- 0.993	0.971- 0.983	0.996- 0.997		0.957- 0.977	0.873- 0.931	0.989- 0.994
	First/ Right	PT/ SPT1		Average of trials 1-3	17	0.998		0.994	0.999	9
Individual Trial			0.992	0.979		0.997	0.994	0.970	0.999	
PT/ SPT2		Average of trials 1-3	0.996	0.988		0.999	0.996	0.913	0.999	
		Individual Trial	0.989	0.780		0.997	0.986	0.939	0.997	
SPT1/ SPT2		Average of trials 1-3	0.997	0.915		0.999	0.999	0.985	1.000	
		Individual Trial	0.982	0.658		0.996	0.996	0.985	0.999	
All		Average of 5 trials	0.998	0.996		0.999	0.998	0.992	1.000	
		Average of trials 1-4	0.998	0.995		0.999	0.998	0.993	1.000	
		Average of trials 1-3	0.998	0.994		0.999	0.998	0.992	1.000	
		Average of trials 1-2	0.998	0.990		0.999	0.998	0.992	1.000	
		Individual Trials	0.988- 0.994	0.946- 0.987		0.996- 0.998	0.990- 0.993	0.964- 0.979	0.998	
Retest/ Right		PT/ SPT1	Average of trials 1-3	17		0.998	0.994	0.999	9	
	Individual Trial		0.971		0.923	0.989	8	0.997	0.984	0.999
	PT/ SPT2	Average of trials 1-3	0.995		0.985	0.998	9	0.995	0.974	0.999
		Individual Trial	0.991		0.971	0.997	8	0.994	0.973	0.999
	SPT1/ SPT2	Average of trials 1-3	0.995		0.986	0.998	9	0.997	0.989	0.999
		Individual Trial	0.972		0.921	0.990	8	0.997	0.988	0.999
	All	Average of 5 trials	0.997		0.994	0.999	9	0.997	0.992	0.999
		Average of trials 1-4	0.997		0.994	0.999		0.997	0.991	0.999
		Average of trials 1-3	0.997		0.994	0.999		0.997	0.990	0.999
		Average of trials 1-2	0.997		0.993	0.999		0.997	0.991	0.999
		Individual Trials	0.978- 0.991		0.952- 0.981	0.991- 0.997		*	0.986- 0.996	0.954- 0.987

Note that all ICC values were found to have a P value = < 0.001.

\*N for retest on right leg was 9 for trials 1 and 2 and 8 for trials 3-5.

### ***Test-retest Reliability***

Tests were mean 12.5 days apart (SD 7.32). Examination of real-time measurements for the typically developing children showed ICCs (1,1) 0.83 – 0.92 for any given individual trial and ICCs (1, 2-5) 0.91 – 0.96 for averages of at least two trials or more. The ICC (1,1) for any individual trial with any of the video measurements had a range of 0.83 – 0.94 and 0.94 – 0.98

for the averages using ICC (1,2-5). Test-retest reliability estimates for children with CP ranged from ICC (1,1) 0.71 – 0.96 and 0.89 – 0.97 for individual trial and averages (using ICC [1,2-5]) with real-time measurements, respectively. Video measurement ICC (1,1) values for children with CP on individual trials ranged from 0.76 – 0.97 and (1,2-5) 0.88 – 0.96 for averages. Test-retest data is presented in Table 5.

The SEM ( $SEM = SD \times \sqrt{1 - ICC}$ ) (Portney & Watkins, 2009) was computed using the video measurements and taking the highest SD (to allow the greatest error) for individual trials and the average of the first 2 trials of each foot. These values were used to compute MDD values ( $MDD = 1.96 \times SEM \times \sqrt{2}$ ) (Portney & Watkins, 2009) for each corresponding SEM value. Table 6 presents SEM and MDD values for both populations.

## **Discussion**

The aims of this study were to determine concurrent validity, as well as test-retest, intra- and inter-rater reliability of using video recordings to assess ADF ROM and determine SEM and MDD in children. Results of this study are interpreted with the recommendation that reliability coefficients of between 0.75 and 1.00 represent a good to excellent relationship between the studied measurements (Portney & Watkins, 2009). The concurrent validity showed excellent relationships when interpreting the ICC values for both the typically developing children and the sample of children with CP. With the lowest ICC value of either test with either foot being 0.95 for any average within typically developing children and 0.89 for any single trial, this study shows that using this method is a valid way to determine ankle DF ROM in the typically developing population. For children with CP, the ICCs are slightly lower, but still show excellent correlations and high reliability (0.94 for any average and 0.89 for any single trial as the lowest ICCs). Nearly every ICC value reported had a 95% confidence interval with the

Table 5. Test Retest Reliability. Table shows data from measured laboratory values and all raters.

Leg	Rating	Comparison	Typically developing children			Children with CP				
			N	ICC	95% Confidence Interval		N	ICC	95% Confidence Interval	
					Low	High			Low	High
Left	Lab	Average of 5 trials	17	0.928	0.807	0.974	9	0.890	0.548	0.975
		Average of trials 1-4		0.922	0.789	0.971		0.907	0.619	0.979
		Average of trials 1-3		0.917	0.775	0.970		0.894	0.564	0.976
		Average of trials 1-2		0.909	0.755	0.967		0.893	0.560	0.975
		Individual Trials		0.841-0.879	0.623-0.704	0.939-0.954		0.715-0.846	0.190-0.491	0.927-0.963
	PT	Average of 5 trials		0.946	0.856	0.980		0.944	0.770	0.987
		Average of trials 1-4		0.955	0.878	0.984		0.946	0.778	0.988
		Average of trials 1-3		0.954	0.876	0.983		0.937	0.743	0.986
		Average of trials 1-2		0.947	0.858	0.981		0.919	0.668	0.981
		Individual Trials		0.850-0.910	0.641-0.769	0.942-0.966		0.803-0.906	0.381-0.663	0.951-0.978
	SPT1	Average of 5 trials		0.955	0.878	0.984		0.933	0.726	0.985
		Average of trials 1-4		0.955	0.878	0.983		0.937	0.740	0.985
		Average of trials 1-3		0.955	0.877	0.983		0.929	0.709	0.984
		Average of trials 1-2		0.944	0.848	0.979		0.907	0.620	0.979
		Individual Trials		0.841-0.893	0.621-0.735	0.939-0.959		0.799-0.902	0.372-0.652	0.950-0.977
	SPT2	Average of 5 trials		0.952	0.870	0.982		0.913	0.642	0.980
		Average of trials 1-4		0.950	0.865	0.982		0.911	0.634	0.980
		Average of trials 1-3		0.948	0.861	0.981		0.905	0.609	0.978
		Average of trials 1-2		0.944	0.848	0.979		0.881	0.512	0.973
		Individual Trials		0.860-0.900	0.662-0.752	0.946-0.962		0.760-0.868	0.283-0.552	0.939-0.968
Right	Lab	Average of 5 trials	17	0.957	0.884	0.984	9	0.965	0.855	0.992
		Average of trials 1-4		0.959	0.801	0.971		0.959	0.830	0.990
		Average of trials 1-3		0.955	0.877	0.983		0.955	0.816	0.990
		Average of trials 1-2		0.950	0.865	0.982		0.956	0.821	0.990
		Individual Trials		0.828-0.921	0.594-0.800	0.933-0.970		* 0.875-0.958	0.570-0.840	0.970-0.990
	PT	Average of 5 trials		0.974	0.929	0.990	9	0.962	0.844	0.991
		Average of trials 1-4		0.974	0.930	0.991		0.957	0.822	0.990
		Average of trials 1-3		0.972	0.925	0.990		0.951	0.799	0.989
		Average of trials 1-2		0.972	0.924	0.990		0.950	0.796	0.989
		Individual Trials		0.915-0.938	0.787-0.842	0.968-0.977		* 0.883-0.960	0.595-0.829	0.972-0.992
	SPT1	Average of 5 trials		0.971	0.921	0.989	9	0.954	0.813	0.990
		Average of trials 1-4		0.971	0.922	0.990		0.947	0.784	0.988
		Average of trials 1-3		0.969	0.917	0.989		0.942	0.763	0.987
		Average of trials 1-2		0.968	0.913	0.988		0.942	0.764	0.987
		Individual Trials		0.907-0.934	0.767-0.831	0.965-0.975		* 0.868-0.953	0.533-0.804	0.968-0.990
	SPT2	Average of 5 trials		0.976	0.935	0.991	9	0.965	0.856	0.992
		Average of trials 1-4		0.978	0.940	0.992		0.959	0.833	0.991
		Average of trials 1-3		0.976	0.934	0.991		0.954	0.812	0.989
		Average of trials 1-2		0.976	0.935	0.991		0.960	0.837	0.991
		Individual Trials		0.902-0.942	0.757-0.852	0.963-0.979		* 0.880-0.971	0.597-0.876	0.972-0.994

All ICC values,  $p \leq 0.001$ . \*N for retest on right leg was 9 for trials 1 and 2 and 8 for trials 3-5.

Table 6. Standard of Measurement (SEM) and Minimal Detectable Difference (MDD) values. The higher SD of the two test sessions was used to reflect the greatest error. Values shown are the lowest and highest values among the three raters for individual trials and for the average of the first two trials.

		Children with Typical Development				Children with CP			
		SEM		MDD		SEM		MDD	
		Lowest	Highest	Lowest	Highest	Lowest	Highest	Lowest	Highest
Individual Trials	Right	2.16	4.66	5.98	12.91	2.16	4.66	5.98	12.91
	Left	3.20	5.07	8.68	12.43	3.20	5.07	8.88	14.06
Average of 2 Trials	Right	2.50	3.04	6.92	8.43	2.18	3.04	6.05	8.43
	Left	2.99	3.04	8.30	8.68	2.54	3.44	7.05	9.54

lower boundary above 0.75, with the lowest single trial ICC being 0.54. These high correlations lead to excellent confidence with the validity of this method. The values in the current study were similar to, and in some cases higher than, the study by Allington *et al.*(2002) where ICCs of 0.965 – 0.977 were reported by measurements obtained via visual estimation.

As with other reported results, intra-rater reliability using post hoc measurement from video recording was high. The SPT raters showed slightly lower ICC values which may indicate that experience is important for improving intra-rater reliability. However, the differences between PT and SPT raters were negligible as all were near 0.99. The reliability estimates obtained in this study were higher than those reported in prior studies, such as 0.973 reported by Glanzman *et al.*(2008) and 0.932 – 0.53 reported by Allington *et al.*(2002) The wider confidence intervals found with intra-rater reliability in the current study were most likely the result of the small sample size used (i.e. 6 TD, 4 CP).

Inter-rater reliability was extremely high using post hoc video measurement. Inter-rater ICCs show that post hoc video measurement has a higher reliability than other methods. Post hoc video measurement has greater inter-rater reliability for children with CP than previously reported inter-rater ICCs (0.872 and 0.900 – 0.954 reported by McWhirk & Glanzman (2006) and Allington *et al.* (2002), respectively).

The test-retest reliability in the current study was also excellent. Children with CP had lower overall test-retest reliability and a wider range of ICCs. Findings were still consistent, or higher than other estimates of test-retest reliability, such as the 0.77 – 0.88 (ICC model 2) reported by Mutlu *et al.*(2007) and the 0.63 – 0.75 reported by Kilgour *et al.*(2003).

Values found for SEM and MDD were only slightly higher with typically developing children than those with CP. None of the other studies examining ADF ROM specifically reported SEM. Thus, the results from this study suggest that ADF in children with CP should include a five degree bandwidth on either side for 68% confidence that the true ROM value is within the measured value. The minimal difference in MDD values between TD children and those with CP challenges the hypothesis that spasticity and hypertonicity may play a role in the error of ROM assessment in children with CP. One possible explanation that altered muscle tone in children with CP had less of an influence on ADF ROM in the current study is that averaging multiple trials may mitigate the role of spasticity during ADF assessment. The MDD of less than 10 degrees for children with CP using an average of at least two trials suggests that a measured change of 10 degrees or greater is likely to be reflective of true change. Single trial MDD values of over 10° suggest that measured values of ADF ROM would need to be greater than 10° to be confident of true change if only using one measurement. MDD values found were similar to the high end of the mean measurement error of 0-10 degrees reported in the study by Allington *et al.* (2002).

Results suggest that for greater reliability, an average of multiple trials should be used. In the current study, using an average of three trials showed minimally greater reliability than two trials, but the reliability of using four or five trial averages was not appreciably greater than the ICC using a two- or three-trial averages. This leads to the recommendation of using the

average of three trials, as the three-trial average yielded the most consistently high ICC values. This three-trial average lends itself to providing much higher reliability than single measurements but that taking the additional time to perform additional measurements may not significantly increase the precision of the measurement.

Though we advocate that the standardized methods used in this study have advantages, there were also a few limitations to this study. First, a relatively small number of children with CP participated in the study. A technical issue caused a loss of data to one child with CP which slightly reduced the amount of data for children with CP included in the analysis. There were few children with CP included in the intra-rater reliability. Due to testing strategies, children with CP typically had a longer time between tests which may have affected the test-retest reliability results compared to the children with typical development. Children with unilateral CP were also included in the group for all the children with CP regardless if the leg used was affected or not. This allowed some ankles reflecting more “typical development” into the data on children who had CP, but was conducted to avoid artificial inflation of the N for the typically developing population.

Other than higher reliability as discussed above, the benefits to post hoc measurement of video recorded ROM also include that video recording could be very valuable in research for blinding of raters to whether they are measuring pre- or post-test ROM. Clinically, this method would create a digital log of progress for children. With prior set-up, this method can also limit the time needed perform direct goniometry with children and possibly help to maintain their engagement in therapy sessions. Video recording makes it much more practical for a single rater to be responsible for measuring ROM, even in cases where multiple therapists may be working with, or assessing the same child. Use of video recording also makes it possible for the assessor

to use two hands to make positioning in correct biomechanical alignment easier, as maintaining neutrality in the subtalar joint is important for ADF (Tiberio, 1987). This type of measurement was determined to be feasible in the clinical setting with only a few minutes of set-up time required and less than a minute, if the treatment table and camera were already in place and allows a single person to perform the rating with comparable to better reliability, depending on the type of reliability reported. In contrast, the Allington *et al.* (2002) study used a two-person approach where one person held the limb and another person took the measurement. The Allington *et al.* (2002) study may suggest greater time-savings using visual estimation rather than goniometry to determine ROM, but the authors of that study also acknowledged the need for direct measurements.

## **Conclusions**

The measurements from this study showed excellent intra- and inter-rater and test-retest reliability with use of post hoc measurement of standardized video recording for passive ADF ROM. Therefore, post hoc video measurements are a reliable way of obtaining ROM data for children with typical development and children with CP. Both inter- and intra-rater reliability was higher with post-hoc video measurement than similar studies have reported. Though inter-rater reliability was high using this method, intra-rater reliability was higher. The test-retest reliability seen with study was also high. Regardless of the application of post hoc video measurement, or standard goniometry of ADF ROM, the results of this study suggest that using an average of three trials generally provides balance between the highest inter-rater, intra-rater, and test-retest reliability with the fewest number of trials.

Future research is needed to address active ROM in children using the video recording method. This future research may lead to methods that will provide high reliability when assessing ROM during motion and activities such as gait.

## Chapter 4:

### NeuroGame Therapy for Ankle Dorsiflexion in Children with Cerebral Palsy

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#### Abstract:

**Background:** A NeuroGame Therapy (NGT) device using surface electromyography (sEMG) biofeedback to control commercially available video games was used to train activation of the tibialis anterior (TA) muscles, hypothesized to improve walking and postural stability in children with cerebral palsy (CP).

**Method:** Nine ambulatory children with spastic diplegic or quadriplegic CP (3 males), 8-16 years old (mean 12.15, SD=3.36) participated in NGT for six weeks. Five assessments of body structure/function domain variables (sEMG, gait, balance, muscle contraction force, and selective muscle activation) and activity domain variables (6-minute walk test, reports of falls) occurred three weeks apart, with two prior, one mid, and two post NGT.

**Results:** Individual analysis showed that each child improved in at least one measure within the body structure/function domain and three children showed slight improvement in the activity domain.

**Conclusion:** NGT was feasible for use in the home in training the TA muscles to dorsiflex the ankles.

## **Introduction**

Cerebral palsy (CP) describes a group of movement disorders resulting from non-progressive injury to the fetal or infant brain (Rosenbaum et al., 2007). CP affects 4-5 per 1,000 children, over 80% have spasticity as the primary presentation, and over 59% of children with CP have a distribution that affects both lower extremities (Aisen et al., 2011; Durkin et al., 2015). Of children with bilateral distribution, nearly 45% are able to walk independently and another 16% rely on an assistive device for primary ambulation (Kirby et al., 2011). The most commonly affected joint is the ankle (Bland, Prosser, Bellini, Alter, & Damiano, 2011). Foot drop or toe drag are common characteristics of the gait patterns of these children (J. R. Davids, Foti, Dabelstein, & Bagley, 1999). As children with CP age, they tend to have a decrease in the amount of ankle dorsiflexion (DF) (Bell, Ounpuu, DeLuca, & Romness, 2002). Due to the abnormalities seen in gait, physical therapy intervention commonly addresses gait training in children with CP (Franki et al., 2012). One such intervention is targeting selective voluntary movement control (SVMC) as a decrease in SVMC has been linked to deficiencies in gait (Fowler & Goldberg, 2009).

We hypothesized that improving selective motor control is a key principle in rehabilitation, which will assist with the ultimate goal of improving participation within the child's home, school, and community. Neural plasticity studies suggest that permanent improvements with motor control can occur with sustained specific and relevant practice (Johnston, 2009; Kleim & Jones, 2008). Direct practice contributes to neuroplasticity in children with cerebral palsy (Sterling et al., 2013; Uswatte & Taub, 2013). There is some evidence that supports plasticity affecting ankle control after treadmill training (Phillips et al., 2007; Willerslev-Olsen, Petersen, Farmer, & Nielsen, 2015) and with use of electrical stimulation to the tibialis anterior (TA) muscle (Damiano, Prosser, Curatalo, & Alter, 2013; Mäenpää,

Jaakkola, Sandström, & von Wendt, 2004). Changes likely result from repetitive motor control practice of activation of the intended motor unit. This “just-right” practice, using the appropriate activity with the right challenge, and the appropriate amount of feedback is the foundation of motor learning.

Practice of selective motor control activity is paramount for the establishment and reinforcement of neural pathways (Flück, 2006) and continued activity is required to maintain these pathways (Damiano, 2006). The amount of practice required to learn and maintain a motor skill may not be possible within the therapy session time a child receives. Given that current clinical therapy regimens that are covered by insurance (1-3 times per week for 45-60 minutes) do not provide enough opportunities for repetitive practice to affect maximum recovery, therapists attempt to add movement practice by recommending home programs. Adherence to home exercise programs that include enough repetition is poor (Chappell & Williams, 2002; Rone-Adams, Stern, & Walker, 2004). One way to enhance the adherence to home therapy programs may be via use of virtual reality (VR) systems (Bryanton et al., 2006). VR is a technology that utilizes computer or other monitor systems to allow participants to use their own movements to control an avatar in two- or three-dimensional environments.

One type of VR application is through the use of surface electromyography (sEMG) biofeedback. Surface electromyography (EMG) has been used with some success to provide information about muscle recruitment to children with CP (Bloom, Przekop, & Sanger, 2010). EMG biofeedback has been used successfully to improve ankle range of motion and gait in children with hemiplegic CP (Binder et al., 1981; Toner et al., 1998). Previous applications of EMG biofeedback used muscle activation to move a bar, generate or maintain a graphic of some type on a monitor (O’Dwyer, Neilson, & Nash, 1994). One drawback to such feedback is that it

offers a less stimulating and interactive environment compared to commercial video games that are commonly enjoyed by children (Nippold, Duthie, & Larsen, 2005). The use of VR has become very popular in rehabilitation as it may influence neuroplastic changes in the cerebral cortex and its use has resulted in improved motor skills in children with CP (Inguaggiato, Sgandurra, Perazza, Guzzetta, & Cioni, 2013; You et al., 2005). These changes may be the result of the opportunities for repetitive sensory practice that VR provides in children with CP (Weiss, Tirosh, & Fehlings, 2014). One potential downfall to VR is that all current systems require active movement to control the avatar. Also, motor skills learned using VR technology may not transfer to other environments (de Mello Monteiro et al., 2014). Thus, training using VR should also focus on the transfer of the activity to the intended environment.

Due to advances in technology, we are able to combine the utility of biofeedback while providing a fun virtual reality experience that is easily used in the home without the need for constant direct supervision by a therapist. Based on the proposed mechanism of improved motor control through neuroplastic changes via focused, salient repetition, a NeuroGame Therapy (NGT) system has been developed. The NGT system is designed to be used as a supplement to, and in conjunction with traditional therapy services. This system promotes salient and motivating practice in individuals with neuromotor dysfunction utilizing sEMG to control popular, commercially available video games, thus providing biofeedback to improve selective motor control. Improvements in upper extremity selective motor control through the use of NGT, have been documented in children with CP (Rios et al., 2013) and adults after stroke and traumatic brain injury (Donoso Brown et al., 2014). In both the Rios et al. and Donoso Brown et al. studies, improvements were seen in individual muscle activity (i.e., as the agonist muscle was activated, there was a decrease in activity from the antagonist muscles) during game play and

some of the participants also demonstrated improved amplitude of sEMG activation during laboratory tests. Neither study reported conclusive improvements in clinical activity outcome measures, with authors citing a lack of sensitivity to change with their chosen measures (i.e., the Shriner's Hospital Upper Extremity Evaluation for the children with CP (Davids et al., 2006), Wolf Motor Function Test (Wolf et al., 2001) and Chedoke Arm and Hand Activity Inventory – 9 (Barreca et al., 2004) for the adults post stroke).

This study expands the current body of knowledge on NGT to address ankle function in children with CP via a home-based motivating VR experience. Additionally, we aimed to bridge the gap between learning a new muscle activation pattern and transferring that skill into the intended environment, thus we paired NGT with a short walking task immediately following game play. Our purpose was to evaluate the feasibility of this home program and the preliminary effect of NGT paired with subsequent walking practice for improving TA selective muscle control and muscle function within gait activity in children with bilateral spastic CP. We hypothesized that NGT would be feasible and that NGT paired with walking would improve active dorsiflexion during the swing phase of gait and thus improve gait speed and reduce falls.

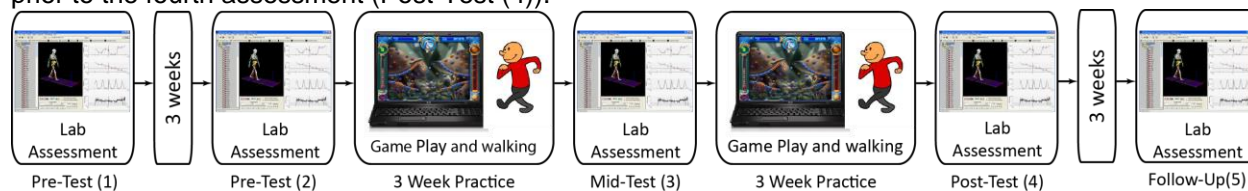
## **Methods**

### ***Study Design***

This study used a repeated-measures, case series design (Figure 1). A total of five assessments were scheduled approximately three weeks apart. This included two pre-tests, a six-week NGT intervention phase with one test in the middle of the intervention, an immediate post-test, and a follow-up test. This design was selected because of the heterogeneous nature of children with CP, in order to compare each child to him/her-self, and the preliminary nature of this investigation. All procedures were approved by the University Institutional Review Board.

Participants' parents gave written informed consent, and participants gave written informed assent prior to participation in the study.

Figure 1. Study design. Study consisted of 5 assessments occurring about three weeks apart. A six-week intervention immediately followed the second assessment (Pre-test (2)) and ceased immediately prior to the fourth assessment (Post-Test (4)).



### ***Participants***

Participants were a convenience sample of ambulatory children with a diagnosis of CP who had both lower extremities affected with an original primary presentation of spasticity, Gross Motor Function Classification Scales (GMFCS) levels I-III (Palisano et al., 1997; Palisano et al., 2008), and mean age of 12.15 years (SD = 3.36 years, range 8 years to 17 years). Recruitment of children was performed via correspondence with pediatric therapists in the local metropolitan area and through a mailing to parents through the local children's hospital. Participants had difficulty clearing their toes during walking, vision and hearing sufficient to play a computer game, and were cognitively able to give informed assent and participate in both research and intervention protocols. Participants were excluded if they had: (1) existence of any behavioral disorder, such as Attention Deficit Disorder (ADD) or Attention Deficit Hyperactivity Disorder (ADHD), or diagnosis of Pervasive Developmental Disorder (Autistic-spectrum or Asperger's Syndrome), (2) any skin condition that precluded adherence of electrodes, (3) flexion contracture at the knee beyond 20 degrees or plantarflexion contracture beyond 10 degrees at the ankle, (4) received neurolytic injections in the previous six months, (5) surgery to the lower extremity within one year, or (6) a history of a grand mal seizure within six months, or a known seizure disorder triggered by video displays.

Twelve participants were enrolled and nine completed all five assessments and the intervention. One participant withdrew due to persistent shunt failure and headaches unrelated to the study. One was not able to participate in the final three assessments due to family emergencies and scheduling conflicts, and another was deemed ineligible following the first assessment due to lack of discernable impairment. The characteristics of the nine participants (with pseudonyms for anonymity) who completed the study are presented in Table 1.

### **Intervention**

The NGT system consists of a laptop computer, NGT console, sEMG leads, and disposable electrodes. The NGT system console uses a custom neurochip circuit (Zanos, Richardson, Shupe, Miles, & Fetz, 2011) to amplify and digitize analog sEMG signals from two muscles and transmit these signals via Universal Serial Bus (USB) to the computer in real-time. Adjusted software controls convert muscle activity into movements used to control the game. The sensitivity of the system is adjustable to detect very low levels of activation, thus allowing persons with minimal muscle activation to participate. The required amount of sEMG activity to produce game movement was adjusted as needed during the intervention phase to facilitate challenging but successful game play (i.e. the 'just-right' challenge). The game system was programmed to automatically adjust the game settings based on sEMG amplitude during maximum voluntary contractions (MVCs) performed prior to each session, so while remote adjustments were possible, they were not used. The investigator could monitor game play adherence, if participants had an internet connection at home.

Participants used NGT in a sitting position and were asked to play with knees extended and resting on a stool to mimic the more straight leg posture seen in balance and walking activities. The children used the muscle activity in each of their tibialis anterior (TA)

Table 1. Demographic and characteristic information for NeuroGame Therapy participants.

Name <sup>1</sup>	Age <sup>2</sup> (yrs:mos)	Gender	Ethnicity	Co-Morbidities <sup>3</sup>	Activities <sup>4</sup>	GMFCS level	Assistive Devices	Gait Pattern	Spasticity <sup>5</sup>
Andy*	10:6	Male	Caucasian	Vision	PT, AHBR, VT, HEP, Sport	II	None reported	Equinous	0/0
Brandon*	13:6	Male	Caucasian	None reported	PT, HT, Sport	III	AFOs, Walker	Jump	6/8
Ethan	8:7	Male	Caucasian	Hearing	PT, HEP, Sport	I	None reported	Equinous	1/5
Francine	12:1	Female	Caucasian	Learning Disability	PT, Sport	III	Forearm Crutches, AFOs	Equinous	4/8
Dianne	11:8	Female	Caucasian	Learning Disability	S-B PT, OT, S-B OT, S-B SLP, HEP, Sport	I	None reported	Equinous	2/9
Helen	16:5	Female	Caucasian	Learning Disability	None reported	I	None reported	Equinous	3/5
Carly	8:5	Female	Caucasian	Vision	PT, S-B PT, OT, S-B OT, HT, H2O	III	AFOs, Walker	Jump	11/10
Irene	17:8	Female	Mixed	None reported	None reported	III	Walker, W/C	Jump	13/11
Gina	8:2	Female	Caucasian	Learning Disability	S-B PT, HEP	II	None reported	Equinous	3/3

<sup>1</sup>Names are pseudonyms used to protect anonymity.

<sup>2</sup>Age listed is age at time of first assessment.

<sup>3</sup>Co-Morbidities are based on parent report from health questionnaire.

<sup>4</sup>Activities are per parent report.

<sup>5</sup>Spasticity is measured using the Composite Spasticity Scale (16 point maximum per leg), reported as Right/Left score.

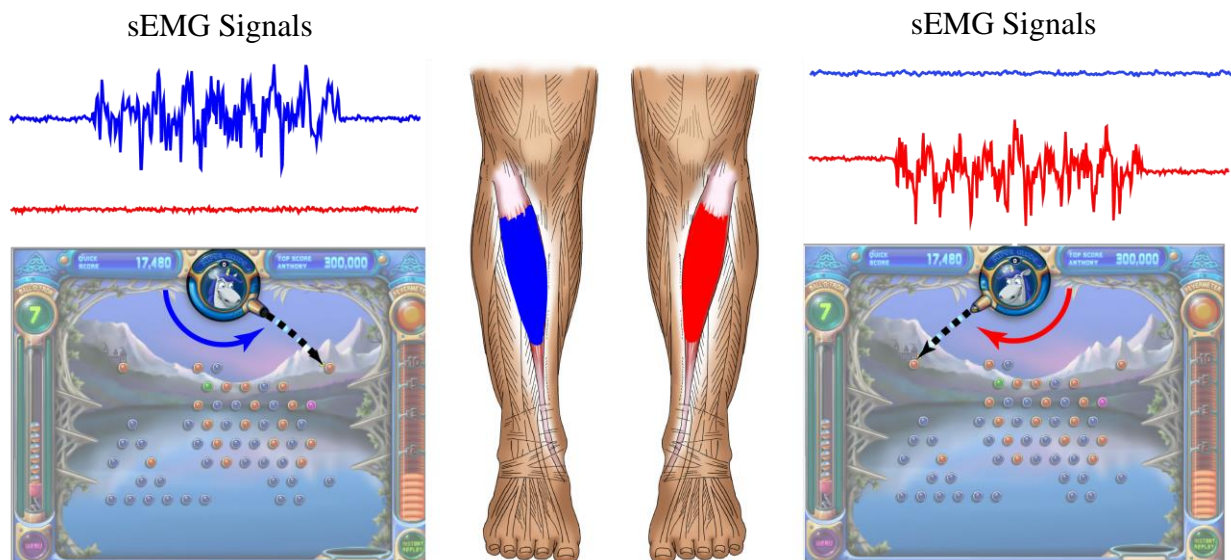
\*These children underwent selective dorsal rhizotomy more than a year prior to this study.

Abbreviations key: AFOs – Ankle foot orthosis, AHBR – adapted horseback riding, H2O – aquatic therapy, HEP – home exercise program, HT – hippotherapy, OT – private occupational therapy, PT – private physical therapy, S-B OT – school-based occupational therapy, Sport – any type of organized sporting activity (e.g., karate, swimming, bowling, baseball, soccer), S-B PT – school-based physical therapy, S-B SLP – school based speech language pathology, VT – Vision therapy, W/C – Manual Wheelchair

Gait patterns: Equinous: toe-walking gait with knee and hip extended during stance, Jump: toe-walking gait with knee and hip flexed during stance.

muscles to perform pre-game maximum voluntary contractions (MVCs). For collection of MVCs, participants were instructed to maximally dorsiflex their ankles during a five second window, followed by an 8 second relaxation period. This was repeated three times for each foot. Then participants used activations of their TAs to play the commercially available computer games, Peggle™ and Peggle Nights™ (PopCap Games, Seattle, WA). In Peggle™ and Peggle Nights™, participants attempt to clear all orange colored pegs from a scene of blue and orange pegs by launching a ball from the top of the screen after rotating the launcher to the desired position. Participants controlled the aim of the launcher using their respective TAs (e.g., contracted their right TA while relaxing their left TA to move launcher to the right) and launched the ball by clicking a button using their hand (Figure 2).

Figure 2. NeuroGame Therapy game control. Active right tibialis anterior contraction (blue) with left relaxation (red) moved the ball launcher (top of screen) to the right, and vice versa. Ball launched using hand control when aligned to clear orange pegs.



Participants were asked to use NGT for up to six weeks, playing the game three to five days a week for 25- 40 minutes per day, followed immediately by a 5 minute walk without wearing AFOs (if typically worn) and practicing lifting their toes. FitBit Zip activity monitors

(FitBit Inc., San Francisco, CA) were worn to allow monitoring of the adherence to the walking portion of the protocol. Participants' parents had intermittent contact with the investigator during the intervention period in order to ensure that the system was working and that the level of challenge was appropriate. Participants were able to contact the investigator at any time they encountered difficulties. These difficulties were typically solved with one or two phone calls during the intervention, but required a home visit with three participants. Contacts ranged from 0 to 5 times (Mean 2.6).

### **Outcome Measures**

**Home therapy outcome measures.** The NGT software captured raw sEMG during each home therapy session. To be included in the analysis, home sessions must have lasted at least five minutes and have displayed modulations in recorded signals from both muscles to assure that the sensors were properly connected to the legs. Some home sessions were shorter than the minimum recommended 25 minutes due to technical difficulties such as battery discharging. Outcome variables included the number of home game sessions, individual muscle bursts per session, and maximal voluntary contraction amplitude. Bursts were defined as activity greater than one second in duration and amplitude exceeding 1.0 standard deviation of the entire game play recording for the individual day.

**Assessment outcome measures.** Assessment outcome measures were collected across the body structure/function and activity levels of the International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001) in order to determine the level of impact from NGT. These included body structure and function and activity tests as described in Table 2 and below.

Table 2. Brief description of outcome measures. Measures used based on the International Classification of Function domains.

<b>ICF Body structure/function domain</b>			
Test	Unit of Measure	Reported value	Psychometric Properties for children with CP
Surface Electromyography amplitude – Maximal Voluntary Contraction <sup>B</sup> (MVC sEMG)	mV	Peak amplitude of muscle activity during maximum voluntary contraction test	Not reported
Surface Electromyography – co-contraction ratio of Tibialis Anterior to Medial Gastrocnemius activity <sup>B</sup> (AROM and Walk sEMG)	Ratio	Ratio of activity detected in tibialis anterior versus activity in medial gastrocnemius during each of two defined events (active dorsiflexion ROM testing and swing phase of gait)	Not reported
Gait* – Velocity	m/s	Average velocity during walking	Test-retest ICC = 0.93 <sup>1</sup>
Gait* – Step Length	Cm	Average step length for each leg	Test-retest ICC = 0.90 <sup>1</sup>
Gait* – Base of support	Cm	Average distance between heels	Not reported
Gait* – Peak swing dorsiflexion (DF during swing phase)	Degrees	Average of peak dorsiflexion angles of each ankle during the swing phase of gait.	Not reported
Gait* – Dorsiflexion at initial contact (DF at initial contact)	Degrees	Average of dorsiflexion angles at initial contact for each ankle	Not reported
Balance	Seconds	Average of three trials of stance on single or both legs, per GMFCS level	Not reported
Spasticity	Score	Based on Composite Spasticity Scale	Test-retest ICC = 0.97 <sup>2</sup>
Passive dorsiflexion ROM <sup>B</sup> (PROM)	Degrees	Average of three passive ROM trials	5 <sup>0</sup> mean measurement error <sup>3</sup>
Active dorsiflexion ROM <sup>B</sup> (AROM)	Degrees	Average of five active ROM trials	Not reported
Muscle Contraction Force (MCF)	N	Average of five trials of dorsiflexion against mounted hand held dynamometer	Test-retest ICC = 0.62-0.91 <sup>4</sup>
Selective Control Assessment of the Lower Extremity (SCALE) <sup>B</sup>	Score	Score for each leg using SCALE grading criteria	Validity: r = -0.83 compared to GMFCS <sup>5</sup>
<b>ICF Activity domain</b>			
Falls	Number	Self-reported falls within previous week	Not reported
6-Minute Walk Test (6MWT)	Feet	Distance walked during 6-minute walk	Test-retest ICC = 0.98 <sup>6,7</sup>

Note: All Gait\* outcomes were computed using kinematic gait analysis over a minimum of 10 strides with each foot. <sup>1</sup>(Klejman, Andrysek, Dupuis, & Wright, 2010), <sup>2</sup>(Poon & Hui-Chan, 2009), <sup>3</sup>(Allington et al., 2002), <sup>4</sup>(Crompton, Galea, & Phillips, 2007), <sup>5</sup>(Fowler et al., 2009), <sup>6</sup>(Maher et al., 2008), <sup>7</sup>(Thompson et al., 2008). <sup>B</sup> – denotes blinding of the rater through altered file names for analysis.

## **Body Structure/Function Tests**

***Surface Electromyography (sEMG):*** Using a Delsys Bagnoli (Delsys Inc., Natick, MA), surface electromyography (sEMG) electrodes were placed over the motor points for the tibialis anterior, medial gastrocnemius, and soleus muscle bellies following skin cleaning and preparation (Botter et al., 2011). The electrode placements were measured relative to bony landmarks and were recorded for consistency in future testing. Data from sEMG were collected at 1000 Hz during laboratory assessments and low-pass filtered at 200 Hz. sEMG data were recorded during a maximal muscle contraction force and active dorsiflexion while supine on a plinth, and when walking at a self-selected pace on a straight, smooth, level walkway 8.8 meters long. Information on the processing of the sEMG data can be found in Appendix A. Analysis for sEMG data was performed by a research assistant blinded to the assessment number.

***Kinematic Measurement:*** Three-dimensional trajectories were collected using a Qualisys, Oqus 300 camera system (Qualisys AB, Gothenburg, Sweden) with eight cameras capturing reflective marker data at 100 Hz (standardized wand length standard deviation during calibration < 0.7 mm). Reflective markers were placed based on the Helen Hayes model (Kadaba, Ramakrishnan, & Wootten, 1990) on the participant's sternal notch, spine (level to marker on sternal notch), and bilaterally on the: acromion process, posterior superior iliac spine, iliac crest, anterior superior iliac spine, greater trochanter, 2.5 cm superior to patella, lateral knee joint, tibial tuberosity, lateral and medial malleoli, third metatarsal head, and posterior calcaneus (level in height to marker on third metatarsal head). Gait measurement consisted of ten trials of walking in bare feet back and forth on a straight walkway 8.8 meters long with a capture volume of the middle 4 meters to avoid capturing acceleration or deceleration. Velocity, step length, and base of support were computed as primary spatiotemporal parameters of interest through use of

the heel and toe markers. Joint kinematics were measured to identify peak ankle DF during the swing phase of gait and to detect the amount of DF at the time of initial contact. Detailed information on kinematic analysis can be found in Appendix B.

***Balance:*** Participants at GMFCS levels I and II performed unsupported single limb stance and participants at GMFCS level III performed unsupported double limb stance. In each test, an average of three trials was reported.

***Spasticity:*** Participants also were evaluated for spasticity using the Combined Spasticity Scale (CSS) during the first and fourth assessment (Poon & Hui-Chan, 2009). The CSS provides a score for each leg based on response to Achilles jerk, modified Ashworth, and clonus assessment.

***Passive Range of Motion:*** Passive ankle DF range of motion was assessed via video recording (Gilbertson et al. unpublished) for a minimum of three trials on each ankle while the participant was supine on a plinth. Passive ROM was video recorded and scored by the primary investigator blinded to the assessment number.

***Active Ankle Dorsiflexion:*** Active ankle DF ROM was assessed using 3D kinematic analysis using markers and the Qualisys system as described above. Within the cameras' view, participants performed five trials of active ankle DF while supine on a plinth with the primary assessor positioning their thigh in order to maintain the ankle position within the sagittal plane and to prevent knee flexion during activity. Each of the children moved their ankles for a minimum of five trials of DF with standardized verbal directions and encouragement from the primary assessor.

***Maximum Contraction Force (MCF):*** MCF was performed unilaterally in supine with knees extended using a digital hand-held dynamometer (MG200, Mark-10, Long Island, NY)

mounted onto an adjustable bracket. Participants performed 5 trials each of left and then right DF, followed by plantarflexion on each side, but only data from the DF were of primary interest in this study. The dynamometer was positioned so force translation would be through the metatarsal heads and this position was measured relative to the participant during the initial testing session for replication on subsequent test sessions. Standardized instructions and encouragement were provided for each trial (i.e. Ready, go, push, push, push, push, push, as hard as you can, as hard as you can, as hard as you can, and relax).

*Selective Control Assessment of the Lower Extremities (SCALE)* (Fowler et al., 2009): The SCALE was used to examine participants' ability to isolate muscle activity to move their individual hip, knee, and ankle joints according to specific administration guidelines. Scores were determined for each leg based on the ability to move each joint through the full range in a given timeframe. Qualitative observations were also noted (e.g., mirroring the motion of one leg with the other or moving other joints other than the specified joint). SCALE testing was video recorded and scored by the primary investigator blinded to the assessment number.

### **Activity tests**

Both participants and their parents completed the Fall History Report form indicating the number of falls and near falls the participant experienced in the previous week (Appendix C). Participants also completed the 6-minute walk test (6MWT) (Thompson et al., 2008) on a standardized looped course of approximately 450 feet. Participants' exact paths of walking were measured using a surveyor's measuring wheel (Stanley Tools, Townson, MD). Participants were provided standardized encouragement at each minute interval during the test.

## Procedures

All testing was conducted without the participant wearing ankle foot orthoses (AFOs) (if typically worn) and using their preferred assistive device (if necessary). Following informed consent/assent in the first assessment (Pre-Test (1)), participants' parents completed a health history form. Subsequent assessments began with both the participant and parent individually filling out the Fall History Report form. This was followed by balance, 6MWT, proprioception, CSS (on just pre-test (1) and post-test) and then the SCALE. After placement of the sEMG electrodes and while supine, participants performed five trials each of maximal voluntary contraction with their ankle against a stabilized dynamometer in the following order: left DF, right DF, right plantarflexion, left plantarflexion. Each ankle was then assessed for passive and active DF ROM with kinematic markers placed on the tested leg prior to active ROM to permit simultaneous kinematic and sEMG data collection. Pelvic, trunk, shoulder, and calcaneal markers were then placed and the ten walking trials were completed after two trials of standing to enable construction of a virtual model for analysis.

Participants were trained to use the NGT system after the second assessment (Pre-test (2)). Training consisted of an explanation/demonstration of computer and game set-up, electrode placement and connection, and program operation. Electrode placement for the tibialis anterior was based on the placement of the sEMG markers during laboratory assessments and was marked using a permanent marker. Participants received one or two training sessions in the lab prior to the investigator delivering the system and providing an additional training session at the participants' home within a week of the second pre-test.

## **Data Analysis**

Data analysis was performed using visual analysis of graphs of each child's performance across the five tests. Visual analysis was performed as this study had a small sample size. Preliminary descriptive review of the data suggested that there were not consistent results with similar changes across participants, thus the data did not warrant group statistical analysis.

## **Results**

Results for each participant, ordered by greatest amount of game play to least amount, are discussed below with selected figures presented for participants who showed interesting results. Figures for all individual results on each outcome measure at each assessment time can be found in Appendix D. The overall results are shown in Figure 3.

Figure 3 utilizes a color scheme to show improvement, slight improvement, no improvement, slight decline, or decline comparing the higher scoring of the two pre-tests to the post-test and follow-up test using green, yellow, white, orange, and red, respectively. These judgments were made by comparing the higher score of the two pre-tests to the post-test and also to the follow-up test. Change was determined by using Minimal Detectable Difference (MDD), if known, or by a greater than 20% difference from pre to post- or follow-up test. Slight change was determined if there was a change between pre and post- or follow-up test values of 10% to 19%. For the 6MWT, these values were determined based on the MDD of 188 feet (Goemans et al., 2013). For ankle range of motion, results were based on 4-6 degrees representing slight change and 7 or more degrees representing change as MDDs for ankle ROM in children with CP have variability within the literature (Allington et al., 2002; Wrobel & Armstrong, 2008). SCALE changes were determined by points where 2 points or more were indicative of change and 1 point indicated slight change. Change for all other measures was based on a percentage of

change from pre- to post- or follow-up test. Thus, a 10-19% change was used as a conservative figure to reflect change. For the purpose of analysis, the mid-test was used for establishment of a trend, but is not reflected in the summary of change or discussed further within this report.

Data from the FitBit activity monitor were not used to calculate steps during focused practice as data is only reported in fifteen minutes intervals and that time span was not specific enough for our purposes. The function of the FitBit was to monitor adherence to the walking portion of the protocol.

### **Andy**

Andy did NGT and walked in 19 sessions, totaling 550 minutes of NGT use during the 6 weeks of intervention. *Post-test results body structure/function:* He showed improvements with his MVC amplitude measured by sEMG unilaterally, his ankle DF angle at initial contact unilaterally, unilateral improvement in balance and a slight unilateral improvement on the SCALE in post-test. He showed regression in gait velocity, unilateral step length, and unilateral PROM. It was noted that there was a smaller standard deviation with ankle DF with both peak DF during swing at initial contact indicating greater consistency in this at the post-test. *Follow-up test results body structure/function:* Improvements were documented in his AROM sEMG unilaterally, ankle DF angle at initial contact unilaterally, and balance unilaterally. Slight regression was seen with gait velocity. Regression was seen in left AROM and muscle contraction force, with slight regression seen in right muscle contraction force. *Post-test results activity:* Andy showed slight improvement with the 6MWT at the post-test. *Follow-up test results activity:* He showed improvement in the 6MWT at follow-up and his self-reported falling reduced from 3-5 to 1 or 2 falls. Andy reported enjoying the game and he or his parents initiated three phone contacts to troubleshoot technical difficulties with NGT.

Figure 3. Results by participant.

Figure legend: Green box highlight indicate 20% or greater improvement between the higher value of the two pre-test data points and the post-test (P) or Follow-up (F) test. Yellow indicates a slight improvement and represents a 10-19% change from the higher of the two pre-test data points. White indicates a change of less than 10% either positively or negatively. Orange indicates a slight regression defined as 10-19% decline between the lower of the two pre-test data points and the post-test or follow-up test. Red indicates a regression (20% or greater change). For the 6MWT, change was based on the 188 foot Minimal Detectable Difference (MDD) with more than 188' equal to green, 94-187' equal to yellow, white for anything less than 94' change, orange a decline of 94-187', and red represents regression of more than 188'. For PROM and AROM, green indicates 7 degrees or greater improvement, yellow 4-6 degrees improvement, white 3 degrees or less change, orange a decline of 4-6 degrees and red a decline of 7 degrees or more. For the SCALE, green indicates an improvement of two points or more, yellow indicates an improvement of one point, white indicates no change, orange indicates a decline of one point, and red indicates a decline in two points or more.

	Body Structure/Function Tests																		Activity Tests				Intervention									
	MVC sEMG		AROM sEMG		Walk sEMG		Gait Velocity		Step Length		Base of Support		DF during swing phase		DF at initial contact		Balance		PROM		AROM		MCF		SCALE		Falls		6MWT		NGT Play	
	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	P	F	Sessions	Minutes		
Andy	*			*														*	*	*	*	*									19	550.1
Brandon	*									*								*			*	*									18	456.4
Ethan		*	*	*	*								*			*						*	*								18	390.6
Francine		*		*			*			*				*						*	*		*								14	356.0
Dianne				*		*							*						*		*	*	*	*							13	317.7
Helen		*	*	*	*	*										*			*		*	*	*	*							11	291.6
Carly			*	*	*	*		*		*					*	*			*		*	*	*	*			*	*			9	216.1
Irene	*	*	*	*	*	*							*	*	*	*		*	*	*	*	*	*	*	*	*					6	144.4
Gina	*	*	*	*	*	*									*				*	*	*	*	*	*	*	*					4	92.8

\* - Denotes unilateral change only,

Abbreviations: 6MWT – 6 Minute Walk Test, AROM – Active Range of Motion, DF – Dorsiflexion, MCF – Maximum Contraction Force, MVC – Maximum Voluntary Contraction, PROM – Passive Range of Motion, SCALE – Selective Control Assessment of the Lower Extremities, sEMG – surface Electromyography

## **Brandon**

Brandon did NGT and walked 18 times and logged 456 minutes of total NGT use during the intervention. *Post-test results body structure/function:* He showed improvements in sEMG amplitude during MVC unilaterally on his right and bilaterally with co-contraction during AROM. Gait velocity was the only spatiotemporal variable of gait to show a difference with slight improvement in this case. Brandon's standing balance without support and his AROM also improved bilaterally. He also showed slight left unilateral improvements with PROM and SCALE. *Follow-up test results body structure/function:* Improvement was seen in contraction during AROM bilaterally and with sEMG during walking unilaterally on his left, and with right step length. He showed slight improvement in AROM and SCALE scores. Slight regression was seen with left muscle contraction force. *Post-test results activity:* Brandon showed slight improvement in the 6MWT. *Follow-up test results activity:* There were no changes maintained at follow-up in activity measures for Brandon. Brandon also reported enjoying the game. His family did not need to contact the investigator during his home NGT use.

## **Ethan**

Ethan had 18 sessions of NGT and walking with a total of 391 minutes of training. *Post-test results body structure/function:* He had left unilateral improvement in his sEMG co-contraction during both AROM and walking. He also had bilateral improvement in balance and slight right unilateral improvement with his muscle contraction force. *Follow-up test results body structure/function:* He showed left unilateral follow-up improvements with sEMG amplitude and co-contraction during AROM and walking as well as balance and muscle contraction force. He showed slight regression with peak swing-phase dorsiflexion on his left. *Post-test results activity:* He had no improvements in the 6MWT or reduced falling. *Follow-up*

*test results activity:* There were no improvements seen in either activity test. Ethan frequently reported enjoying the game. His mother contacted the investigator four times for NGT support, primarily related to battery charging issues.

### **Francine**

Francine did NGT 356 minutes over a total of 14 sessions of playing and walking. *Post-test results body structure/function:* There were no positive changes in any of the three sEMG variables or spatiotemporal parameters of gait seen with Francine. She showed a slight improvement in balance, but no other positive changes in body structure/function tests. Francine showed slight regression with her base of support and with her right AROM. She showed regression with the co-contractions associated with AROM sEMG and with gait velocity. *Follow-up test results body structure/function:* Improvements with left MVC amplitude and right AROM co-contraction were noted with sEMG. Slight improvement with right sEMG co-contraction during walking, peak dorsiflexion during swing, and AROM were observed. The slight widening of Francine's base of support seen at the post-test was observed again at the follow-up test. Regression with right muscle contraction force was also observed. *Post-test results activity:* Francine showed improvements in her 6MWT. *Follow-up test results activity:* The changes in 6MWT were retained at follow-up. She reported no falls at any time during the study. Francine also requested a copy of the game for use after the study. Her father contacted the investigator four times during NGT play for technical support, usually involving the battery life or charging issues.

### **Dianne**

Dianne played NGT and walked 13 times, and had a total of 318 minutes of practice. *Post-test results body structure/function:* She showed no positive change with sEMG or any

spatiotemporal variables of walking, but did have slight improvements in right muscle contraction force and the SCALE on her left. Slight regression was observed in the amount of dorsiflexion at initial contact that was measured in her right ankle. *Follow-up test results body structure/function:* Dianne showed improvements in right muscle contraction force and bilaterally on the SCALE. She showed regression with co-contractions with left AROM sEMG and with right walking sEMG at follow-up. *Post-test results activity:* Changes were not seen in activity level testing at post-test. *Follow-up test results activity:* The only change that was observed in follow-up activity tests for Dianne was a regression in 6MWT distance. No falls were reported by Dianne at any point during this study. She requested a copy of the game for play after the study as she enjoyed it. Her mother contacted the investigator five times for NGT support with battery problems. Dianne also required one additional home visit to assist with electrode placement.

## **Helen**

Helen played NGT a total 292 minutes and walked for 11 sessions. *Post-test results body structure/function:* She did not show any positive changes with any sEMG or spatiotemporal gait variables. The only positive change seen was improved bilateral muscle contraction force. Helen showed regression with sEMG co-contractions for left AROM and right walking. *Follow-up test results body structure/function:* Helen showed improvement with her sEMG amplitude during MVC on her right leg. The bilateral improvements with her muscle contraction force were retained. There were no changes in gait variables. Regression was seen with sEMG co-contraction ratios for Helen's AROM bilaterally and walking on her right leg. Her left single leg balance was reduced at follow-up as well. *Post-test results activity:* Helen did not demonstrate any appreciable changes in activity tests at post-test. *Follow-up test results activity:* There were

no changes in 6MWT or falls reported during the follow-up test. Helen enjoyed the game and wanted to play more. Her father called one time for technical support during NGT play.

### **Carly**

Carly did NGT for a total of 216 minutes and walking for 9 sessions. *Post-test results body structure/function:* She showed improvements in unilateral sEMG co-contraction with left dorsiflexion during walking and bilaterally with muscle contraction force. Carly also showed slight improvement in the SCALE. She had reduced right AROM sEMG co-contractions, bilateral balance, and left AROM during the post-test. *Follow-up test results body structure/function:* Carly maintained the improvement in muscle contraction force bilaterally. She showed slight increases with sEMG amplitude bilaterally with MVCs and with co-contraction during walking unilaterally on her right foot. Carly showed slight regression with left step length and regression with right AROM sEMG co-contractions as well as gait velocity and balance. An increase in base of support (3.4 cm) was noted as regression. *Post-test results activity:* There was a slight increase in falls with a single reported fall compared to previous reports of zero. Slight regression with the 6MWT was also observed. *Follow-up test results activity:* The slight regressions seen in the post-test were both nullified and no positive or negative changes were seen in follow-up activity tests. Carly responded positively when talking about the game. There were no phone calls while Carly had the NGT system at home.

### **Irene**

Irene used NGT and walked 6 times and recorded 144 minutes of total NGT time. *Post-test results body structure/function:* Improvement was seen with left AROM sEMG co-contraction. There were not changes seen with any spatiotemporal gait variables. She showed improvement with her balance as well as her left leg on the SCALE. She had regression with

sEMG amplitude with MVCs and muscle contraction force bilaterally. *Follow-up test results body structure/function:* Irene had improved sEMG amplitude on her right during MVCs. She also had improvements with sEMG co-contraction ratio with left AROM. She had improved left PROM and her balance and left leg SCALE improvements were retained. Slight regression was seen with right dorsiflexion in both swing and at initial contact. Reduction in sEMG co-contraction was observed on the right foot during walking. Bilateral muscle contraction force was also reduced. *Post-test results activity:* Irene showed no changes in any activity measures. *Follow-up test results activity:* No changes were observed in activity measures. Irene stated she enjoyed the game. Her father contacted the researcher three times during the NGT intervention for technical support. An additional home visit was required to solve a charging and battery problem.

## **Gina**

Gina's NGT console malfunctioned during play and these technical issues erased Gina's home play data so her total NGT play is not known. Prior to the console malfunction, she had only logged 4 gaming and walk sessions for a total of 93 minutes, so results of game play data should be taken with caution. Her mother, however, reported adherence with doing NGT 2-3 times per week. She wore her FitBit activity monitor throughout the day, so walking practice after playing NGT was difficult to detect and provide a retrospective game-play amount to verify parent report. *Post-test results body structure/function:* Gina had improvements with her right sEMG amplitude during MVCs and with her left leg sEMG co-contraction during walking. No changes were observed with spatiotemporal parameters of gait. The only other test within the body structure/function domain that showed improvement was the SCALE, where she showed a two-point improvement on her right side. She demonstrated a slight decline with right sEMG co-

contraction during AROM and a decline in right AROM. *Follow-up test results body structure/function:* Gina had improvement with her left sEMG co-contraction during walking. She also showed improvements with left leg balance. Slight decreases were seen with her muscle contraction force on her left. Regression was seen with left MVC amplitude and right AROM sEMG co-contractions. Both ankles showed reduced AROM. *Post-test results activity:* There were no changes seen within the activity domain testing. *Follow-up test results activity:* As with post-testing, no changes were seen with activity tests. Gina reported enjoying the game, but said it was really hard. Her mother called the investigator three times about NGT issues including electrical interference with sEMG signals and difficulty with the software, and battery charging. An additional home visit was required for Gina due to difficulties with electrical interference.

## **Discussion**

The purpose of this study was to evaluate the feasibility and the preliminary effect of NGT paired with subsequent walking practice for improving TA selective muscle control and muscle function within gait activity in children with bilateral spastic type CP. Related to feasibility, overall, NGT was feasible as a biofeedback system for engaging the tibialis anterior muscles in children with CP who had spasticity affecting both lower extremities. All children participated in home game play and indicated enjoyment with the intervention and most wanted to keep a copy of Peggle for use after the study. NGT systems overall functioned well within the children's homes except for persistent problems with battery charging. Families needed occasional ongoing assistance (0-5 phone contacts) with three additional home visits required.

## **Body Structure/Function changes**

While there is variability among the children and across the assessments, all showed improvement in at least one of the body structure/function outcome measures used. In addition, all nine children showed improvements at the follow-up assessment with at least one measure. There appeared to be variability between the two pre-tests with many participants having better results on pre-test (1) compared to pre-test (2). This could be due to motivation of the participant as a learning effect would be expected to have the opposite effect and yield better results at pre-test (2). With downward trends prior to intervention, the positive changes seen lead to the assumption that NGT may be causing an effect.

The measure with the greatest amount of improvement was the co-contraction ratio with sEMG while performing AROM with three children showing improvement at the post-test and five showing improvement at the follow-up session. As one of the purposes of this intervention was to increase recruitment of the tibialis anterior muscle, this result is appropriate. One child (Francine) actually showed bilateral regression at post-test but had unilateral improvements at the follow-up. The other four children showed regression at follow-up, but many of the participants scored very small numbers to begin with (e.g., less than 4.0), so 20% change is small. Also, because the co-contraction is expressed as a ratio, changes to either the numerator or denominator can affect the product.

Given that muscle recruitment of the TA was a primary goal of the NGT practice designed for this study, it was surprising that there was not more evidence of improvement in sEMG amplitude during maximal voluntary contraction or with active movement. This may be related to the amount of NGT practice. In this study, participants used NGT for a total of 1-9 hours, with only 3 participants using NGT for more than 6 hours. In comparison, Rios et al.

(2013) reported NGT use from 4 to 10 hours and Donoso Brown et al. (2014) reported NGT use from 5-21 hours. The positive effects seen in the Rios et al. (2013) and Donoso Brown et al. (2014) studies are likely, at least in part, due to the increased amount of game play. Though overall more NGT time was reported, both of the other studies had shorter (no more than 4 weeks) at-home NGT use. It appeared that changes seen with sEMG outcomes were associated with the amount of game play. Children in our study who played the minimum recommended time (i.e., 3 times per week) showed the greatest improvements in sEMG outcomes, while the others did not show the same improvements.

Part of the lack of improvement in sEMG variables may be due to the method of analysis, i.e., analyzing change based on the amount of co-contraction of the agonist TA compared to that of the antagonist medial gastrocnemius. In the other two NGT studies (Donoso Brown et al., 2014; Rios et al., 2013), sEMG activity practice was associated with agonist/antagonist pairs where participants were required to actively relax the antagonist while actively contracting the agonist to enable game control. In this study, neither the medial gastrocnemius nor soleus sEMG was utilized during game play, rather this study required quiescence of the contralateral agonist muscle. Though Toner *et al.* (1998) reported a decrease in antagonist activity as a result of agonist training, our study only supports the results found by Toner et al. to a minimal extent.

Based on the recruitment of the TA for NGT game play and the addition of a walking practice following NGT play, the anticipated result was an improvement in DF at initial contact or during mid-swing phase at both post-test and follow-up sessions. Only one participant showed unilateral improvements with DF at initial contact and none within the swing phase of gait. Though instructed to practice lifting their toes during the walk, there were no data collected to determine if the children actually practiced this skill. Their ankle dorsiflexors may have been

fatigued from NGT and thus less capable of rehearsing the desired movement. The lack of improvement with DF during the swing phase of gait indicates that children may not be clearing their toes more while walking and are still at greater risk of tripping and falling. Though one child, Andy, reported fewer falls, most children did not fall frequently to begin with and thus had no room for improvement. Andy was the only child to show improvements in DF at initial contact so there may be an association between those two improvements.

The lack of improvement with the swing phase DF parameter may also be a result of the complexity of walking where changes in other kinematic aspects such as hip and knee angle can either assist or negate ankle movement changes. Another possibility is the children's position during NGT. Though we asked children to play the game with their knees straight and feet on a stool, data do not exist to monitor adherence with this aspect of the study and direct observation of game play during two home visits to troubleshoot NGT problems with two participants suggested this portion of the protocol was not always followed. The knees straight position was chosen to put the legs in a position similar to that of initial contact during the gait cycle. This could have had an effect on the recruitment of the TA outside of NGT play. Due to the play and recruitment of the TA in a mid-range of plantarflexion, there was also a lack of work at the end of the range of motion, or attempt to achieve a new range of motion. This likely also relates to the lack of changes in range of motion.

Though five children showed at least slight improvement in balance, only four maintained these improvements at follow-up. Three of those children were among the four youngest children in the study, so these improvements may be due more to natural maturation as opposed to NGT. The fact that two of the four children used NGT the least points to other factors being involved. However, the other two were in the top three NGT users. Improved TA

control should theoretically lead to improved balance via improved ankle control which may be substantiated by the fact that the other child was the oldest in the study, so maturation was not likely a factor. Three of the four also showed improvement with AROM sEMG co-contraction, so muscle isolation may be the biggest key to the improvement of balance.

Though some slight changes were seen in the SCALE at post-test, few were seen at follow-up. The SCALE may not have been a sensitive enough measure to show subtle change in individualized joint movement as a result of practice with TA recruitment. Most children scored a partial ability score with ankle movement indicating that they were able to selectively move the ankle through at least a partial range. Though this range may have increased some, the measure did not reflect an improvement. The qualitative aspect of the SCALE during reciprocal ankle plantarflexion/dorsiflexion did indicate less mirroring on the contralateral limb and fewer movements of other joints, or increased speed of recruitment for several participants. Due to the lower sensitivity of the measure to small differences, administration of the SCALE could have also been performed with the kinematic marker set applied to better quantify the results.

### **Activity changes**

We anticipated finding improvements in the 6MWT by virtue of the addition of the walking practice following NGT play. Data gathered from the FitBit activity monitor were only specific to 15-minute intervals of time and so were not detailed enough to determine the actual amount of walking in the 5 minutes following game play. Thus, number of steps during post-NGT focused practice were not calculated. While all children showed some activity on the FitBit after the NGT play, the amount of walking may not have been enough to influence carry-over of TA control to an improved gait pattern.

None of the participants had frequent falls (i.e., more than 3 per week) and were therefore near the ceiling of that outcome measurement. The questionnaire also only covered falls within the past week, so may have been more dependent on the timing of assessments as opposed to the true number of falls across time. More specific, sensitive and standardized testing of falls needs to be developed.

## **Limitations**

This research had several limitations related to the NGT system and to the assessment measurements. There were some technological issues experienced with NGT systems such as the laptops not charging correctly or other difficulties using the system, which may have decreased motivation and therefore adherence with game play. The sEMG electrodes used for game play were applied at home using pre-determined and marked sites. The markings wore off quickly with many participants (especially those who swam frequently) and this may have influenced electrode placement and therefore the game play specificity, though mitigation of this was attempted through providing pictures of the appropriate set-up and training parents in how to take measurements to re-locate the correct attachment location. Outcomes could have also been affected by the design of the NGT practice itself. The game did not require reciprocal bilateral muscle activity similar to alternate bilateral muscle activity bursts for walking. Within the NGT protocol used, for success the participants could have used one long burst of muscle activity to move the launcher to one side of the screen and then clear pegs by shooting the ball after small launcher movements (requiring less muscle activity) towards the other side.

During assessments, though procedures were taken to maintain as much consistency as possible, sEMG electrodes can be placed in slightly different places from session to session, which could have an impact based on proximity to motor end plate. A slight change with

application of the kinematic markers could have an influence on range of motion data. Set-up of the participant and the mounting bracket used to house the hand held dynamometer used in MCF testing was also difficult to replicate even with multiple set-up measurements taken to ensure higher consistency of testing. Slight changes in position would affect the angle of the ankle and could therefore have consequences on the muscle contraction force.

### **Future Research**

Future research using NGT on the lower extremities could focus on influencing the amount of gastrocnemius/soleus activity during TA activity by using one channel on the agonist muscle while applying the other channel on the antagonist muscle for game play and increasing the amount of movement practice after NGT. This could be targeted to children with unilateral CP and have a similar application to the NGT studies utilizing the agonist wrist extensors and antagonist wrist flexors where success was not only predicated upon activation of the desired muscle group, but inhibition of the antagonist group. Other studies could determine if NGT is more beneficial to children at a specific GMFCS level, or children who have different types of gait (i.e. equinus or jump). Using NGT on younger children who may not have as established patterns of movement is another possible area of study.

### **Conclusion**

NGT was a feasible and accepted method for home practice of tibialis anterior muscle recruitment for children with CP who have spasticity affecting both lower extremities. Small changes were seen across all children in some outcomes with the exception of no improvement found in PROM. It appears that children who played the game the most often showed the greatest improvements with sEMG variables, which matches with motor control and learning research studies (Johnston, 2009; Kleim & Jones, 2008). Changes in outcomes other than sEMG

and balance did not appear to have demographic commonalities among participants, such as age, gender, or GMFCS level. Though NGT appeared to influence changes in outcomes measured at the post-test, few of these changes were maintained at follow-up. This suggests that NGT may be beneficial to train voluntary selective control of muscles, but a more intense bout of NGT training may be necessary followed by greater practice of a functional task. Repeated episodes of use of NGT may also be necessary to maintain the selective motor control over longer periods of time.

## **Chapter 5:**

### **Summary**

The incidence of children with cerebral palsy (CP) remains high with 4-5 per 1,000 affected (Aisen et al., 2011). Due to the physical limitations that accompany a diagnosis of CP, many intervention studies have been conducted to help inform clinicians of the best treatment strategies. Many studies have required children with CP to perform repetitive movements outside of therapy sessions with goals of enhancing strength, coordination and/or mobility, but adherence to home exercise programs has been poor (Chappell & Williams, 2002). Use of virtual reality (VR) or other video game platforms have become popular as they tend to be more engaging for the children in both home and clinic settings (Bryanton et al., 2006). A problem with many current VR and gaming platforms is that they require active movement of impaired limbs to generate movement of the avatar on screen. As a potential bridge to the gap between difficulty creating active movement and requiring active movement to control an avatar, a NeuroGame Therapy (NGT) system was devised. This NGT system utilizes surface electromyography (sEMG) to provide real-time biofeedback to assist in training muscles that are difficult to activate where muscle activation allows control of a popular and commercially available video game. The overall purpose of this dissertation was to evaluate the effectiveness of NGT to improve activation and control of the tibialis anterior (TA) muscles in children with CP.

To examine the effectiveness of the NGT system in recruiting the bilateral TA muscles in children with CP for the ultimate purpose of improving walking, selection of appropriate gait outcome measures was required. A review of the gait measures used with children with CP and an examination of their psychometric properties found that the 6-Minute Walk Test (6MWT) had

the highest amount of evidence of validity and reliability supporting its use in children with CP, therefore it was selected as one of the outcome measures. Measuring other spatiotemporal parameters as well as some kinematics of gait was also desired for outcomes in the NGT study. Since the gold standard of 3-dimensional kinematic (3DK) analysis was possible for the spatiotemporal and kinematic variables of interest, 3DK was selected rather than one of the other kinematic based gait measures evaluated in the review paper.

Beyond the outcomes of gait, range of motion (ROM) was also determined to be an important body structure outcome of the NGT study. Assessment of ROM is typically done in real-time. Within research this can introduce bias due to lack of blinding and it is also often difficult for one person to perform reliably in children with CP. An alternative method for ROM assessment was devised and studied to determine its concurrent validity and test-retest, intra-, and inter-rater reliability. A standardized method of video recording the assessment of ROM and measuring the values post-hoc was devised. Both children with CP and those who were typically developing were enrolled in the study. The results of that study using Intraclass Correlation Coefficient (ICC) analyses showed evidence of concurrent validity when comparing the values measured post-hoc to those gathered in real-time using standard goniometry procedures. There were also high ICCs when comparing the post-hoc measurements from videos of a licensed physical therapist to two different physical therapy students and when comparing the measurements of these individuals to themselves with a second measurement of the same videos. This study also examined test-retest reliability with assessments at least 3 days apart and found little difference in children with CP and in the children who were typically developing. Based on these analyses, taking an average of three trials was determined to yield the highest reliability

with the fewest trials. The results of this study provided evidence for utilizing this ROM method within the NGT study.

Within the NGT study nine participants completed the full 12-week protocol, which consisted of two pre-tests, 6 weeks of NGT paired with walking practice and two post-tests. The children participating had CP affecting their bilateral lower extremities and difficulty clearing their toes when they walked. The NGT was focused on bilateral activation of the TA muscles to improve active dorsiflexion in attempt to ultimately reduce toe drag while walking and decrease falls. Many measurements were made at the body structure/function level (i.e., sEMG amplitude during maximal voluntary contraction, sEMG co-contraction ratio of the TA compared to the medial gastrocnemius during active ROM and walking, gait velocity, step length, base of support, amount of ankle dorsiflexion during swing phase and at initial contact, balance, passive and active dorsiflexion ROM, muscle contraction force, and selective motor control) and at the activity level (i.e., 6MWT and self-reported falls), which were examined individually for improvements from pre to post-tests. Each participant showed some improvement in at least one body structure/function measure at the immediate post-NGT assessment, however there were other individual improvements and no clear similar improvements across all participants. Co-contraction between the TA and medial gastrocnemius during AROM was the only outcome in which five of the nine participants showed improvement at the follow-up assessment, but only three children showed improvement at the post-test. Oddly, every child who didn't show an improvement with this variable showed regression, at least unilaterally. Amplitude of sEMG during maximum voluntary contractions had all but one participant show improvements at either post-test or follow-up, but none of the children had discernable change in both assessments. Four children showed some improvement in balance on the post-test, and four showed

improvement on the follow-up. Muscle contraction force was the only other variable where at least three children showed improvement at the follow-up test. The selective control assessment had six children show at least slight improvement at the post-test with three children retaining gains at the follow-up. Though many children showed improvements, only two participants showed carryover into improved walking distance on the 6MWT. The results of NGT use in this study showed similarities to previous studies of NGT applied to improve wrist dorsiflexion in children with CP and adults post-stroke (Donoso Brown et al., 2014; Rios et al., 2013), but overall findings were not as consistent or robust.

The three participants who showed the most improvement were the ones who used NGT for 391 to 550 minutes. These three participants represented every ambulatory level of the GMFCS, were all male, and were all involved in recreation or sports outside the home. This level of NGT repetition combined with the outside sports activities may represent the amount of practice needed to effect outcome. The finding that similar outcomes occurred with children from each GMFCS level suggests NGT may be effective for children at any GMFCS level rather than having a greater effect on children at one specific level of functioning. Given that all of these children were male may suggest that males enjoyed the video game aspect more than the females in the study, or were more motivated to play NGT.

### **Future Directions**

Through the review of gait measures currently being used for children with CP, it is clear that the psychometric properties of these measures needs to be more carefully examined on children with CP. Evidence should be generated to either support the 10-minute walk test, or to have more conclusive evidence that the 6-minute walk test is just as valuable for assessment without the extra time. It is also important to determine if passive range of motion and active

range of motion can both be accurately assessed using the video recording with a post-hoc measurement method.

NeuroGame Therapy had mixed results with this sample of children. Though the results were mixed, NGT may be a promising new technology that has a place within the habilitation or rehabilitation of children with bilateral CP, and potentially other children with CP who have trouble with gait. Specific follow-up studies could be conducted to further evaluate the use of NGT to improve selective muscle activation in children. Some questions that could be answered through future studies are described below.

Research related to specific follow-up to the current study:

- 1) Would alterations in the study design provide results that are easier to interpret? Since there were only two assessments prior to NGT use, the baseline of children's function was not well established. Conducting an experiment with study design more similar to single subject study design may be more effective. Collecting baseline data until a clear trend has been established prior to introducing NGT may lead to a better understanding of the effect of NGT. At a minimum, at least three assessments prior to baseline seem pertinent to establish a more reasonable trend.
- 2) How can the methods used in the data collection be improved to increase reliability or sensitivity? Using markers on the skin leaves room for error and dealing with markers where 1 mm difference in placement from one time to another could make a huge difference in the outcomes. Finding ways to attach electrodes and reflective kinematic markers to improve test-retest reliability is important. One idea may be to make an orthotic out of Aquaplast® (Patterson

Medical, Bolingbrook, IL) or another quick and easy-to-mold hard material that has holes where the electrodes or markers should be placed. This might improve placement reliability. This could also be used to improve reliability of home NGT electrode placement. A device could also be used to ensure that the reflective marker on the calcaneus is level with the one on the 3<sup>rd</sup> metatarsal head for improved reliability of dorsiflexion measurements. Beyond electrode and marker placement, increased reliability with determining changes on the SCALE could be used via utilizing three-dimensional kinematic analysis during administration of this measure. The other measure that appeared to have the most problem with test-retest reliability was measured maximum contraction force. Some ways to improve reliability with this measure may be to include a heel cup in the bracket that housed the hand-held dynamometer so the child's foot always started in the same position. Also, putting the children's lower extremity in straight knee splint might mitigate increased forces into the dynamometer as a result of knee flexion. Though the primary investigator held the children's knees straight, the amount of support may have varied between sessions and participants.

- 3) What measures were truly necessary to capture NGT effect? Though most outcome measures were useful in understanding the NGT, several measurements appeared to be extraneous and did not seem retrospectively relevant to be completed. The measures that appeared to be most relevant to NGT included: sEMG for MVC and walking, kinematic data for step length and dorsiflexion both during swing and at initial contact, maximal contraction

force, the ankle portion of the SCALE, reported falls, and the 6MWT. Gait velocity was not a target of NGT and it could be assumed that increased 6MWT scores would indicate a change in gait velocity. As participants were not using their full available ROM or going into the end range of their dorsiflexion ROM during NGT, it is not surprising that neither PROM nor AROM changed. Thus, if the protocol remains the same, these measurements are not necessary. The sEMG co-contraction during AROM is also less useful and changes in sEMG should be seen through the MVCs or during walking. Though gait velocity and base of support may not be directly relevant to NGT, these data are very easily computed given the kinematic data collected during walking, so there would be little reason not to compute them, but reporting them may not be as relevant in future studies. It is felt that the assessment of balance was less relevant for these children as many of the gains seen were still minimal and those changes might be evident through adding a stance time variable to the kinematic gait analysis, which would not take extra assessment time. The SCALE was not a very sensitive measure, but there were some good anecdotal data that came from the assessment of the ankle portion of this test. Completing the SCALE, as noted above, with kinematic markers during administration of this measure might increase the sensitivity of the measure to change.

- 4) As it seems that children who used NGT more, potentially were more active walkers, and showed more changes, what factors could lead to increased NGT adherence? Would less overall time having NGT at home increase the amount

of game play? Rather than six weeks of NGT play, perhaps using the four weeks of the other studies is a more reasonable amount of time and less daunting for the participant. Both the Rios et al. (2013) (4-10 hours of total game play) and Donoso Brown et al. (2014) (5-21 hours of game play) studies had more overall game play in the shorter intervention window than this study (1-9 hours). Another difference between this and the other two studies was the amount of training in the laboratory. The other studies required participants to come to the university generally at least three more times for game training whereas this study only had one game training session in the lab and then a game training session combined with the home delivery within a few days of the single laboratory training. This was in effort to boost participant retention. Perhaps more training in the lab or home would add to NGT comfort and thus more home use. The protocol itself may have needed to be more explicit about the amount of game play where participants could be told they need to use NGT a minimum of 5 times per week rather than being told to do NGT 3-5 times per week. Removing the variability within the expectation of use may be helpful. Adding reminder emails or phone calls to the parents or participants on a weekly, or more often, basis may have improved adherence. Children may also benefit from a more explicit incentive such as a specific dollar amount or other toy or item given for each time they did NGT. Perhaps a system where the child receives a sticker or ticket for each time they used NGT and then got to redeem them at the end of

the study for some prize contingent on the number of NGT sessions (i.e., better prizes for more NGT).

The other factor that was discussed was the limitation of the technical issues with some of the NGT systems. This may have dis-enticed participants from playing the game and simply having updated computers could have led to increased adherence.

- 5) Along the line of NGT adherence is the game itself and the question: Is Peggle™ the most appropriate game for children with bilateral involvement to play? Peggle™ allows the participant to take as long as they wanted to line up a shot and it doesn't require the person to use both legs for any given shot. Data from game play suggests that there was fairly equal recruitment during the course of play though. Walking requires quick movements to lift toes prior to heel strike and thus, training without a timing component or reciprocal activity may not be achieving the intended results. By playing games similar to a ski slalom or other game which require alternating activation of each TA under a time constraint may be more similar and have a greater transfer to walking. The last factor to enhance NGT home game play may be to ask if children might have benefitted from a variety of games rather than playing Peggle™ the whole time? Even if the games were not commercially available and had less exciting graphics, effects, and music, the novelty of several new games might have been more stimulating and have increased NGT adherence. Perhaps a study with a consistent period of training (1-week intervals) could

be used: one that plays Peggle™ and one that plays a variety of non-commercial games, changing each week.

- 6) The last question to be considered with modifications to this protocol related to game play is: are we providing the “just-right” challenge during game play? This protocol used an algorithm to determine the amplitude gains or adjustments based on the MVCs for each NGT session. This may not have been the best for the children as it could have made things too difficult at times and therefore frustrated the child. Other times, it may have been too easy and did not challenge the participant enough to effect the neuroplastic changes for which NGT was designed. Perhaps reverting back to the previous protocols of the investigator determining the initial level of gain and then making remote adjustments at appropriate intervals would have yielded greater results.

Research related to better carry-over to function:

- 1) How is NGT best used to influence functional change? Protocols could be developed to compare NGT paired with 5 minutes of walking to: NGT without paired walking and walking without NGT. This comparison may provide insight into whether it is NGT that is assisting with functional change, or whether it is simply practice with the function. Another concept that may be incorporated is to develop a game timed to walking on a treadmill such that success in the game would be contingent on alternating TA activity that corresponds with that specific portion of the gait cycle (i.e., when the right leg is in swing phase and right TA activation is the goal, the game would be set to

require a movement to the right at that same time, and vice versa). Perhaps playing NGT in stance and asking participants to lift their leg to activate the TA might be enough to influence walking.

- 2) How much NGT game play is required to affect change in movement behavior and how frequently does NGT need to be used to maintain change? Previous studies using NGT have varied in the length of the intervention phase when participants used NGT within their home. To further evaluate the intensity of NGT use, individuals could start NGT and assessments could be performed at regular intervals until a plateau was reached in the outcome measures used. This timing of a plateau could be very informative for determining the duration of NGT game play needed to affect maximum recovery and possibly show discrimination between children in different GMFCS levels. If the technology was available to the researcher, pairing NGT with brain imaging studies such as diffuse tensor imaging would also allow for examination of neuroplastic changes that may also inform future research about the intervention time-frame needed.

Research related to the broader application of NGT use in children with CP:

- 1) Are children in one particular Gross Motor Function Classification System (GMFCS) level better candidates for NGT than others? This question is much more reliant on a large sample size to be able to analyze performance on each group of children, according to their GMFCS levels and ability to independently operate the NGT system by placing their own electrodes. Though our study didn't suggest greater benefit of NGT by any particular

group based on GMFCS level, this may be a factor. By understanding if children at a specific GMFCS level benefit from this type of intervention more than children at other GMFCS levels, future intervention research could be more directly and specifically designed for maximal benefit. With a large enough sample size, a randomized controlled trial would be a better design where a control could just get their typical therapy.

- 2) Another question could be do children with a specific gait pattern benefit more from NGT than others? In this study, the majority of the participants had an equinous gait where hips and knees are extended and the foot is plantarflexed. However, a few had more of a jump gait where the ankle stays plantarflexed and the hips and knees remain flexed. A study comparing these two groups could also be very useful to determine if NGT is more effective for targeting dorsiflexion during gait.
- 3) Is NGT more effective for children with unilateral CP versus bilateral CP? sEMG is a very promising technology and may be of benefit to other groups of children. NGT may be more appropriate for children with unilateral CP rather than bilateral CP as these children tend to have asymmetrical gait, and training the TA to reduce toe-drag may lead to a more symmetrical gait pattern with these children. Recruitment of children with unilateral CP may be easier as well which would hopefully lead to a larger sample size and therefore, more robust data for analysis.
- 4) Is NGT feasible with younger children (i.e., working towards habilitation vs. trying to change movements that have become routine)? NGT experimentation

with a younger population may yield different results as these children would not be as set in their movement patterns. Initially, it was thought that older children would be more able to operate the NGT system, which is why the sample for this NGT study included children from 8-18 years. It seems that with technology becoming much more accessible, children at younger ages are learning to use computers. Using NGT requires few skills beyond turning on a computer and launching a program. Young children would likely need their parents there to provide assistance as needed with the electrode placement, but could otherwise potentially operate NGT independently.

## **Conclusions**

A single “best” measure for the evaluation of gait in children with CP does not exist as all measures should be chosen based on the child and intended goal for intervention. Some measures have greater responsivity based on the parameter of interest. The psychometric properties of each tool are different and should be considered when choosing a specific tool for each patient population.

Assessment of ankle dorsiflexion ROM in children via post-hoc video analysis is feasible and supported by psychometric testing. High levels of test-retest, intra-rater, and inter-rater reliabilities were found, especially when averaging three trials. Being able to utilize a video camera for assessment of ROM saves time for the child during testing and also allows for blinding of the rater to reduce potential bias during research.

Though NGT is feasible in children with CP, the effect on the body structure/function impairments and walking activity within this study was small. All participants showed some improvement in at least one of the outcomes, but the outcomes with improvement were

somewhat inconsistent across participants and evidence of retention of those improvements was sparse. The sEMG variables of amplitude in MVC and co-contraction during AROM were the only outcomes in which more than half of the participants showed improvement at the follow-up test. Both the balance and SCALE showed at least slight improvement with five or more of the nine children at the post-test. Muscle contraction force had nearly half of the participants show improvement and the only variable where all of the children who showed initial change retained that improvement at the follow-up assessment. Walking sEMG was also improved or slightly improved with nearly half of the participants at follow-up when only 3 children showed changes at post-test. There was not much effect seen in spatial, temporal, or kinematic parameters of walking suggesting that there was less carry-over of learned muscle use into the functional task. The three children who played NGT the most showed the most positive change across all measured variables. Based on these results, it is suggested that a higher intensity of NGT may be necessary to initially learn to recruit the muscles of interest. It is likely that children would still need to use NGT occasionally to maintain the active muscle recruitment or improved functional skill gained during the more intensive training.

## **Appendix A: Information on surface electromyography (sEMG) analysis**

Data for sEMG recorded in the laboratory were processed using custom LabView (National Instruments Corporation, Austin, TX) software. The sEMG level during maximal voluntary contraction (MVC) was calculated for each muscle as the average of the peak amplitudes over 5 trials per assessment was performed in both dorsiflexion and plantarflexion. To calculate normalized co-contraction ratio over the period of tibialis anterior (TA) activation, the integrated signal for the TA was divided by the TA MVC and then divided by the integrated signal for the ipsilateral medial gastrocnemius over its respective MVC. This made a ratio greater than one indicative of agonist activation.

### **sEMG during active movement**

After testing, the sEMG and video data were manually synchronized with the aid of a light emitting diode (LED) (placed in the view of the camcorder) that indicated the initiation of EMG data capture. The active range of motion task start frame was defined by a trained research assistant as any frame prior to initiation of muscle activity or movement. The stop frame was determined automatically by the program as a value equal to 3 seconds after the event initiation determined using the threshold method described later. For the walking sEMG, the step was determined by the primary researcher as events within the kinematic analysis. The events of toe off and initial contact were manually chosen by the primary investigator using the following criteria: toe off defined as the frame when the toe marker showed an upward and forward trajectory and the knee maintained a stable angle, and initial contact was defined as the frame when the heel (or toe depending on the child) remained constant in both the sagittal and vertical planes while the other foot or ankle markers continued to move downward in the vertical plane.

Each of these defined events had a time signature associated with it and those time signatures were used for event analysis in the sEMG program.

The EMG signals were sampled at 1 kHz using a Delsys DS-160 EMG amplifier digitized by a National Instruments USB-6009, 14-bit A/D converter mated to an Apple MacPro running custom LabVIEW data acquisition and analysis software. Signals were rectified and low-pass filtered using a forward/reverse pass 4th order Butterworth filter at a frequency of 20 Hz (per pass).

To determine an EMG contraction event, the signal's onset and termination were determined automatically using a threshold method. A burst of muscle activity was determined for each muscle group (right and left TA) and was defined as the EMG signal exceeding the threshold. The threshold was governed by the mean quiescent EMG value (the lowest value for muscle activity observed), plus five standard deviations of the quiescent signal. This value was multiplied by 5 to identify and isolate only high intensity contractions and reliably identified muscle activity bursts for each task.

The agonist/antagonist ratio for the period of the agonist burst was then calculated (the agonist co-contraction ratio). For the tasks chosen, the agonist was the TA and the antagonist was the medial gastrocnemius. This was normalized to activity during the Maximum Voluntary Contraction (MVC) for that session, to control for variations across sessions. Higher normalized co-contraction ratios signify increased independence of agonist and antagonist.

### **Data processing of sEMG during game play**

sEMG data from the game play sessions were down sampled to 500 Hz and low-pass filtered at 200 Hz before rectification and binning into 10 ms windows. Sections of data greater than 3 standard deviations above the mean were removed as likely due to non-physiological

signals (e.g., wire movement artifacts). Wavelet analysis was used to establish a reliable baseline level of EMG, which varied in the home due to changes in environmental noise and variations in electrode placement. The Daubechies wavelet family with an order of 44 (Wavelet Toolbox, MatLab release 2011a, The Mathworks, Natick, MA) was used as the mother wavelet function to perform feature extraction from the signal recorded during each game play session [29, 30]. Daily baseline values were calculated using the mean signal amplitude during all times where features were not detected, and this was subtracted from the rectified signal. The maximum muscle signal for each session was then calculated as the average of the three largest peaks for each muscle. A burst of activity in each muscle was subsequently defined as features in the binned data that exceeded 15% of the maximum activity for at least 0.5 s.

## **Appendix B: Information on 3-dimensional kinematic (3DK) analysis**

Kinematic analysis was performed using Visual 3D (C-Motion, Inc., Germantown, MD). Marker position data was filtered using a fourth order Butterworth lowpass filter with a 6 Hz cutoff. Custom Visual 3D analysis coding was used to calculate spatiotemporal and kinematic parameters of interest during the walking and active ROM tasks.

During walking, initial contact and toe off events were manually identified by the primary investigator using the following criteria: initial contact was defined as the frame when the heel (or toe depending on the child) remained constant in both the sagittal and vertical planes while the other foot or ankle markers continued to move downward in the vertical plane, and toe off defined as the frame when the toe marker showed an upward and forward trajectory and the knee maintained a stable angle. Spatiotemporal parameters included gait velocity, step length, and step width (base of support). For walking, spatiotemporal and kinematic parameters were calculated on a step or stride basis, as appropriate, and averaged across steps or strides for a single participant. Velocity was calculated as the displacement of the heel marker from one initial contact to the next initial contact on the same foot, divided by the time between initial contact events. Step length was calculated as the displacement of the heel marker from toe off to the next initial contact on the same foot. Step width (base of support) was defined as the distance (perpendicular to the direction of travel) between the heel marker on one foot at initial contact and the heel marker on the opposite foot at the subsequent initial contact.

Kinematic parameters during walking included peak dorsiflexion during the swing phase of gait and the degree of ankle dorsiflexion at initial contact. Dorsiflexion was defined as the sagittal plane angle between the shank (defined by knee joint, tibial tuberosity, and lateral malleolar markers) and the foot (defined by the heel, lateral malleolar, and 3<sup>rd</sup> metatarsal head

markers). Peak dorsiflexion during swing phase was defined as the maximum dorsiflexion angle during swing phase (from toe off to initial contact on one leg). Dorsiflexion during initial contact was defined as the computed dorsiflexion angle at the frame manually defined as initial contact.

For active ROM trials, peak ankle dorsiflexion was determined for each trial. Values were reported as averages across all trials of movement for a given participant.

University of Washington  
Department of Rehabilitation Science

### Neurogame Therapy Study

Participant report form

Date today: \_\_\_\_\_

A fall is when you touched the ground with a part of your body other than your feet.  
In the past week, how many times do you remember falling?

\_\_\_\_\_ 0 times      \_\_\_\_\_ 1 or 2 times      \_\_\_\_\_ 3 to 5 times      \_\_\_\_\_ more than 5 times

If you fell, where did you fall?      \_\_\_\_\_ inside      \_\_\_\_\_ outside      \_\_\_\_\_ both

If you fell, where did you fall the most?      \_\_\_\_\_ inside      \_\_\_\_\_ outside

If you fell, please tell me the type of surface(s) that you fell on? (examples might include carpet, tile, sidewalk, grass, bark chips)

\_\_\_\_\_

If you fell, were you wearing ankle or foot braces at the time?      \_\_\_\_\_yes      \_\_\_\_\_no      \_\_\_\_\_both

If you fell, were you using a walker or other support?      \_\_\_\_\_yes      \_\_\_\_\_no      \_\_\_\_\_both

A near fall is when you stumble and have to brace yourself on another person, a wall, or another object like a railing or piece of furniture. Using a walker or other walking aide does not count.  
In the past week, how many times do you remember having a near fall?

\_\_\_\_\_ 0 times      \_\_\_\_\_ 1 or 2 times      \_\_\_\_\_ 3 to 5 times      \_\_\_\_\_ more than 5 times

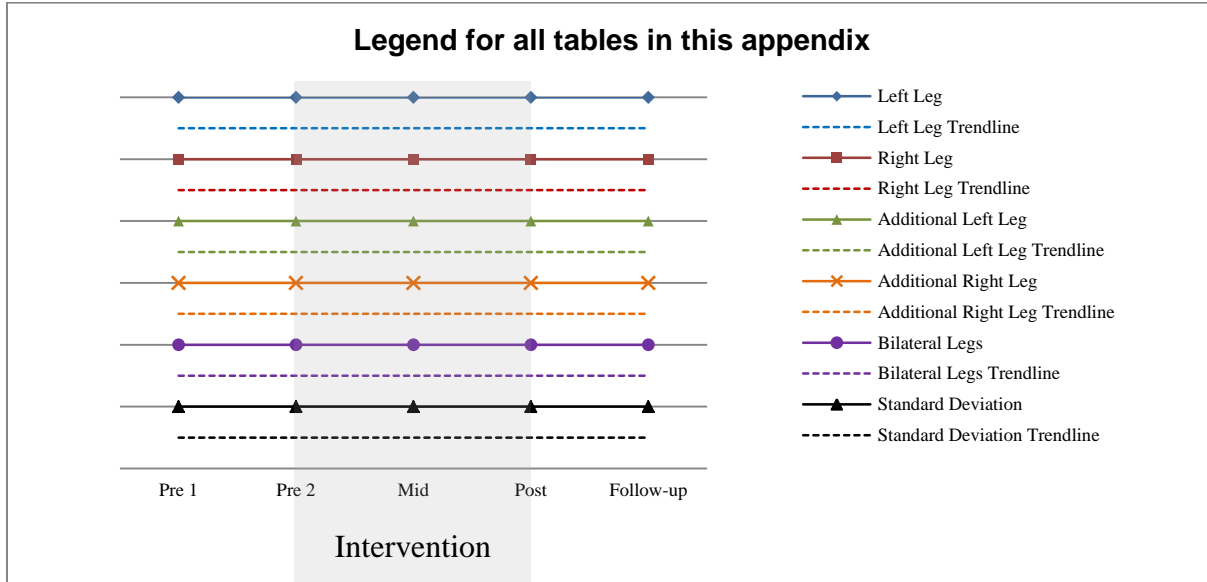
Where did any near falls occur?      \_\_\_\_\_ inside      \_\_\_\_\_ outside      \_\_\_\_\_ both

Where did the most near falls occur (if any)?      \_\_\_\_\_ inside      \_\_\_\_\_ outside

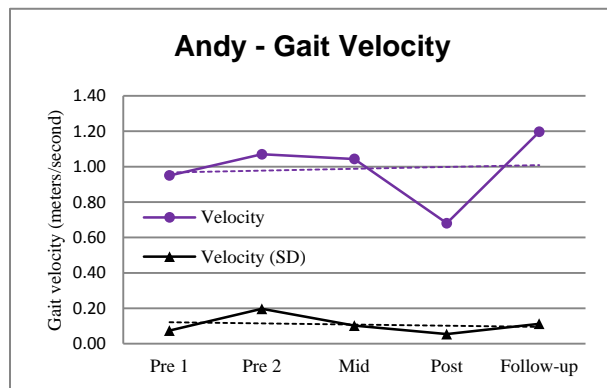
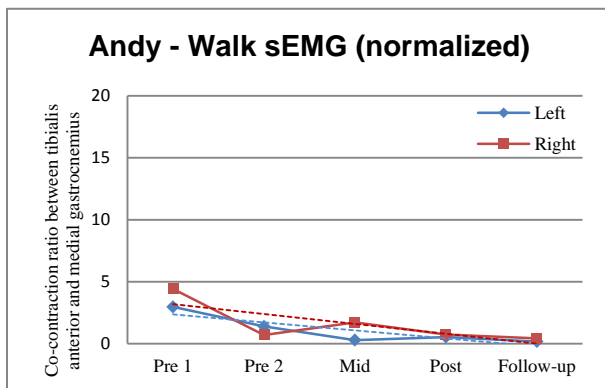
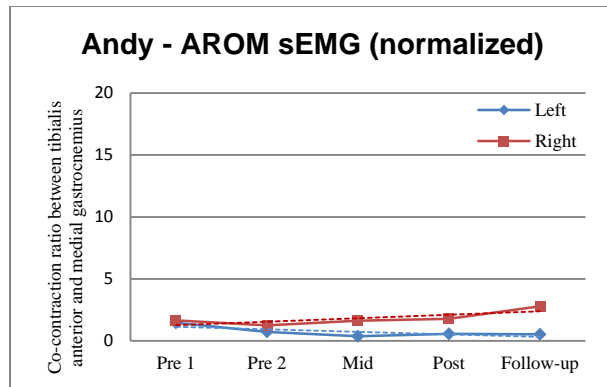
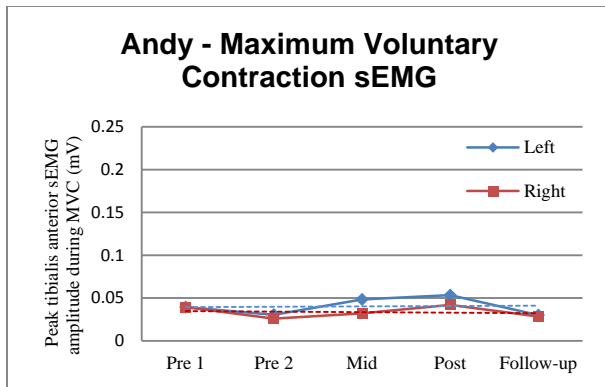
On what surface(s) did near falls occur? \_\_\_\_\_

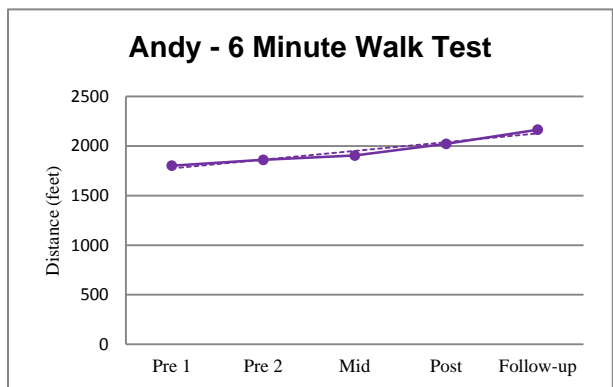
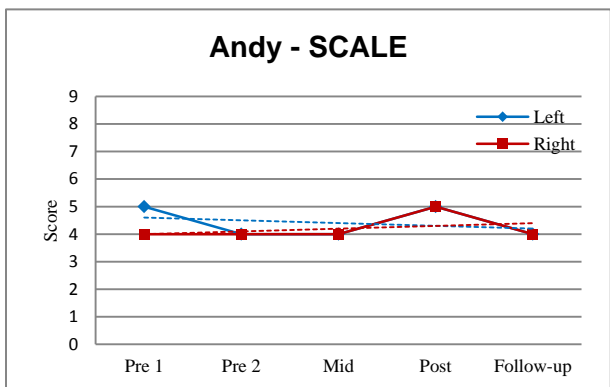
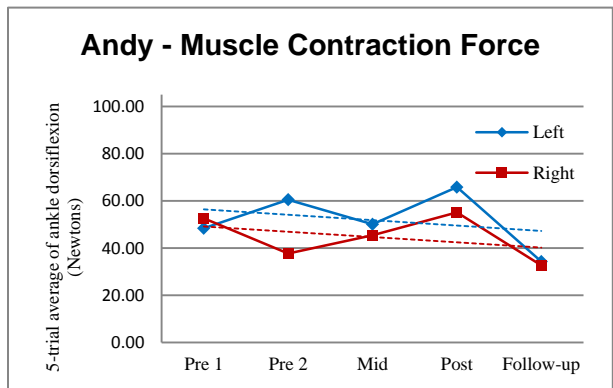
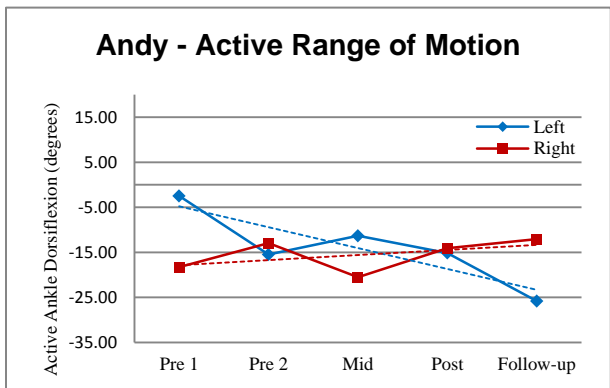
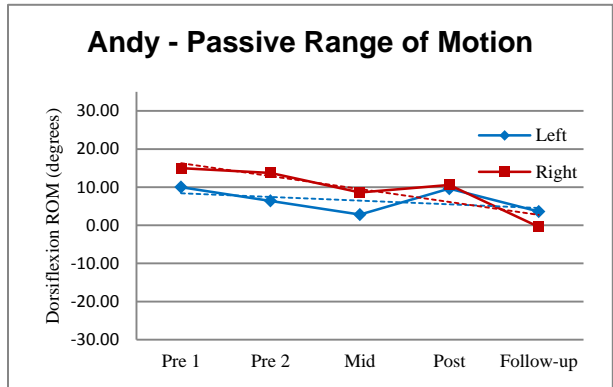
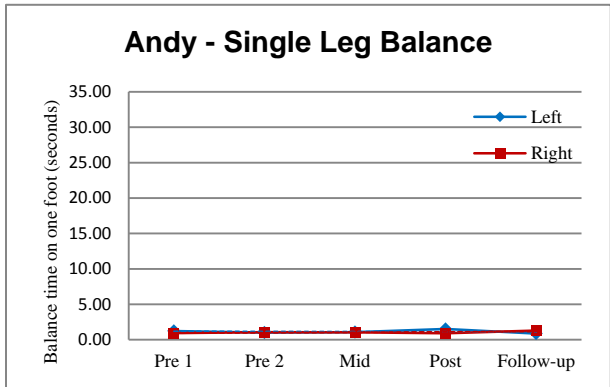
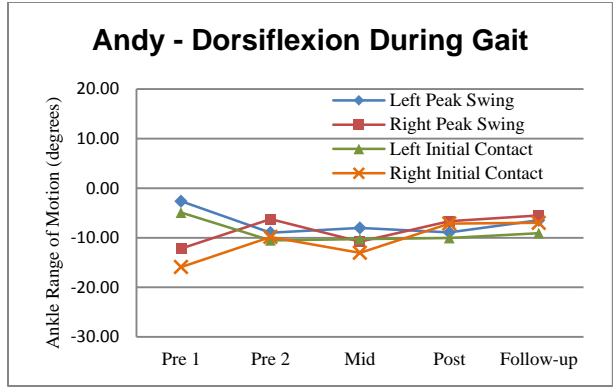
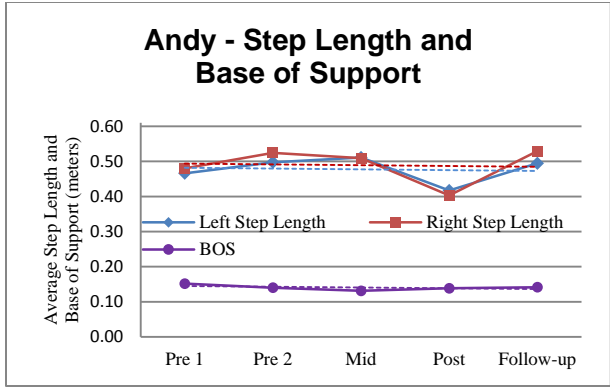
Were you wearing ankle or foot braces during any near falls?      \_\_\_\_\_yes      \_\_\_\_\_no      \_\_\_\_\_both

## Appendix D – Participant Data Graphs

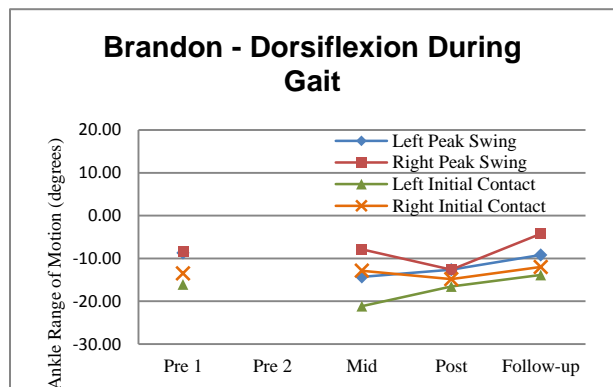
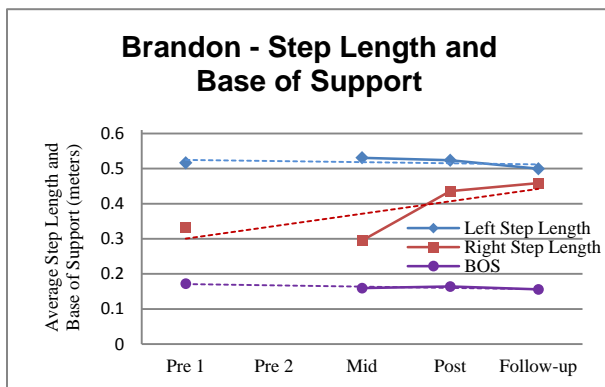
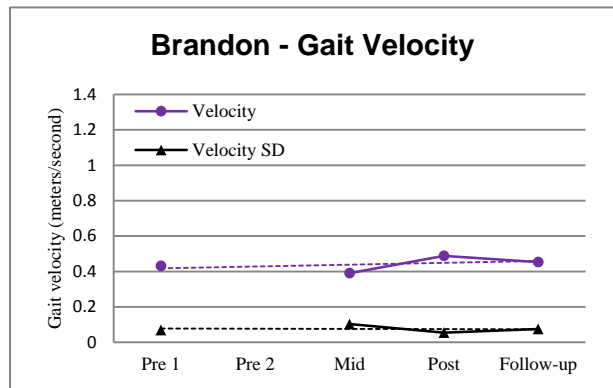
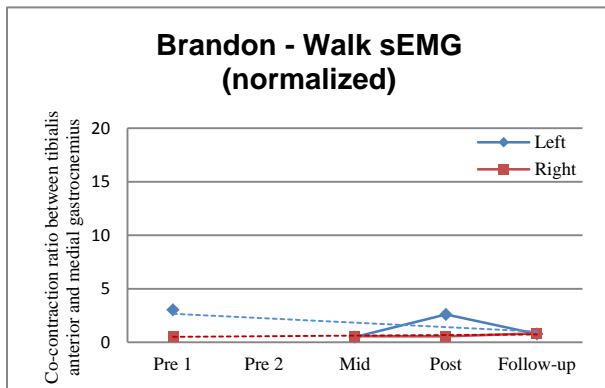
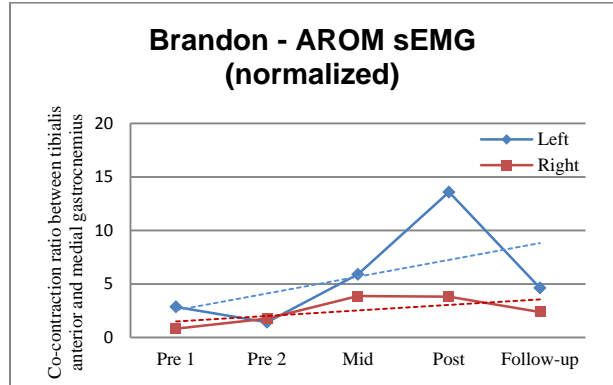
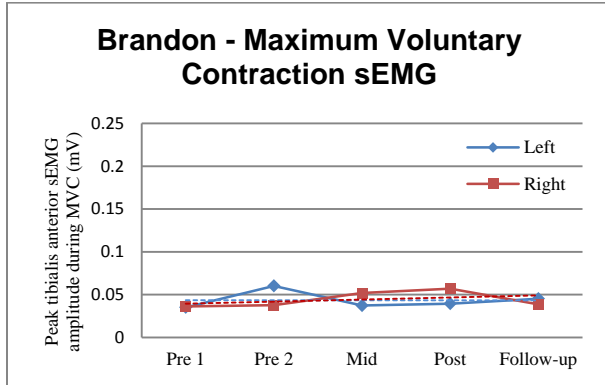


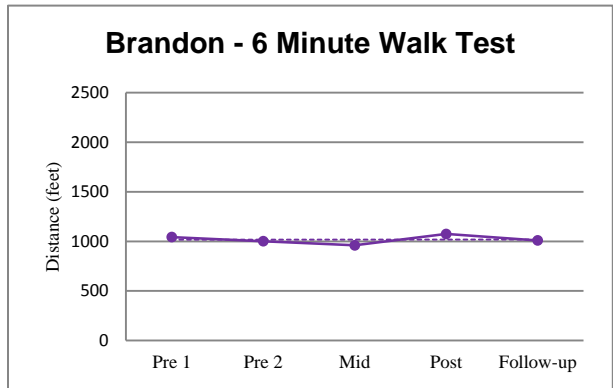
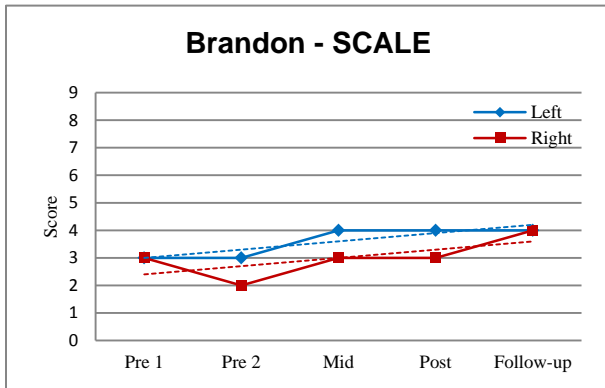
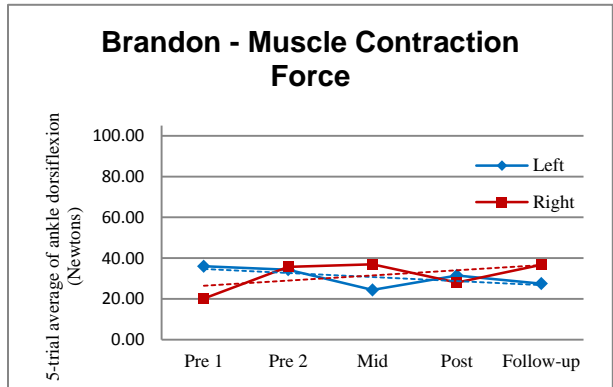
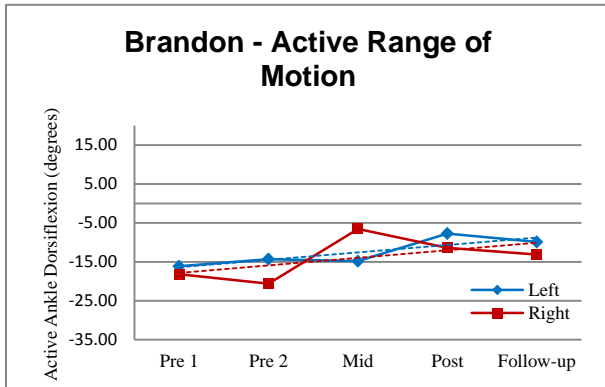
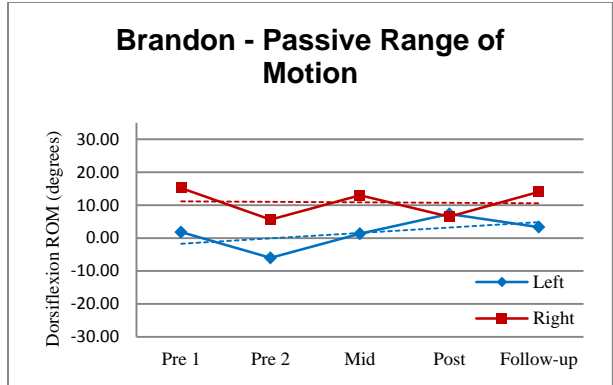
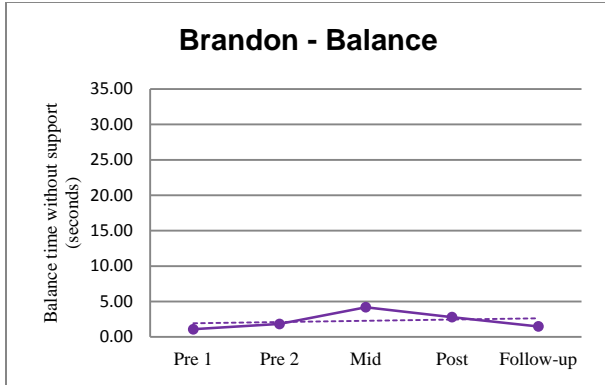
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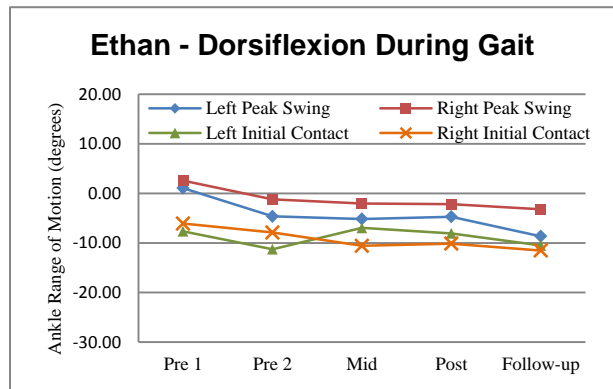
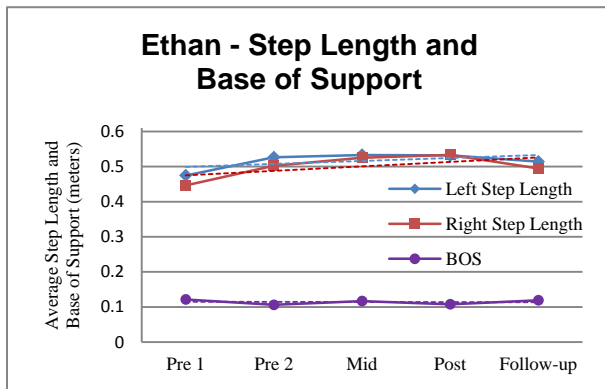
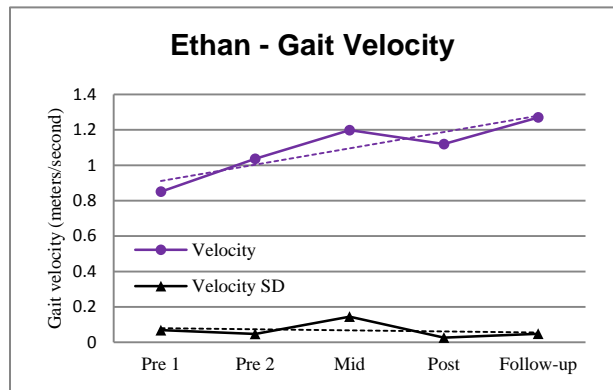
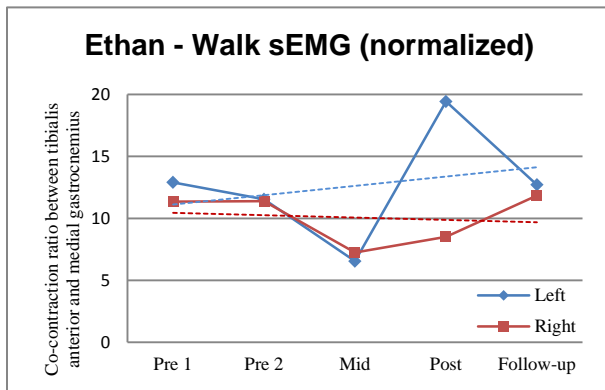
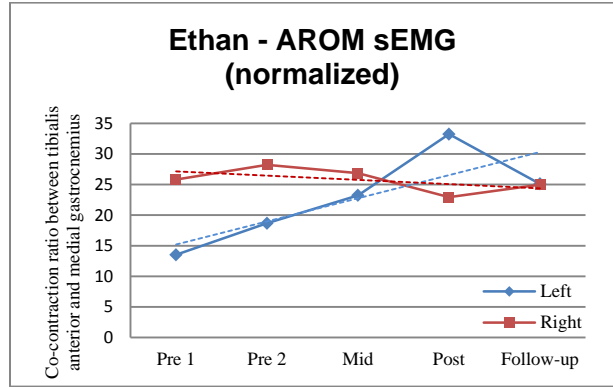
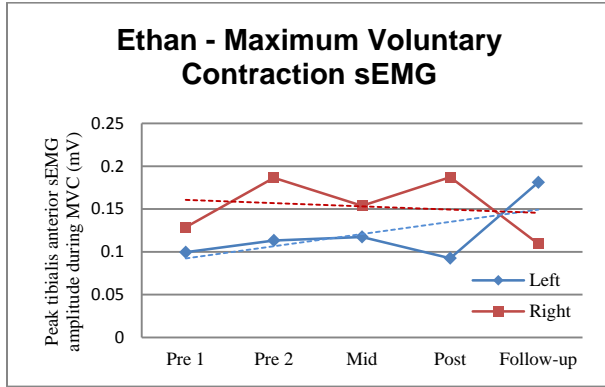


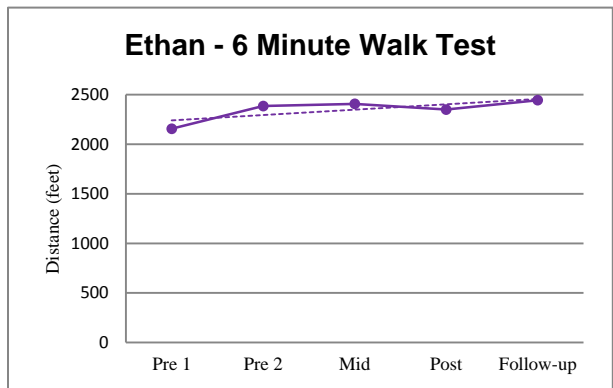
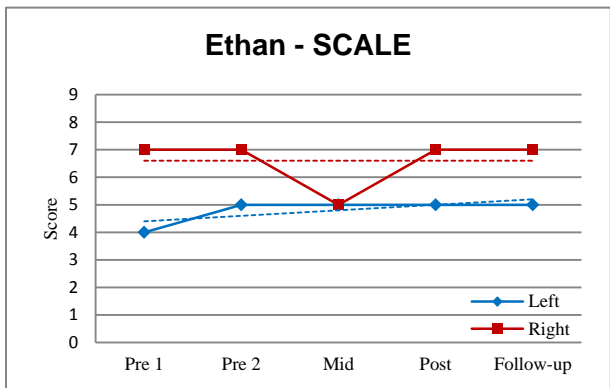
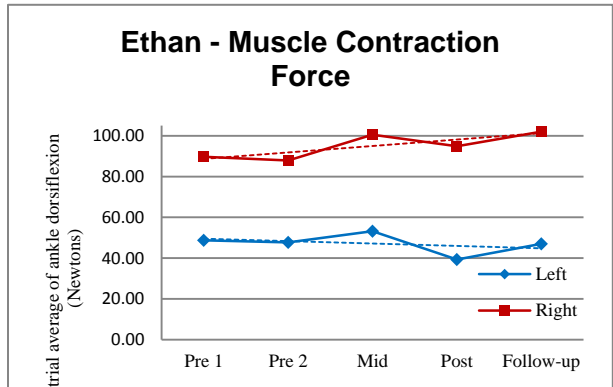
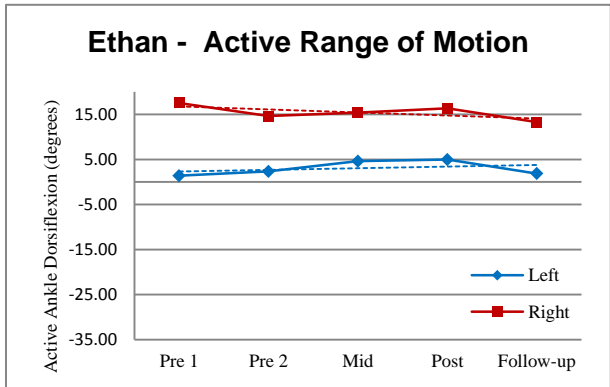
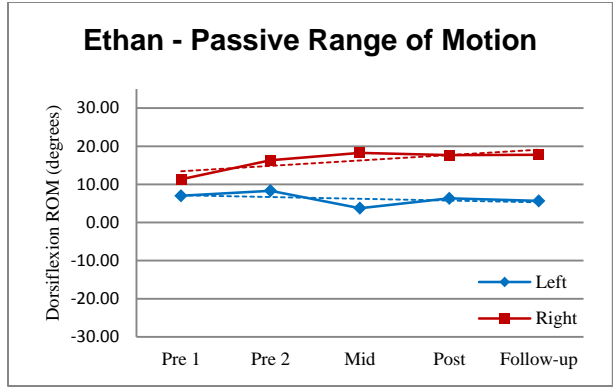
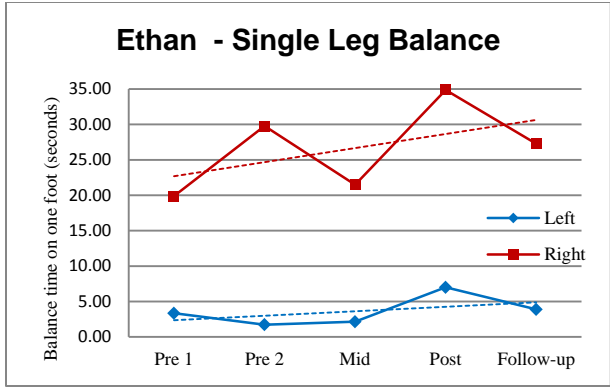
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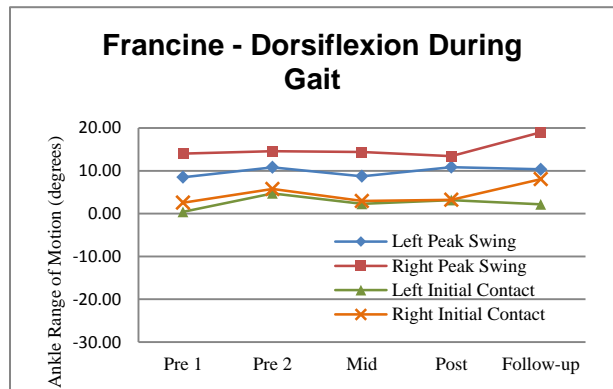
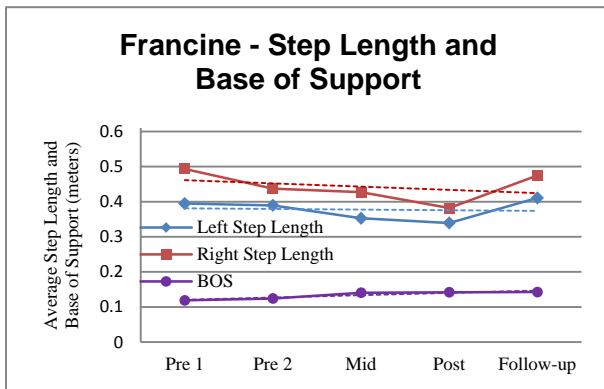
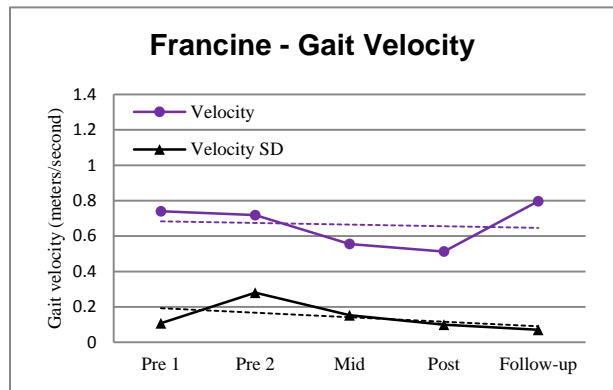
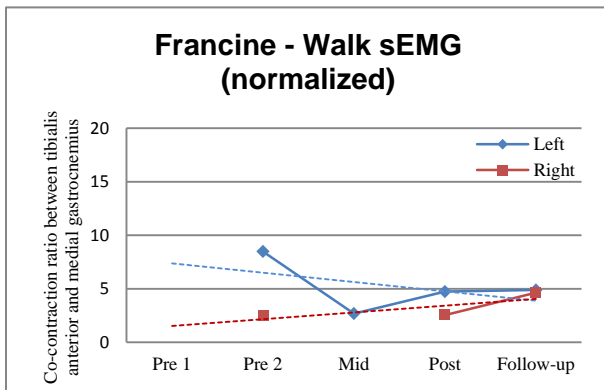
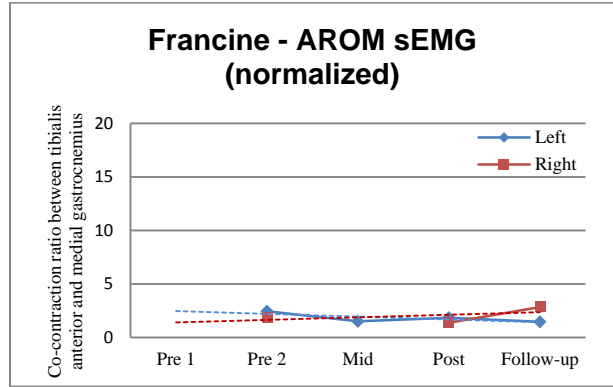
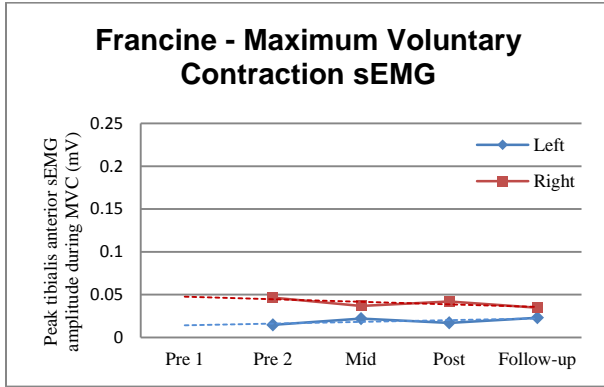


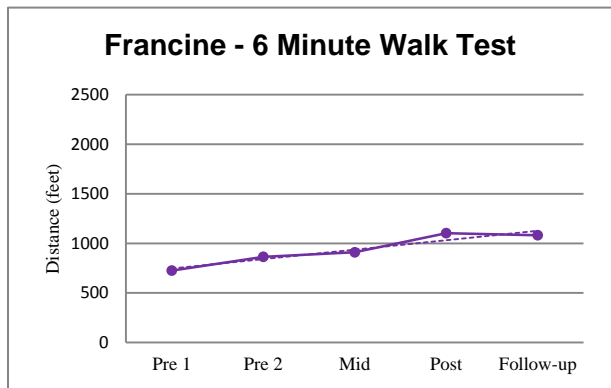
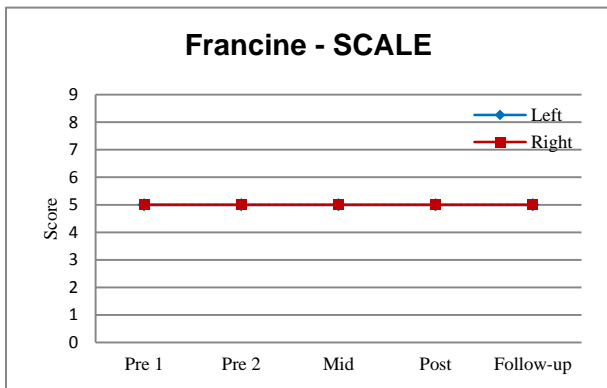
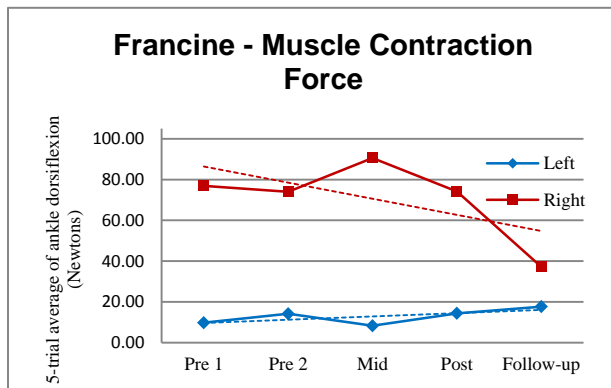
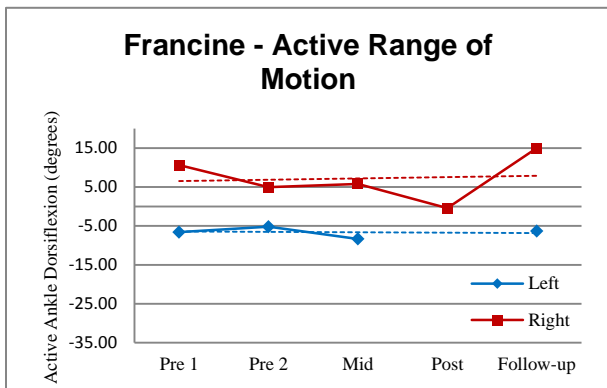
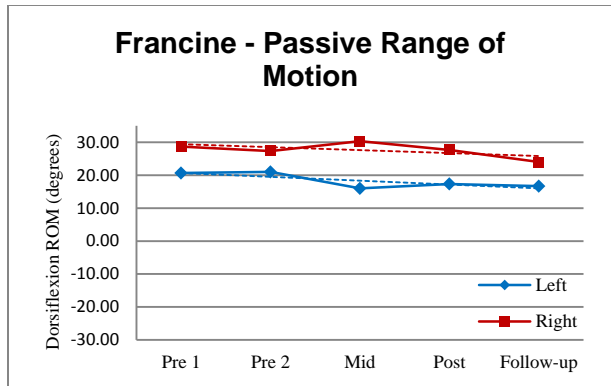
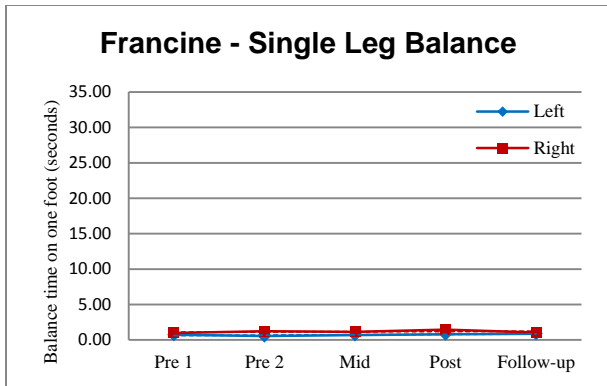
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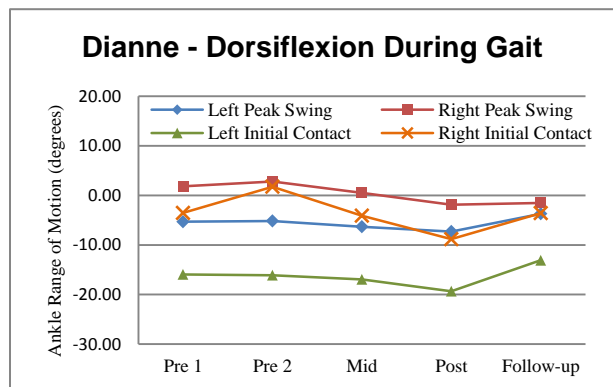
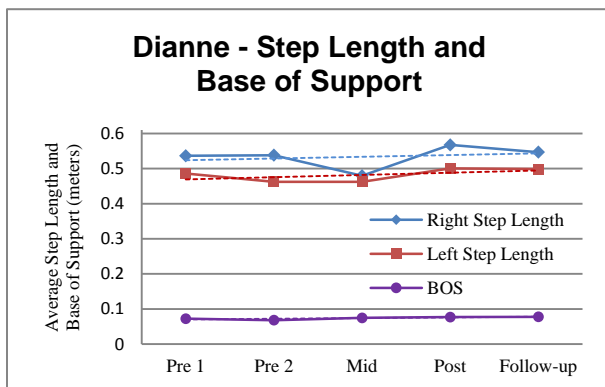
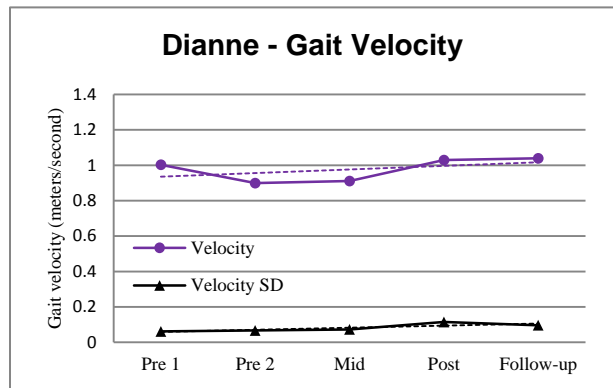
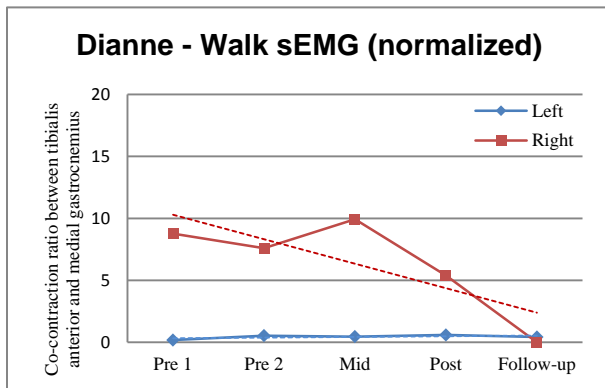
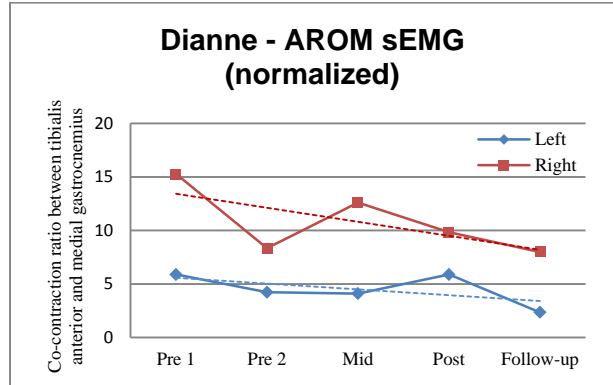
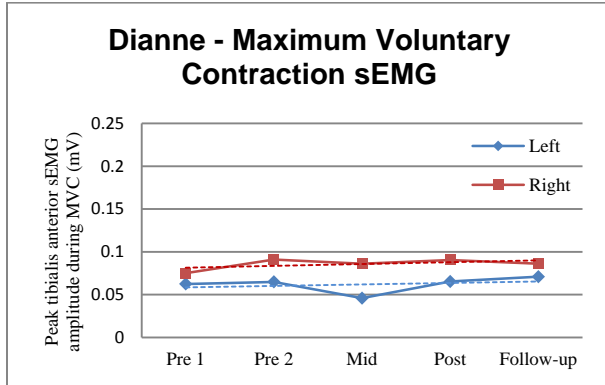


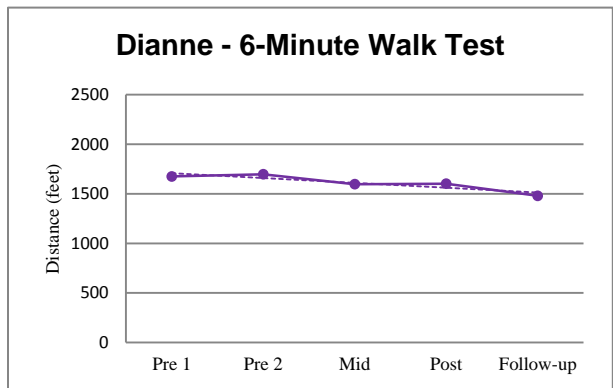
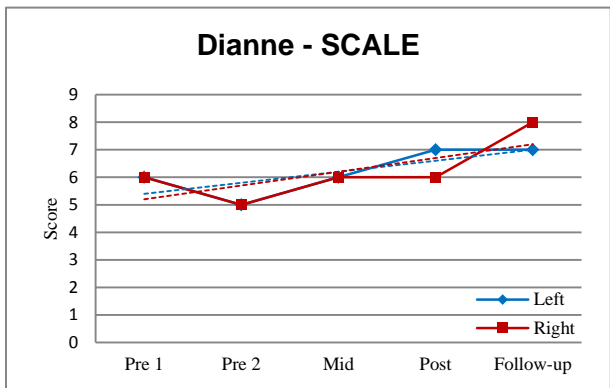
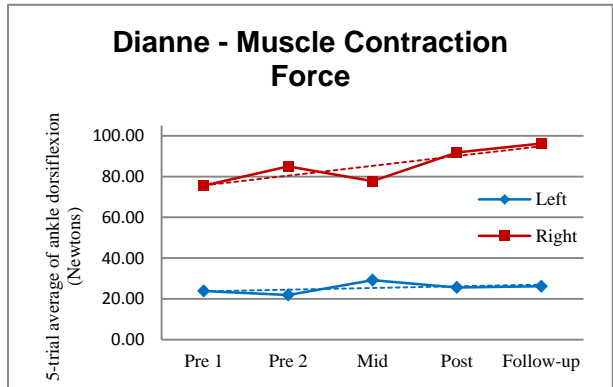
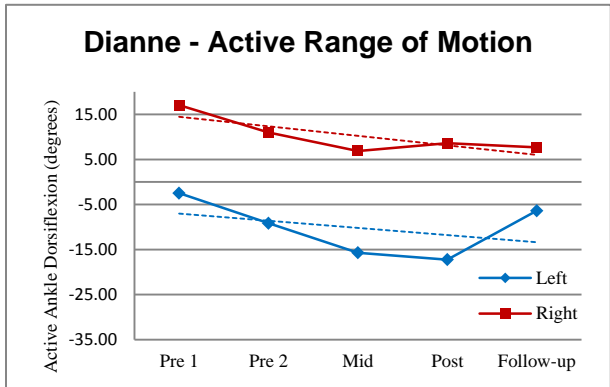
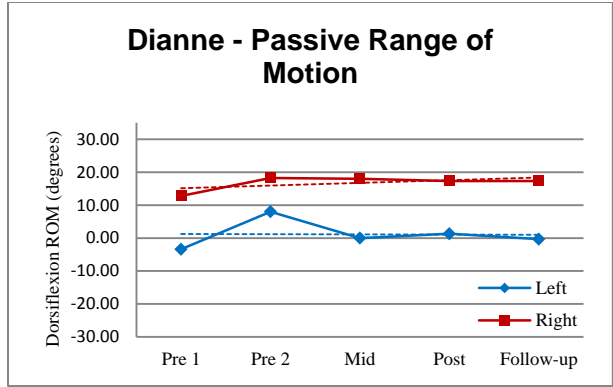
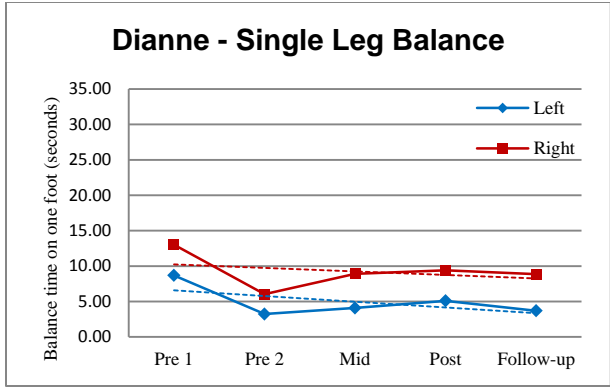
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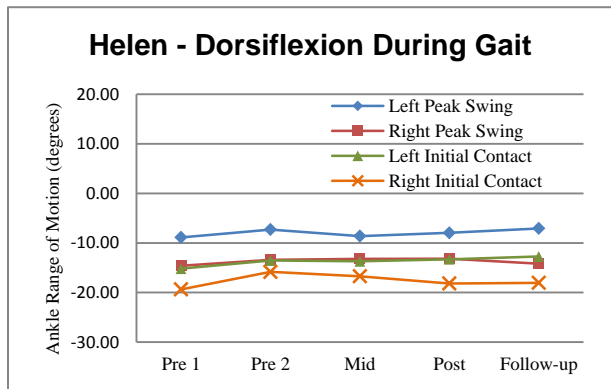
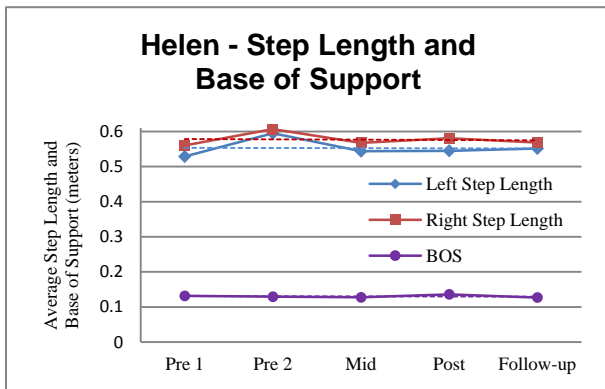
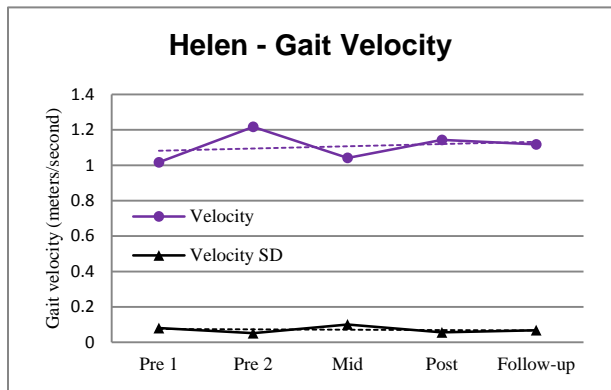
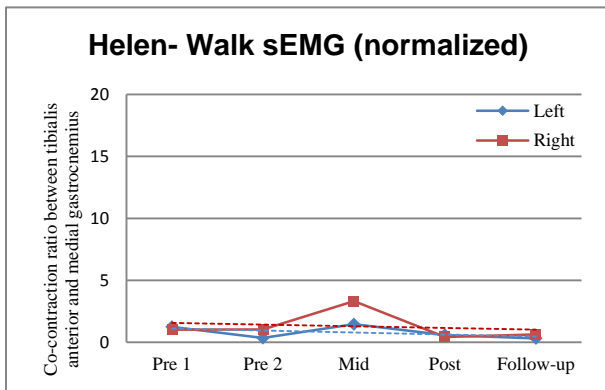
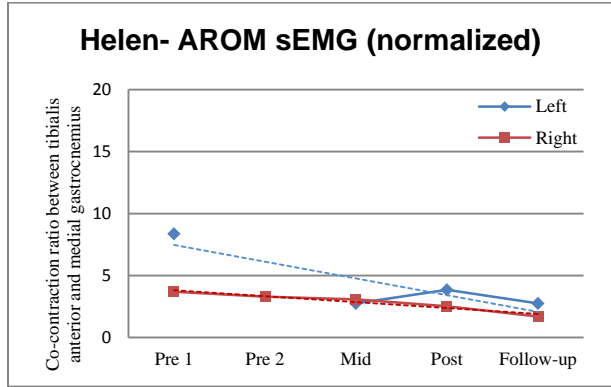
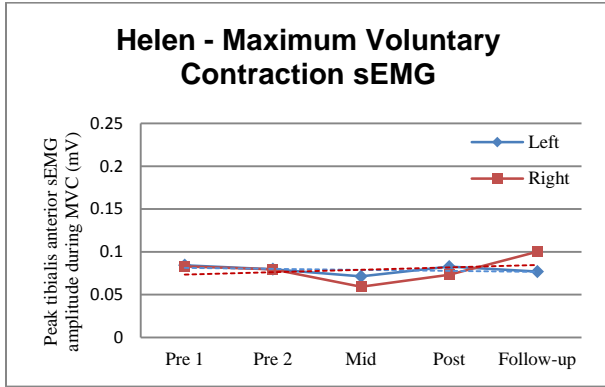


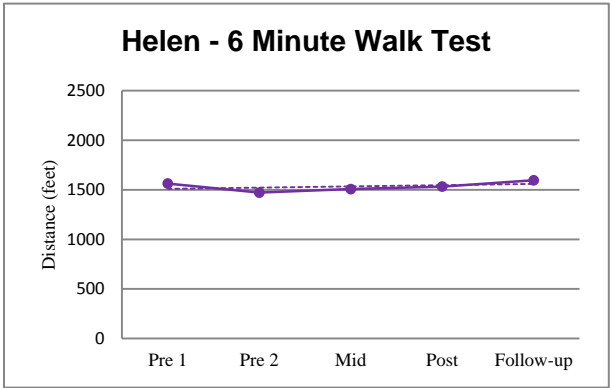
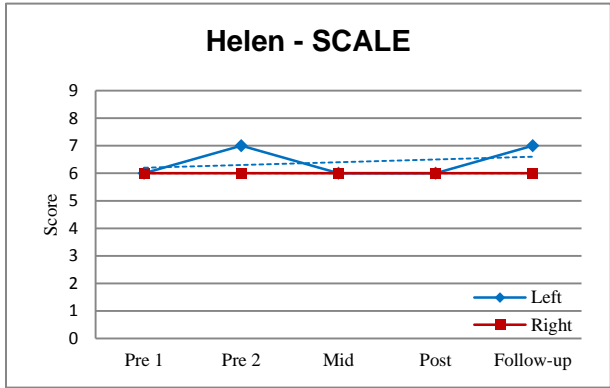
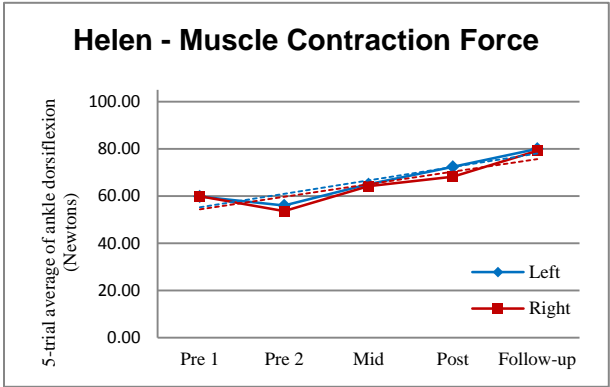
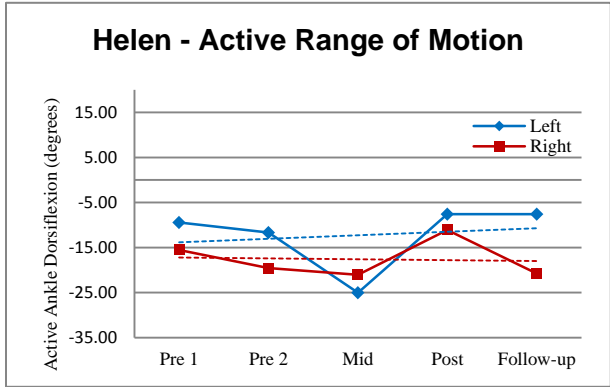
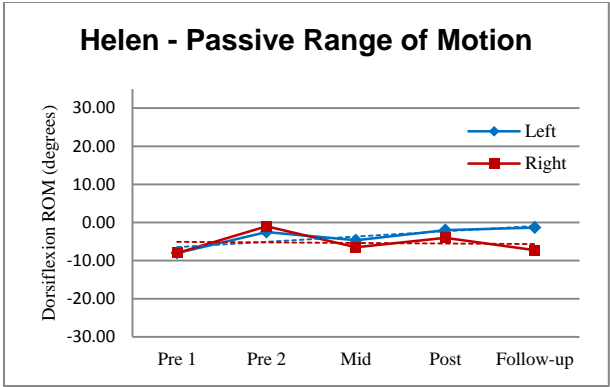
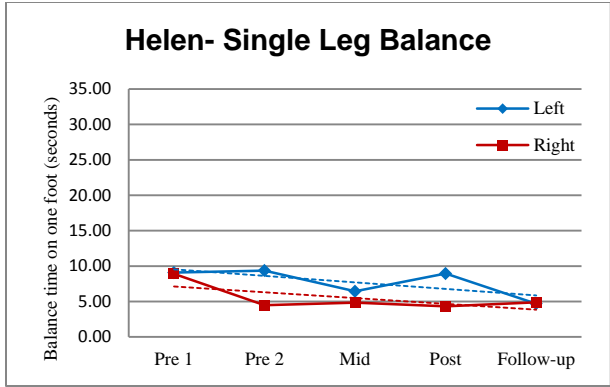
## Dianne Results



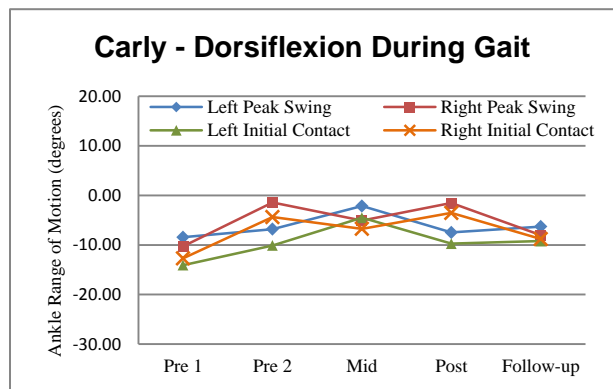
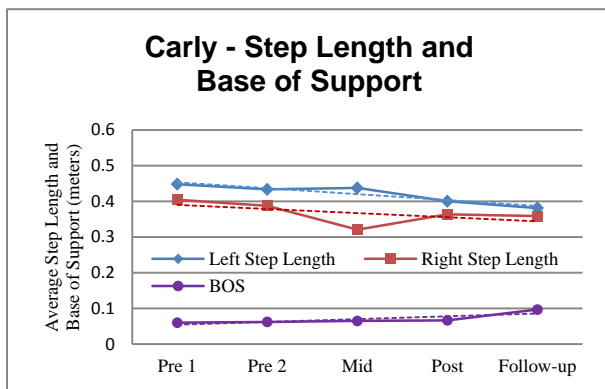
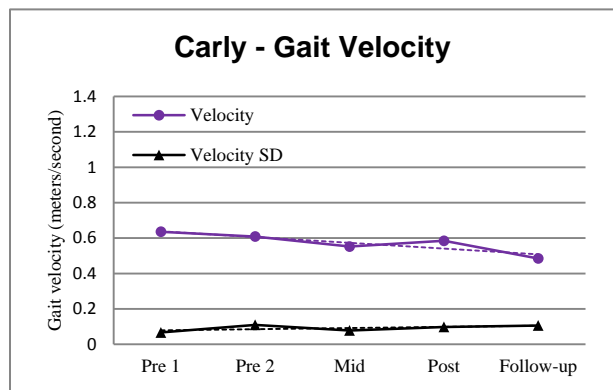
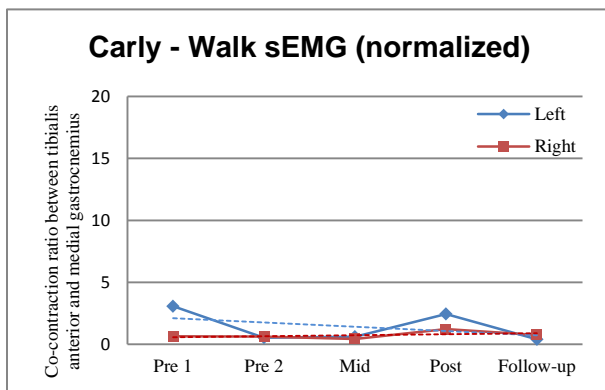
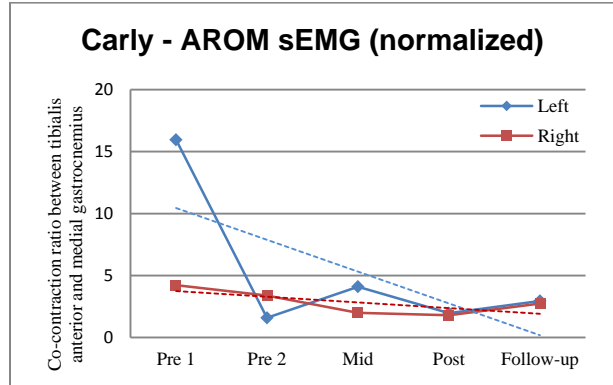
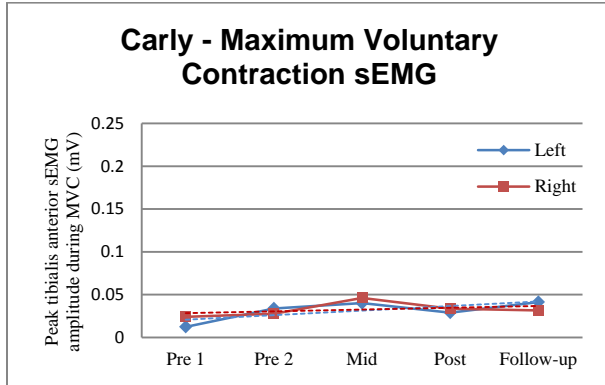


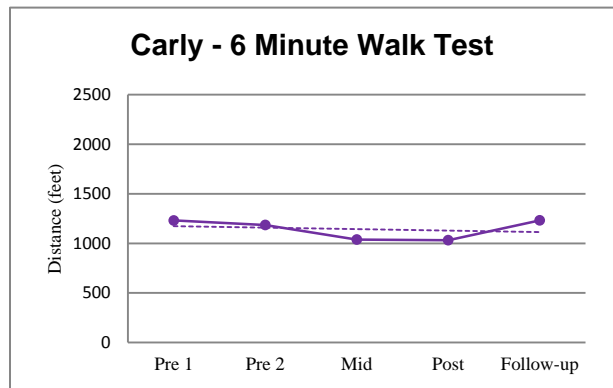
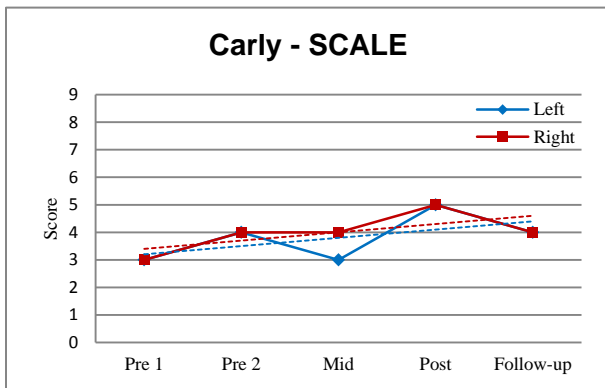
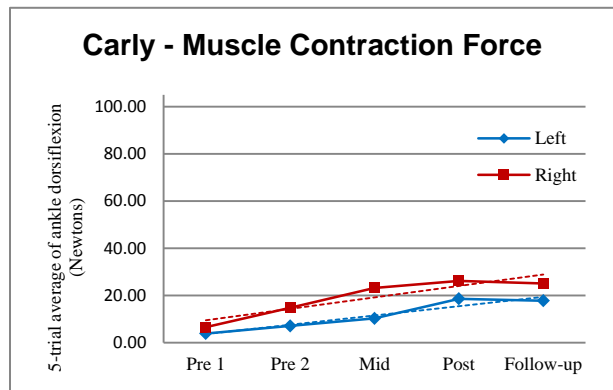
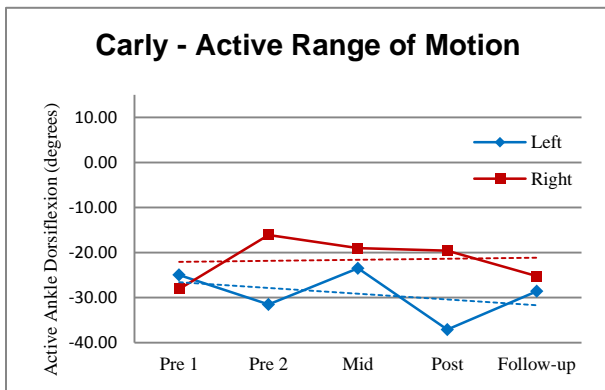
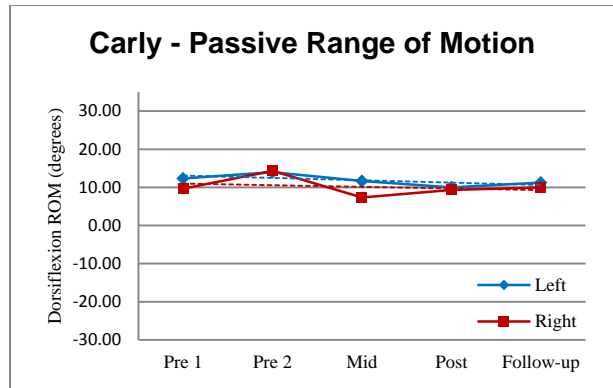
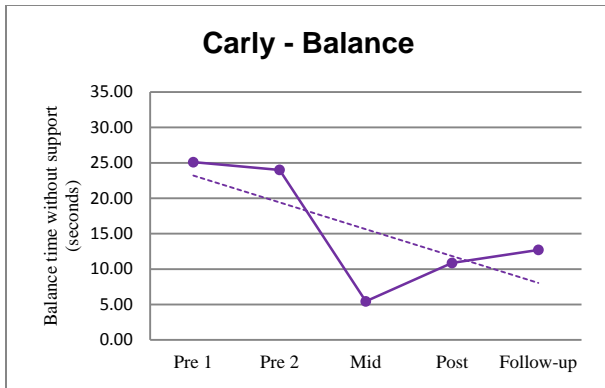
# Helen Results



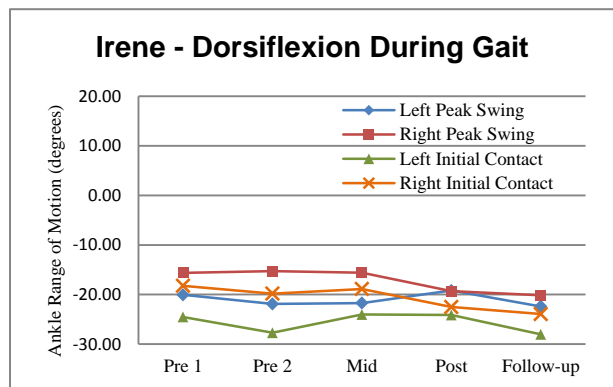
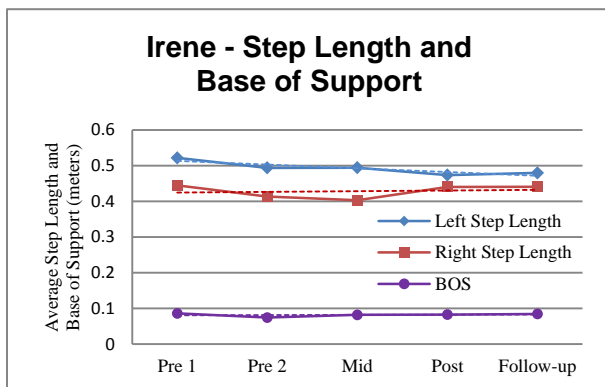
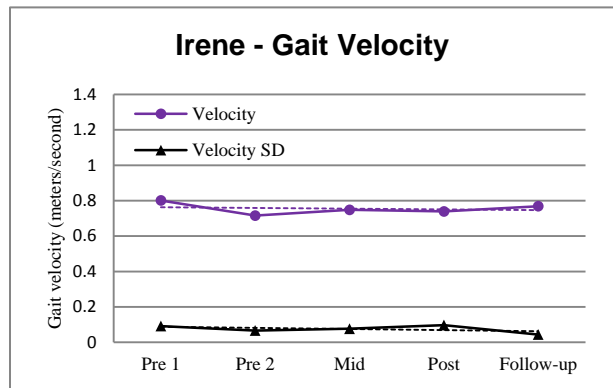
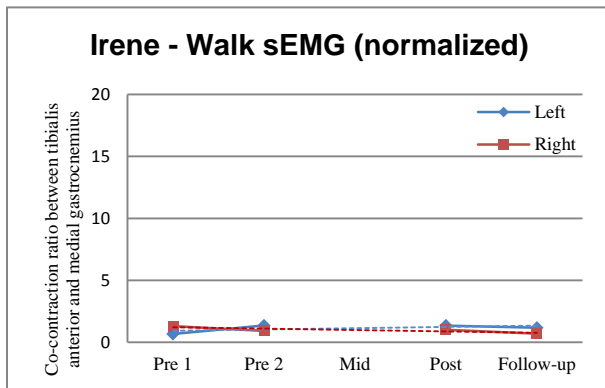
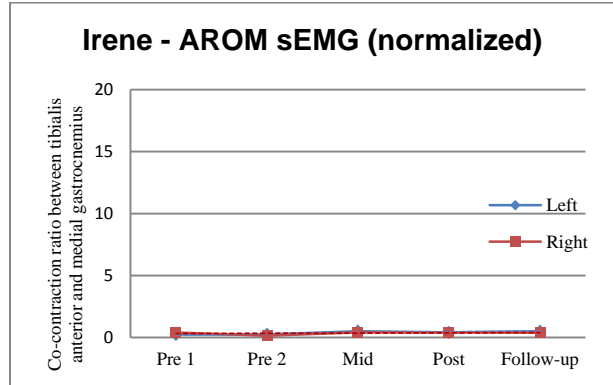
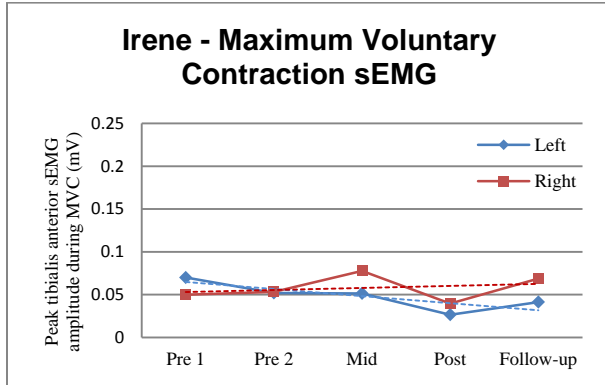


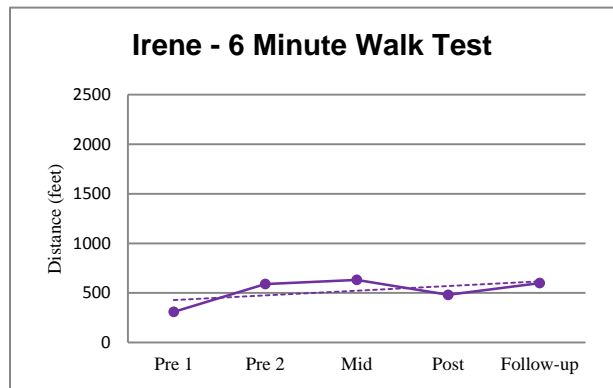
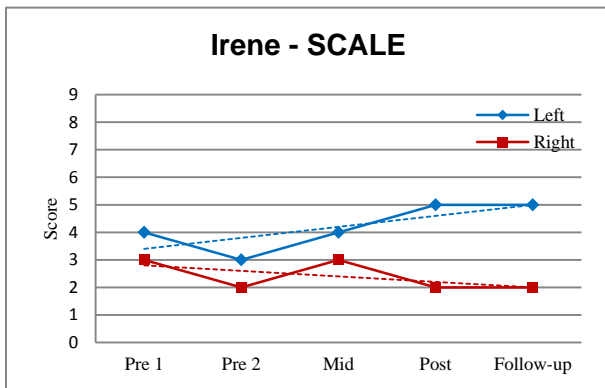
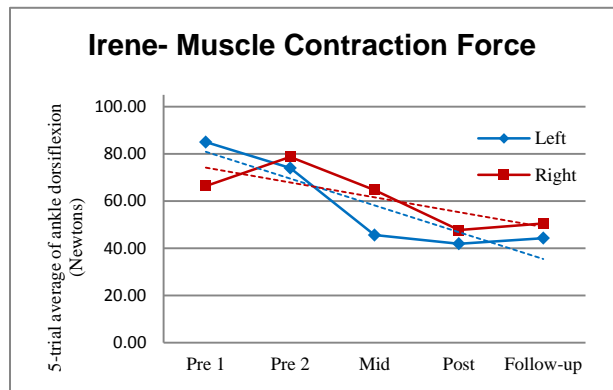
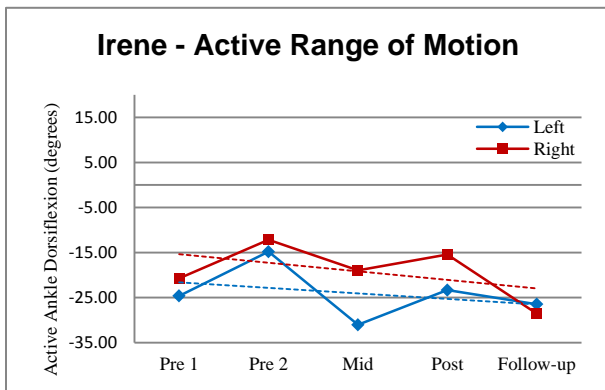
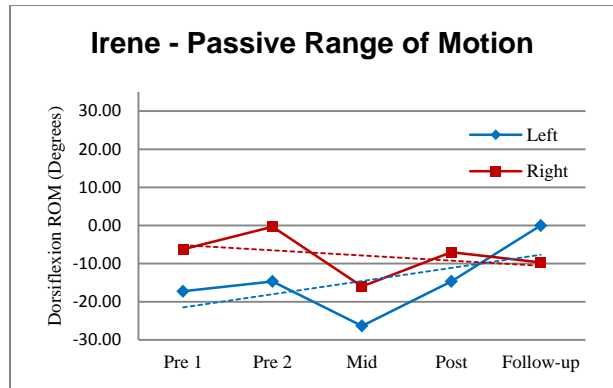
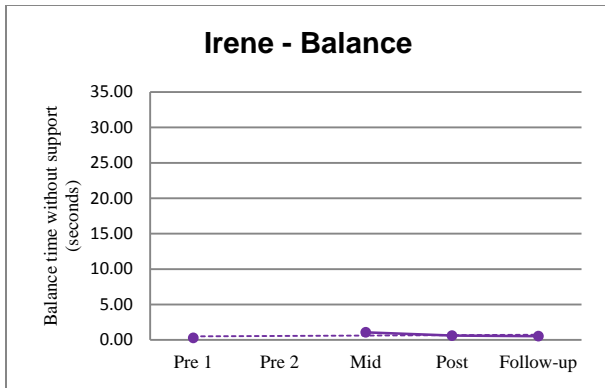
## Carly Results



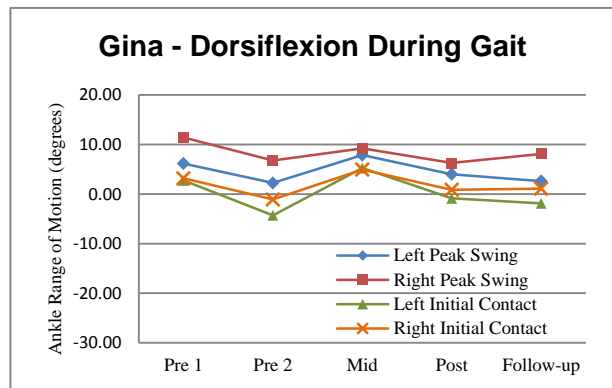
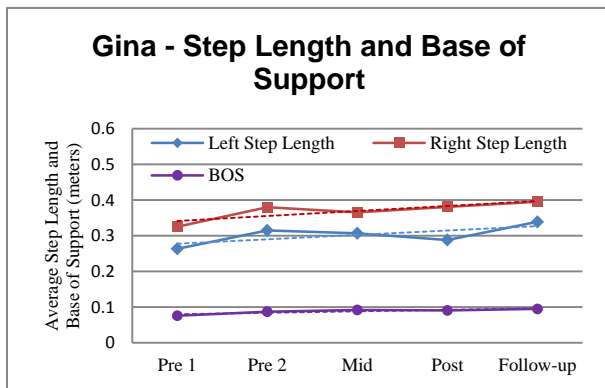
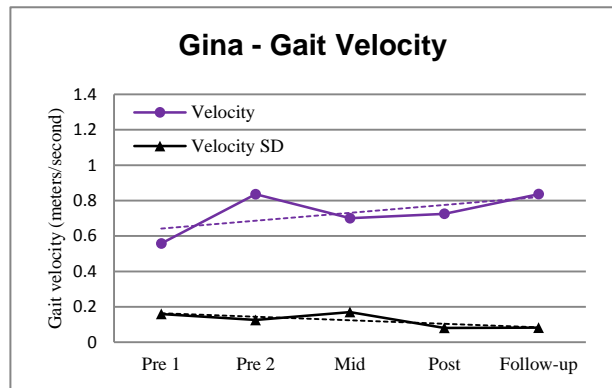
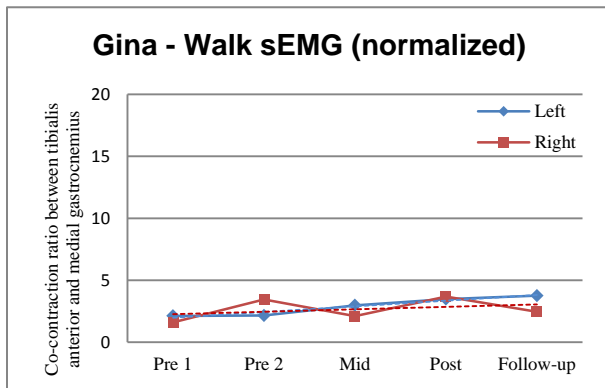
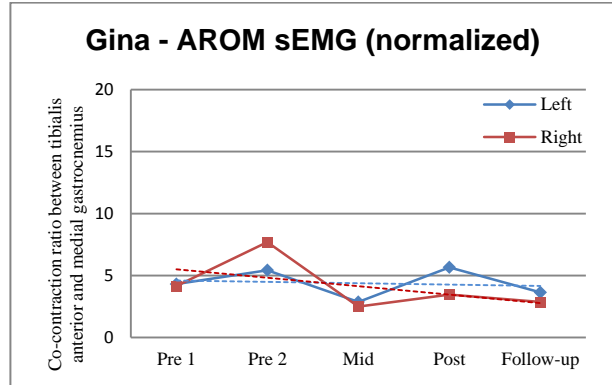
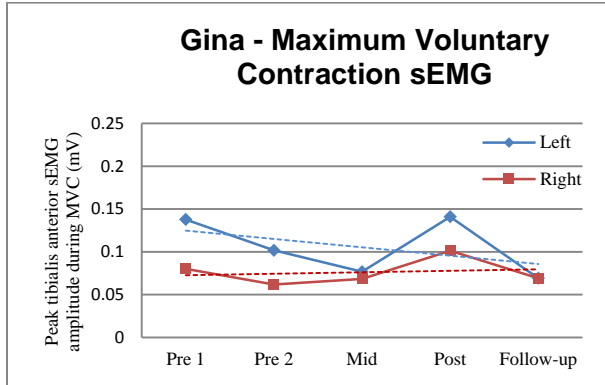


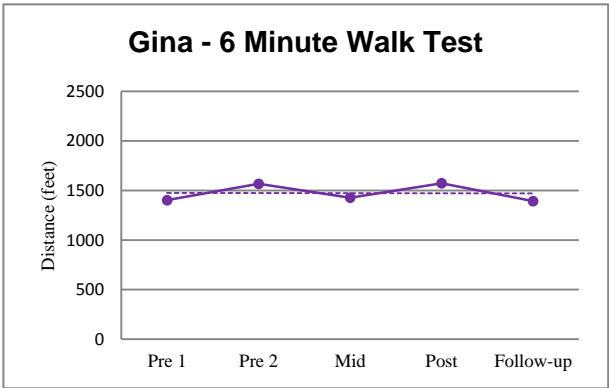
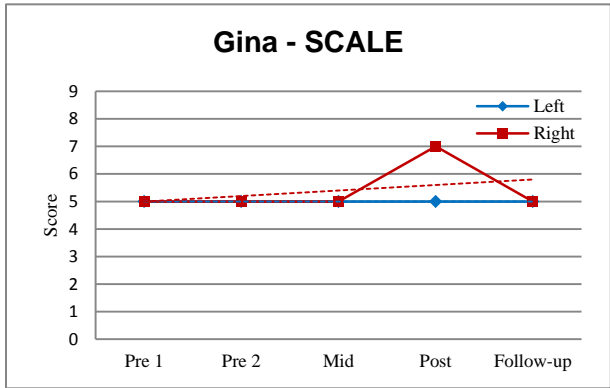
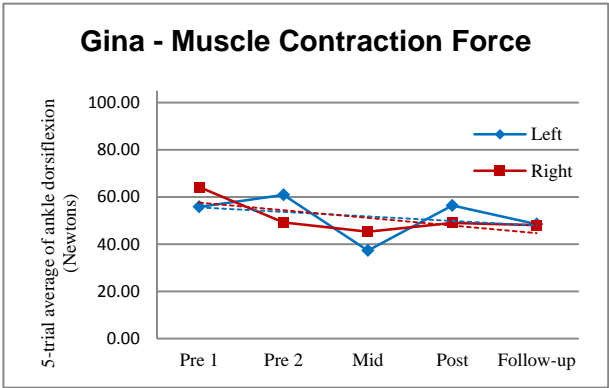
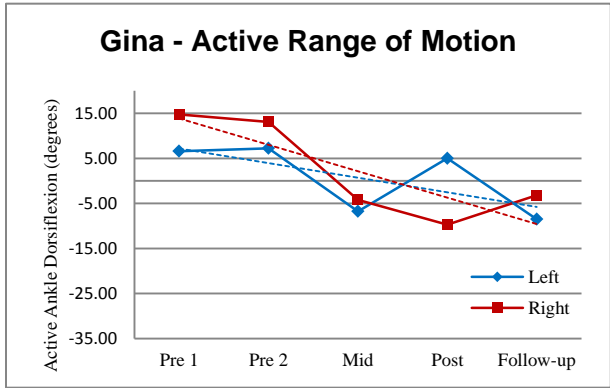
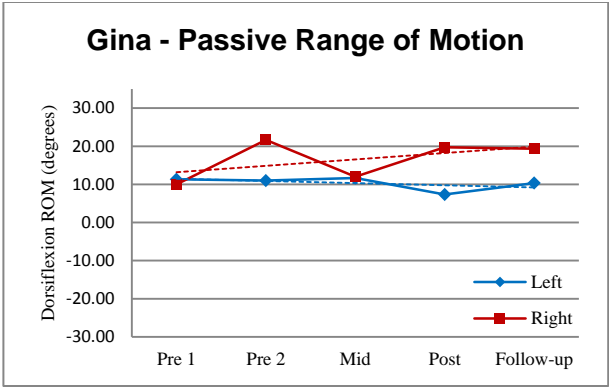
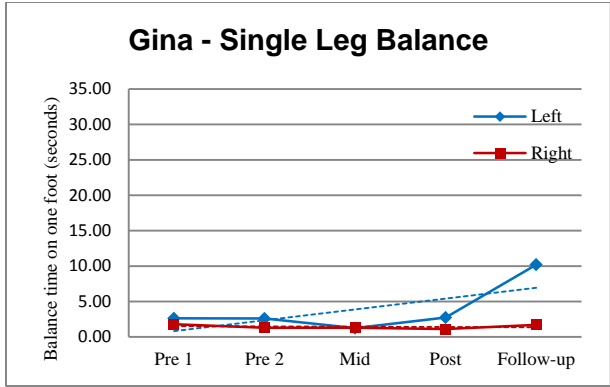
## Irene Results





## Gina Results





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