AN INVESTIGATION OF TRUNK CONTROL IN INFANTS AND TODDLERS WITH DOWN SYNDROME

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MEGAN B. FLORES, MPT

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DEDICATION

To the person who has supported and encouraged me throughout this process with unending patience, love, and humor. Thank you, Mario Flores.

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ABSTRACT

MEGAN B. FLORES

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Infants and toddlers with Down syndrome (DS) display gross motor developmental delays and decreased trunk control. Trunk control is a precursor to upright gross motor skills, but this relationship has not been fully explored in infants and toddlers with DS. The Gross Motor Function Measure (GMFM) is a reliable and valid measure of motor skills for children with DS. The Segmental Assessment of Trunk Control (SATCo) shows promise as a simple measure that can be employed by a physical therapist to measure segmental changes in trunk control, but it has not been studied in this population. The purposes of this study were: 1) to examine the reliability of the SATCo, 2) to explore the relationship of the SATCo with the GMFM, and 3) to explore the impact of a dynamic standing device (Upsee) on trunk control and motor skills in infants and toddlers with DS. Eighteen children with DS, ages six to 24 months, were tested once on the GMFM and twice on the SATCo by two different physical therapists. SATCo scores among live and video recorded sessions were compared between three raters. The SATCo and GMFM were compared using Spearman's rho correlation and linear regression. A subset of six children with DS participated in a A_1BA_2 withdrawal/reversal single-case

experimental design study to explore the impact of a dynamic standing device home program on trunk control and gross motor skills in infants and toddlers with DS. Interrater reliability of the SATCo was moderate to good and intrarater reliability was good to excellent. The SATCo showed a good to excellent significant correlation with the GMFM and was a significant predictor of total GMFM scores in infants and toddlers with DS. After six weeks of using the dynamic standing device as a home program, three children had significant increases in GMFM scores and three children did not show significant changes. SATCo scores did not change significantly in any of the children and no adverse effects were reported. For infants and toddlers with DS, the SATCo is a reliable and valid tool and a home program using a dynamic standing device may improve gross motor skills.

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CHAPTER I

PROSPECTUS

INTRODUCTION

Down syndrome (DS) is a common cause of developmental disability, occurring in approximately 12.6 per 10,000 live births in the United States.[1] Typically developing infants are able to sit without support at approximately six months of age, and walk independently by 12 to 15 months of age. Infants with DS show gross motor skill delay, attaining independent sitting at around 11 months and independent walking at approximately 24 months.[1-4] Trunk control is a precursor to upright gross motor skills,[5] therefore, assessing trunk control in infants and toddlers with DS is essential when applying interventions for improving the acquisition of gross motor skills.

Physical therapists must employ effective tools to measure the incremental changes that occur in infants and toddlers with DS, both at the body structure and function level and the activity level of the World Health Organization's *International Classification of Functioning, Disability and Health* (ICF).[6] The Gross Motor Function Measure (GMFM) has proven to be a reliable and valid measure of motor skills at the activity level of the ICF for children with DS.[7-9] However, there are limited sound outcome measures of trunk control at the body structure and function level of the ICF for infants and toddlers with DS. The Segmental Assessment of Trunk Control (SATCo)

shows promise as a simple measure that can be employed by a physical therapist in any setting to measure segmental changes in trunk control.[10] Although the original sample for the reliability and validity study of the SATCo included children with cerebral palsy (CP) in Gross Motor Function Classification Scales (GMFCS) levels I through V, as well as a few other diagnoses, none of the children had a diagnosis of DS.[10] Hansen et al[11] examined the reliability of the SATCo in children with CP in GMFCS levels I through V (intrarater interday, interrater interday and live versus video reliability). Cardoso de Sa et al[12] studied the interrater reliability of the SATCo for children with Duchenne muscular dystrophy. Both articles found the SATCo to be a reliable outcome measure for these populations.[11,12] Additionally, Curtis et al[13] examined the predictive effect of SATCo and age on GMFM score in children with CP, finding that both were significant predictors of gross motor function. To date, the psychometric properties of the SATCo have not been analyzed in children with DS.

In addition to assessing trunk control in children with DS, intervention strategies for improving trunk control in this population should be investigated. The health care costs for infants and toddlers with DS have been estimated at 12 to 13 times higher than for children without DS, emphasizing the need for early, effective intervention.[14] Ulrich called for the identification of effective home programs to supplement physical therapy as a financially feasible option for "generating sufficient therapy that adheres to principles for optimizing development of neuromotor control."[15]

To address this issue, a home program intervention using upright mobility through treadmill training proves effective for accelerating the acquisition of motor skills in

infants with DS.[2,16] Studies published by Ulrich et al[2,16] and Wu et al,[17] determined that in-home use of treadmills for infants with DS is feasible. Furthermore, infants with DS who participated in the treadmill training home program were able to walk earlier than they typically would when compared to infants with DS who did not participate in the program.[2,16,17] However, pediatric treadmills are expensive. Alternative forms of upright mobility that are more affordable and less cumbersome are needed. The Upsee* shows promise as a dynamic standing device that can be used at home by the parent and child to promote upright mobility. This device uses a child harness connected to an adult hip belt with shared sandals for promoting upright activity in young children. In a case series by Ardolino et al,[18] dynamic upright mobility through use of the Upsee was found to improve trunk control and gross motor skills in children with trunk hypotonia. Casey et al[19] found the Upsee was a feasible means to promote physical activity and social engagement in a young child with Pitt Hopkins syndrome. The evidence suggests that this dynamic standing device may lead to improved trunk control, which is an important factor for mobility.[5] The impact of dynamic standing through use of the Upsee has not been studied in infants and toddlers with DS.

PURPOSES AND HYPOTHESES

Overall, the purpose of this dissertation was to investigate trunk control in infants and toddlers with DS. The purpose of the first study was to determine the reliability of the SATCo in infants and toddlers with DS. The hypothesis was that the SATCo will demonstrate good ($\kappa > 0.8$) interrater and intrarater reliability. The purpose of the second

* Firefly by Leckey, Lisburn, Northern Ireland

study was to examine the validity of the SATCo with the GMFM and determine whether age and SATCo score predicts GMFM score in infants and toddlers with DS. The hypotheses were that the SATCo will show concurrent validity with the GMFM ($r < 0.7$), and both age and SATCo scores will be significant predictors of GMFM scores ($R^2 > 0.4$, $p < 0.05$). The purpose of the third study was to explore the impact of a dynamic standing device (Upsee) on trunk control and motor skills in infants and toddlers with DS. The hypothesis was that participants will demonstrate a greater change in scores on the SATCo and the GMFM during the intervention phase than during the baseline phases (at least two data points outside the 2-standard deviation band).

PARTICIPANTS

To obtain a kappa statistic of 0.8 at 90% power, a sample size of 17 participants was required to detect statistical significance at $p \leq 0.05$ for reliability studies.[20] A priori power analysis using G*Power[†] for a Pearson correlation test with a large effect size of $\rho = 0.7$, power = 0.8 and $p = 0.05$ reveals that a sample size of 11 participants was required. For linear regression with an effect size of 0.7, power = 0.8, and $p = 0.05$, a sample size of 18 participants was required. Therefore, for Studies One and Two, a sample of 20 children with DS were recruited in case of attrition. The age before children with DS develop the ability to sit independently to an age well after they can maintain independent sitting balance will capture the full range of the SATCo scale. Children were recruited from local pediatric clinics and DS parent support groups. For inclusion in Studies One and Two, participants must speak English, have a diagnosis of

[†] G*Power Version 3.1.9.2, Franz Faul, Universitat Kiel, Germany

DS, and be between the ages of six to 24 months. Participants were excluded from the study if they have a diagnosis unrelated to DS that limits gross motor movement, or medical restrictions that contraindicate movement or handling.

Study Three will have slightly different participant criteria. Recruiting infants and toddlers with DS between the ages of nine to 18 months old presents challenges for a traditional randomized controlled study; therefore, participants (n = 5 to 10) will serve as their own control in an A_1BA_2 single-case experimental design. Single-case designs use purposeful manipulation of independent variables across multiple phases, with frequent measurement of dependent variables, making this design a rigorous alternative to a large clinical trial.[21,22] Participants from Studies One and Two were offered the opportunity to also participate in Study Three. In addition to meeting the inclusion and exclusion criteria for Studies One and Two, participants were included if they: 1) are between the ages of nine to 18 months; 2) are able to bear weight through lower extremities when supported; and 3) display evidence of gross motor developmental delay and are not independently walking as measured by the GMFM from Study One. Children were excluded from Study Three if they had: 1) severe hypertonicity in the lower extremities, 2) a history of multiple lower limb fractures, 3) severe lower extremity asymmetry including hip dislocation, or 4) an unstable medical condition requiring rapid repositioning for treatment (such as epilepsy or breathing difficulties). Additionally, the adult using the device cannot be pregnant or suffering from back pain, mobility, or balance problems. The child and adult must both be able to stand upright in the device with the child's

shoulders below the level of the adult's navel. Participants were screened prior to beginning the study.

INSTRUMENTATION

The SATCo and the GMFM are the primary outcome measures for all three studies. If the SATCo is found to be unreliable or not valid, the GMFM would be the primary outcome measure for Study Three. The SATCo uses a dichotomous scale to assess discrete levels of trunk control in children with motor disabilities.[10] The child is seated on a bench with his or her feet on the ground and the pelvis held in a neutral position. The tester gives manual horizontal support at the trunk, starting at the shoulders and moving segmentally down the trunk through seven levels. The child is tested on static, active, and reactive trunk control at each level, for a total of 20 items. The child is given a score of "present" or "absent" control for each item. Scores range from 0 to 20 for the items, and 0 to 7 for the level of trunk control. In the original sample of typical children and children with CP, intrarater reliability for the SATCo was high, (ICC = 0.98) as was interrater reliability (ICC = 0.84).[10] Concurrent validity was established with GMFM Dimension B (r = 0.73 to 0.83) and with Alberta Infant Motor Scale (AIMS) sitting (r = 0.86 to 0.88).[10]

The GMFM is a measure of gross motor function for children with CP that has also been shown to be reliable and valid for children with DS under six years old.[7,23,24] The examiner scores a child's capabilities across five dimensions of functional movement: A) Lying and Rolling, B) Sitting, C) Crawling and Kneeling, D) Standing, and E) Walking, Running, and Jumping. Each dimension is made up of several items on

an ordinal scale. The child receives a score of 0 (does not initiate), 1 (initiates), 2 (partially completes), or 3 (completes). This outcome measure can be administered by direct observation or by parent report. For children with DS, the GMFM has strong interrater reliability (ICC = 0.96 to 0.98) and test-retest reliability (ICC = 0.95 to 0.96), with evidence of responsiveness and validity with the Motor Scale of the Bayley Scales of Infant Development.[7,9,24]

Study Three employed home use of the Upsee, a dynamic standing and walking device worn by both the child and parent. The device consists of a child harness connected to an adult hip belt. Both parent and child wear connected sandals. As the parent bears weight through his or her lower extremities, the child is encouraged to stand and take steps.

PROCEDURES

Dual Institutional Review Board approval will be obtained from Texas Woman's University and the University of St. Augustine for Health Sciences. Parental consent was also obtained prior to any data collection. Parents completed a short demographic survey about their children.

Two experienced physical therapist raters with at least three years working in pediatric physical therapy were recruited. They were given all outcome measure protocols, manuals, and supporting literature to review.[10,24] The raters met with the principal investigator for training on the outcome measures to ensure consistency of scoring. Demographic information about each physical therapist rater was obtained.

Doctor of Physical Therapy students were also recruited and trained as research assistants for the purposes of video recording and to assist the raters as needed.

Studies One and Two employed a methodological research design. Each child participant was tested on the SATCo twice and GMFM once, with at least 20 minutes between each SATCo test. The participant was tested in the same environment by a different physical therapist rater each time, within 48 hours of the first test. For studies One and Two, the child participants were recruited on an on-going basis and scheduled for two testing sessions to occur at a location of the parent's choosing (either in the home, daycare, or on campus at the University of St. Augustine for Health Sciences). At the first session, Rater One tested the child on the SATCo through all seven levels, which took about 10 minutes, and the GMFM, which took about 30 minutes. The child was video recorded during the testing session to ensure consistency of the testing environment and account for discrepancies in scoring that might occur. The second testing session occurred at least 20 minutes, but no more than 48 hours, after the first day of testing, which allowed the participant and parent some flexibility. Rater Two then tested the child on the SATCo through all seven levels. The child was again be video recorded during the testing session. To ensure consistency of the testing environment, the same research assistant, testing equipment/toys, and space was utilized. After a period of at least two weeks passed, the raters re-scored their own SATCo testing sessions, by watching themselves test each participant in the video recordings. This information was used to assess intrarater reliability of the SATCo. A third rater also watched and scored the videos for Rater One and Rater Two to assess live versus video interrater reliability of the

SATCo. The live scores of the GMFM and SATCo from the first day of testing will be used for data analysis in study two. Participants will be barefoot for all testing.

Study Three employed an A_1BA_2 single-case experimental design to determine how a dynamic standing device (the Upsee) impacts trunk control and gross motor function in infants and toddlers with DS. Caregivers of the participants administered the home program with professional supervision by a physical therapist. The independent variables were the condition of no intervention and a dynamic standing home program. Dependent variables included assessment of trunk control over time as measured by the SATCo, and assessment of gross motor function over time as measured by the GMFM. For Study Three, a sub-sample of participants were recruited until at least five children and parents have completed the protocol. To ensure consistency, the same physical therapist performed testing on the same child from the start of Study Three until the end. For the A_1BA_2 design, there were two conditions: two baseline phases (A_1 and A_2) lasting four weeks each, in which participants continued with their usual therapy and no additional home program, and a six-week dynamic standing phase (B) employing a home program intervention. GMFM and SATCo scores were administered and recorded weekly through all three phases by the physical therapist, with each testing session lasting approximately 40 minutes, for a total of 14 testing sessions for each child. Testing was performed on the same day and time each week and at the same location of the caregiver's choice, either at the University of St. Augustine for Health Sciences or in the child's home. Throughout the study, the participants continued with their usual therapy sessions.

On the first day of the intervention phase, the principal investigator (PI) delivered the home program device (Upsee) to the parent and child. The PI provided training and education to the parent on the device and home program. The dynamic standing device were worn 30 minutes per day for the duration of the intervention phase. The parent was contacted by the PI weekly to check the fit of the device, ensure compliance, and answer any questions about the home program. Parents kept a daily log sheet to record duration of the device use, activities performed, and subjective observations of the child during the intervention. Study Three was expected to last 14 weeks.

DATA ANALYSIS

Data was analyzed using IBM SPSS Statistics 25 software.[‡] To test the hypothesis that the SATCo will demonstrate good interrater and intrarater reliability ($\kappa > 0.8$), Cohen's Kappa was calculated for each category of the SATCo (static, active, and reactive) and the overall score from 0 to 20. A p-value < 0.05 and confidence intervals not containing the null value of zero will indicate significance. To test the hypothesis that the SATCo will demonstrate concurrent validity with the GMFM dimension B, Spearman's rho was calculated for each category of the SATCo and dimension B of the GMFM. To test the hypothesis that age and SATCo scores have a predictive effect on GMFM in infants and toddlers with DS, a linear model was used: $\text{GMFM-66}_i = b_0 + b_1\text{SATCo}_i + b_2\text{Age}_i$.

To test the hypothesis that the six-week dynamic standing home program will demonstrate greater SATCo and GMFM scores than the baseline phases, visual analysis

[‡] IBM Corporation, Armonk, New York

of graphed data points were utilized, with the 2-standard deviation (2SD) band method for data analysis. The mean and standard deviation of each phase were calculated for each outcome measure. Statistical significance was indicated if two consecutive data points in a phase fall outside the 2SD band.[25-27] Between-phase comparisons were made and positive changes that are immediate, readily discernible, and maintained over time will be indicative of improvement.

CHAPTER II

REVIEW OF THE LITERATURE

INTRODUCTION

Infants and toddlers with DS display gross motor developmental delays and decreased trunk control.[3,8,28] Physical therapists should employ tools to measure deficits, demonstrate progress toward goals, and justify the need for physical therapy services. Measurement of gross motor function in this population can be achieved using the Gross Motor Function Measure (GMFM).[7] However, a tool for measuring trunk control has not been studied in this population. Additionally, to improve gross motor skills and trunk control in infants and toddlers with DS, effective home programs administered by parents and supervised by physical therapists should be identified.[15] For the purposes of this dissertation, "infants" is operationally defined as children below the age of 12 months, and "toddlers" are defined as children below the age of three years.

In this study, the reliability and validity of the SATCo was investigated in infants and toddlers with DS (between the ages of six to 24 months). The effectiveness of a home program using a dynamic standing device was also examined. This chapter summarizes the existing literature, including motor development, outcome measures, and home program interventions as they relate to trunk control and gross motor skills in children with DS.

MOTOR DEVELOPMENT IN CHILDREN WITH DS

DS is a genetic disorder of chromosome 21, which is usually identified in-utero or immediately after birth, with implications of developmental delays. In the United States, an estimated 12.6 per 10,000 live births are diagnosed with DS.[1] Babies with DS develop at a slower rate than those who are typically developing. Several studies have identified the ages at which most children with DS achieve gross motor developmental milestones. In a non-linear growth curve analysis study, Palisano et al[8] created gross motor curves for children with DS, based on their GMFM scores. A sample of 121 children with DS, ages one month through six years, was tested using the GMFM. The authors found that the probability that a child with DS would roll from supine to prone by six months was 51%.[8] Other estimates included: sitting independently by 12 months (78% probability), crawling by 18 months (34% probability), and walking by 24 months (40% probability).[8] They concluded that children with DS require increased learning time for new movements as the movement complexity increases.[8]

A study by Pereira et al[3] examined gross motor skills in 20 infants with DS between three and 12 months old and compared them to 25 typically developing infants using the Alberta Infant Motor Scale (AIMS). The AIMS is an observational outcome measure that evaluates motor development in supine, prone, sitting, and standing. This outcome measure demonstrates excellent interrater reliability (ICC = 0.98), and content validity.[29] The AIMS can be used in infants from birth until they achieve independent walking to identify infants with motor delay.[29] The authors discovered that the median age for infants with DS to develop sitting balance without support was ten months, while

typically developing infants achieved this skill by six months.[3] Not one infant with DS was able to achieve independent standing by 12 months of age, a skill typically developing infants were able to achieve at a median of 11 months.[3] As motor skills measured by the AIMS increased in complexity, the age difference between typically developing infants and those with DS also increased.[3] A lower percentage of babies with DS were able to achieve the more complex motor skills, such as reciprocal creeping, that are indicated by a higher AIMS score.[3]

In a similar investigation, Tudella and colleagues[4] used the AIMS to assess 19 infants ages three to 12 months with DS and 25 typically developing infants. Because the AIMS is a normative scale, the authors were able to determine age percentiles for the infants with DS. Babies with DS demonstrated the same sequence of development as their typically developing peers, but they showed a much slower rate of acquiring motor skills.[4] The majority of infants with DS were below the normative fifth percentile from three months to 12 months old.[4]

These three studies demonstrate that infants with DS demonstrate delays in gross motor skill acquisition compared to typically developing babies. This motor delay increases as gross motor skills become more complex. For example, typically developing infants are able to sit without support at approximately six months of age and can walk independently by 12 to 15 months of age. However, infants with DS attain independent sitting at approximately 11 months of age and walk independently at approximately 24 months of age.[3,4,8]

Gross motor development can have an impact on other domains of development. Houwen et al[30] recently published a study examining the interrelationships between the three domains of motor, cognitive, and language development in children with intellectual and developmental disabilities. They tested these three domains of development in 77 children with intellectual and developmental disability, including children with a diagnosis of DS, between 1 and 10 years of age.[30] The authors also tested the same domains of development in 130 typically developing children between three months to four years of age.[30] The main outcome measure used in the study was the Bayley Scales of Infant and Toddler Development, third edition (Bayley-III), which assesses developmental function across five scales: cognitive, motor (fine motor and gross motor), language (expressive communication and receptive communication), social-emotional, and adaptive behavior in children ages one to 42 months.[31] The Bayley-III shows evidence of reliability, Cronbach's $\alpha = 0.86$ to 0.91, and concurrent validity in infants with developmental delays.[31] The authors found correlations between the motor, cognitive, and language scales, with stronger associations in children with intellectual and developmental disabilities ($r = 0.61$ to 0.94), than in typically developing children ($r = 0.24$ to 0.56).[30] Based on their findings, the authors suggest early interventions to improve motor development may have an impact on other domains of development.[30]

Physical therapists can help accelerate the rate at which children with DS achieve gross motor developmental milestones, which may impact not only their gross motor development, but other areas of development as well. Early intervention to accelerate the acquisition of gross motor skills is supported by the literature;[15-17,32] however, physical

therapists require effective tools to measure any incremental changes that occur as a result of their treatment.

OUTCOME MEASURES

To measure change, physical therapists should employ outcome measures that accurately assess a child's body structure and function, as well as their functional activities as defined by the World Health Organization's ICF model.[6] When examining infants and toddlers with DS, measurements of trunk control (at the body structure and function level of the ICF) and gross motor skills (at the functional activities level of the ICF) can provide physical therapists with a more complete picture of the child's abilities. This section reviews outcome measures of trunk control, including the Level of Sitting Scale (LSS), Seated Postural Control Measure (SPCM), Sitting Assessment of Children with Neuromotor Dysfunction (SACND), and SATCo. Outcome measures for gross motor skills (AIMS, Bayley-III, and GMFM) will also be reviewed.

Measures of Trunk Control

Infant development tends to follow patterns. For example, children generally develop gross motor skills before they develop fine motor skills.[33] Most development occurs from a proximal to distal distribution. Children will typically develop strength and coordination in their proximal muscles prior to gaining these skills in distal muscles. Postural control "involves controlling the body's position in space for the dual purposes of stability and orientation."[34] In this dissertation, the operational definition for trunk control is the ability to regulate proximal muscles to maintain upright posture. As stated earlier, measuring trunk control falls under the body structure and function level of the

ICF. Many outcome measures examine sitting or standing balance, but do not measure control at the level of the trunk.

There are several measurements of trunk control for children, but most have been developed and tested in school-aged children. The Pediatric Reach Test, Trunk Control Measurement Scale, and Trunk Impairment Scale have evidence of good psychometric properties; however, they are intended for children older than five years of age. To complete these assessments, children must be able to understand and follow simple directions. Therefore, these outcome measures would not be appropriate for the target population of infants and toddlers with DS. Though not specific to DS, the LSS, SPCM, SACND, and SATCo have been tested in children and youth who were younger than three years old, with a variety of neuromotor disorders. These outcome measures may be appropriate for measuring trunk control in infants and toddlers with DS.

The Level of Sitting Scale

The LSS was originally developed to assess seated posture and motor control to inform clinicians in the evaluation of seating systems for children with neuromotor disabilities.[35] To assess a child's level of sitting, the child is first asked or assisted into a sitting position on a bench or table with thighs supported and feet unsupported.[36] If the child is able maintain sitting independently for 30 seconds, he or she is then requested to shift and re-erect his or her trunk.[36] This can be accomplished by reaching for a toy outside the child's base of support. There are eight levels of sitting, with Levels Six through Eight representing a child's ability to maintain sitting while shifting the trunk in different directions.[36] Level Five indicates the child can maintain static sitting for 30

seconds independently.[36] At Level Four, the child requires support at the pelvis to maintain static sitting balance.[36] If the child needs support at the shoulders or anywhere on the trunk, he or she is scored as Level Three.[36] Level Two indicates head support in addition to trunk support. Level One is a child who is not able to be placed in upright sitting.[36]

The original sample for interrater and test-retest reliability included 40 non-ambulatory children with cerebral palsy (CP) in GMFCS levels IV and V and "similar disorders", ages one to 19 years.[35] A follow-up study assessing validity included 114 non-ambulatory children with neuromotor disability.[36] According to Koo and Li,[37] interpretation of interclass correlation coefficiency (ICC) reliability is: poor (< 0.5), moderate (0.5 to 0.75), good (> 0.75 to 0.9), and excellent (> 0.9). For children with neuromotor disabilities between the ages of one and 19 years, the LSS demonstrates moderate interrater reliability ($\kappa = 0.58$ to 0.62) and moderate test-retest reliability ($\kappa =$ 0.54 to 0.55).[35] Construct validity reveals a negative correlation with LSS score and the amount of external support required for sitting (Spearman's rho = -0.42, $p < 0.05$). [36]

There are several advantages to using the LSS to measure trunk control. The LSS measures both static and active control in a seated position. The measure is relatively quick and easy to administer, requiring very little equipment. The population used to develop the LSS included children with varied neuromotor disabilities and was tested in children as young as one year old. However, this measure may not be ideal for measuring trunk control in infants and toddlers with DS. The LSS was developed to assist clinicians in evaluating seating needs. Children with DS are not expected to require

special seating as they age. Additionally, the LSS does not measure discrete levels of trunk control. Children score level two if they require head support and level three if they require support anywhere on the trunk. A child who requires support at the upper trunk may have less trunk control than a child who requires support lower on the trunk, but the LSS would not be sensitive to these differences. Furthermore, the LSS does not measure reactive control.

The Seated Postural Control Measure

The LSS was developed as part of a larger outcome measure, the SPCM, which is an observational scale for use with children who have adaptive seating needs.[35] The SPCM contains 22 postural alignment items and 12 functional movement items.[35] Each of the 34 total items is scored on a four-point scale.[35] The alignment domain contains items that assess body alignment using visual inspection or palpation.[35] Examples in this section include pelvic obliquity (>25° = severe, 1 point; neutral = normal, 4 points) and lateral head tilt (>35° = severe, 1 point; neutral = normal, 4 points).[35] The functional movement domain involves a task that is meaningful to the child, measuring the degree to which the child achieves the movement goal, not the quality of the child's movement in attempting the task. In this section, children are asked to reach, pick up an object (for example, a toy or pen), and use the object in a meaningful way.[35] Essentially, the SPCM is a longer, more detailed version of the LSS. The SPCM can be administered with the child sitting on a bench or while using his or her prescribed seating system.

The SPCM has an interrater reliability of poor on the alignment subscale, $\kappa = 0.41$ to 0.47, and moderate to good on the functional movement subscale, $\kappa = 0.79$ to 0.87.[35]

Test-retest reliability was poor on both the alignment subscale, $\kappa = 0.36$ to 0.41, and the functional movement subscale, $\kappa = 0.28$ to 0.29.[35] Items were generated through consultations with therapists who were experienced with seating needs in children.[35] Items were then externally evaluated by seven seating experts to establish content validity and clinical utility.[35]

As with the LSS, this outcome measure is not ideal for measuring trunk control in infants and toddlers with DS. Although the SPCM is relatively easy to administer and requires no special equipment, its psychometric properties are on the lower end of the spectrum. One advantage of the SPCM is the measurement of both static and dynamic control, however, there is no distinction for quality of movement. The SPCM may be more detailed than the LSS, but it demonstrates lower interrater reliability overall. After the initial study, no follow-up studies were performed in the pediatric population.

Sitting Assessment of Children with Neuromotor Dysfunction

As described by Reid,[38] the SACND was originally designed to assess quality of static and dynamic sitting in children with neuromotor disabilities. To complete the outcome measure, the child is asked to maintain sitting on a bench for five minutes at rest while listening to a story or watching a video ("Rest" module).[38] The child is then asked to perform reaching for five minutes toward objects on a board in multiple directions: forward, up, down, and to each side using his or her dominant hand ("Reach" module).[38] The Rest and Reach modules each contain four constructs of mature seated postural control: proximal stability, postural tone, postural alignment, and balance. [38] Constructs are given a score from one to four, with a lower score indicating higher ability and

"normal" behaviors.[38] Operational definitions are provided for the constructs on the SACND.

Interrater reliability for both the combined Rest and Reach modules of the SACND was excellent (ICC > 0.99).[39] Additionally, interrater reliability was excellent for the Rest module subscales (κ = 0.91 to 1.0) and for the Reach module subscales (κ = 0.96 to 1.0).[39] Test-retest reliability was good for the Rest module subscales (κ = 0.87 to 1.0) and excellent for both the Reach module subscales (κ = 0.91 to 1.0) and for the Rest and Reach modules (ICC > 0.99).[39] Internal consistency was reported by Reid for both the Rest module (Cronbach α = 0.47 to 0.49) and the Reach module (Cronbach α = 0.71 to 0.78).[38] Content validity for this outcome measure was established by using the items and scoring from four key constructs derived from a literature review.[38] In addition, 13 pediatric occupational therapists agreed on the importance of each concept (67%), clarity of wording (70 to 73%), and reflection of operational definition on scoring (66 to 69%).[38] To support construct validity, the author reports no difference on 14 out of 32 items between the Rest and Reach subscales, indicating postural control was the same for static and dynamic sitting for these items.[38] However, 16 items had lower scores for the Rest module and two items were lower for the Reach module.[38]

Two studies have used the SACND as a primary outcome measure.[40,41] In an experimental crossover design, Reid[40] examined the effects of a saddle seat on postural control in children with "moderate" spastic CP, ages four to nine years. The Rest module of the SACND was used to measure static postural control in six children, ages four through eight, during two different experimental conditions: flat bench and saddle seat.[40]

Children sat on both surfaces during two separate sessions and the order of the experimental conditions were reversed during the second session.[40] Results indicated no significant difference for seat condition order (p = 0.07 to 0.7).[40] Paired t-tests were performed to determine whether there was a significant difference between the two seat conditions for total Rest module score.[40] The saddle seat had a significantly improved SACND Rest module score, more so than the flat bench (p = 0.007).[40]

In a case study, Knox[41] evaluated the SACND as a measurement tool in a 3-year-old child with spastic diplegic CP. The SACND was administered to the child before and after a 2-week intense physiotherapy intervention.[41] The child improved in every subscale for both modules of the SACND. The biggest improvement occurred on the Reach module.[41] The author concludes that the SACND was sensitive to change for this one child, providing an objective assessment of progress.[41] However, sensitivity for this outcome measure has not been examined in a larger population, therefore, more studies are warranted.

The SACND is a measure of static and dynamic sitting balance. The operational definitions and pictures that are provided give the rater the ability to examine the quality of postural control, which is important for discriminating trunk control from sitting balance. However, the test does not differentiate between different levels of trunk control. Alignment of the trunk is observed with no distinction between head, thoracic, or lumbar control. Children receive a score of one on the balance subscale of the Rest module if they can statically sit with hands resting comfortably on the lap. The score increases, indicating decreased balance, if the child uses his or her arms to assist with upright

sitting. Children with head and thoracic trunk control may be able to sit without upper extremity support if given external support at the pelvis or lumbar spine, but this outcome measure does not differentiate the amount of control at different levels along the trunk. The SACND does not have a category for reactive postural control. If a child is pushed or nudged unexpectedly, would he or she be able to maintain seated postural control? Another concern in using this outcome measure is the endurance required for administration. Although ten minutes is not a burden on the examiner, the child is asked to sit for five minutes during each module, which would be difficult for children under two years of age. For these reasons, the SACND may not be the ideal outcome measure for infants and toddlers with DS.

Segmental Assessment of Trunk Control

The SATCo was designed and refined by Butler et al[10] to assess discrete levels of trunk control in children with motor disabilities. To complete this assessment, the examiner places the child sitting on a low bench with his or her feet on the floor. The child's pelvis is secured in a neutral position, either with a positioning strap or with an assistant's hands.[10] The tester gives manual support horizontally around the trunk, starting at the shoulders and moving segmentally down the trunk through seven levels.[10] At each level, the child is tested on static trunk control (ability to hold head in neutral for five seconds), active trunk control (ability to turn head left and right), and reactive trunk control (ability to maintain control with perturbations at the sternum, thoracic spine, and at each acromion).[10] The child's arms must not be used to maintain sitting balance or upright posture.[10] Levels two through seven of the SATCo contain three items each

(static, active, and reactive trunk control) and level one contains two items (static and active trunk control).[10] Therefore, the child can receive an item score from zero to 20. The level of trunk control is the highest level at which the child is able to maintain control in all three aspects. Higher levels indicate better trunk control.[10]

Initial psychometrics of the SATCo indicate this outcome measure is a good tool for measuring discrete levels of trunk control in different populations. Reliability and validity of the SATCo were established through rating eight typically developing children, ages three to nine months of age, as well as 24 children with neuromotor disability, ages 18 months to 17 years of age.[10] Twenty-one of the 24 children with neuromotor disability were diagnosed with different types of CP across all five GMFCS levels, but none of the children tested had a diagnosis of DS.[10] Intrarater reliability for the SATCo was excellent (ICC = 0.98) across all data sets and all aspects of control.[10] Interrater reliability for the SATCo was good (ICC = 0.8 to 0.84) for the total and both subsets of infants with typical development and children with neuromotor disability.[10]Concurrent validity was established with GMFM Dimension B (sitting) (r = 0.73 to 0.83) and with Alberta Infant Motor Scale (sitting section) (r = 0.86 to 0.88).[10] A recent study by Cardoso de Sá et al[12] found excellent interrater reliability ($\kappa = 0.901$) when rating the SATCo in 50 children with Duchenne muscular dystrophy. Responsiveness and sensitivity to change of the SATCo has been studied in children with spinal cord injury (SCI).[42] In a sample of 21 children with SCI, regardless of injury severity or initial score, SATCo scores increased significantly after one, two, and three months of activity-based locomotor training ($p < 0.05$). [42] The SATCo demonstrated a

large effect size (> 0.8) for responsiveness to change from baseline to discharge after the intervention.[42]

Curtis et al[13] studied the relationship of the SATCo and the GMFM in children with CP, ages one to 14 years. The authors performed a retrospective cross-sectional study on 92 children with spastic, ataxic, or dyskinetic CP across all five GMFCS levels to determine the predictive effect of SATCo level, age, and neuromotor disability on GMFM scores. They found that both SATCo level and age were significant predictors of GMFM score ($p < 0.001$ and $p = 0.033$, respectively). The article supports the hypothesis that trunk control influences gross motor function in children with CP.[13] For the target population of children with DS, the relationship between trunk control and gross motor function has not yet been investigated.

Of the trunk control outcome measures analyzed thus far, the SATCo is unique in that it measures static, active, and reactive trunk control. Like other measures previously discussed, the SATCo is relatively quick to administer and requires very little equipment. The instructions clearly describe how to administer the SATCo, with photos of common compensations to look for during item administration. The SATCo measures trunk control throughout the spine, from the pelvis all the way up to head control. One disadvantage to this measure is that the strapping system is cumbersome. Although the instructions demonstrate how to use the strap, many clinicians prefer to have an assistant hold the child's pelvis in a neutral position. However, this method then requires two people to administer the test, utilizing more clinic resources. Another consideration is that

the SATCo only measures trunk control during sitting. Other postures, such as supine, prone, quadruped, or standing are not analyzed with the SATCo.

Conclusion: Measuring Trunk Control

In a systematic review of clinical measures of sitting balance for children with CP, Banas and Gorgon[43] found that four clinical measures were suitable for use in clinical practice based on their psychometric properties: SACND, SATCo, Pediatric Reach Test, and Trunk Control Measurement Scale. Two of these outcome measures were not included in this literature review because either they were developed for older children, or they grossly measured sitting balance instead of trunk control. To date, none of these outcome measures has been studied in infants and toddlers with DS. Based on the available evidence from the literature, the SATCo appears to be the most feasible outcome measure for quantifying discrete changes in head and trunk control in the target population of infants and toddlers with DS.

Measures of Gross Motor Function

Commonly used norm-referenced tests of gross motor function in children include: the Peabody Developmental Motor Scales, second edition (PDMS2), the Buininks-Oseretsky Test of Motor Proficiency, second edition (BOT-2), the Test of Infant Motor Performance (TIMP), the AIMS, and the Bayley-III. The PDMS2 can be administered to children from birth through five years of age but is not sensitive to change over a six-month period for infants with neuromotor delay, and a large cohort ($N \geq 68$) is needed for research studies.[44] The BOT-2 has evidence of good psychometric properties; however, it is intended for children over the age of four years old, which

would exclude infants and toddlers. The TIMP is a test of posture and movement for infants less than four months of age, designed to predict gross motor delays later in life.[45] The TIMP is also outside the age range for the target population and cannot be used over time to evaluate gross motor skills. Therefore, of the five common norm-referenced tests of gross motor function, the AIMS (developed for infants and toddlers ages birth to 18 months old) and the Bayley-III (intended for children ages one month to 42 months) are the most appropriate for the target population.

One more gross motor assessment, the GMFM, would be appropriate for infants and toddlers with DS. This criterion-referenced test has been studied for reliability and validity in children with DS of all ages.[7] To assess gross motor function in infants and toddlers with DS, the three most relevant outcome measures are the AIMS, the Bayley-III, and the GMFM. Each of these were explored in more detail to determine the best outcome measure for this study.

Alberta Infant Motor Scale

The AIMS was developed by Piper and Darrah[26,46] to provide a standardized, performance-based, norm-referenced assessment of the motor skills in developing infants. Typically developing and motor delayed infants (birth to 18 months of age) were included in the construction and development of this outcome measure, $N = 2{,}202$.[26,46] The assessment features pictures and descriptions of typical infant motor behavior that allows the examiner to check off skills based on observation of the child's performance.[26] A manual for the administration and interpretation of the AIMS is available to aide in scoring.[46] Although developed for infants from birth through 18 months of age, the AIMS

is commonly used for infants and toddlers not yet walking but should not be used for infants who demonstrate pathological patterns of movement (abnormal movements that are not observed in typical development).[46] Children with DS tend to display immature motor abilities resulting in gross motor delay, so the AIMS can be used in this population. The examiner evaluated infant positions in four subscales: prone, supine, sitting, and standing. The AIMS scoresheet consists of 58 items across the four subscales that address weight bearing, posture, and antigravity movements. The child obtains a score of zero (not observed) or one (observed) for each item. The total is calculated into a raw score, which can then be converted into a percentile rank and compared to normative age-equivalent peers. The test takes 10 to 20 minutes to complete and requires no special equipment.[46]

As a norm-referenced test to identify gross motor delays in infants, the AIMS has demonstrated good psychometric properties. Interrater reliability for the AIMS is excellent[26] (ICC = 0.98) and predictive and content validity have been established.[26,47] Concurrent validity has been verified through comparison of scores with the PDMS2[48] (Spearman's rho = 0.90 to 0.99) and the BDMS III (Spearman's rho = 0.94 to 0.97).[31] The AIMS has also been used in numerous studies[3,4,49,50] to evaluate infants and toddlers with DS.

There are several advantages to using the AIMS for infants and toddlers with DS. This outcome measure requires very little equipment (flat surface for infant, small toy) and takes less than 20 minutes to administer. The examiner observes the infant and scores him or her appropriately. No special training of examiners is required for item

administration, just a general knowledge of typical infant development. However, one drawback to this outcome measure is that the highest gross motor skill possible is independent walking. The test does not assess children on their gross motor skills once they are able to walk independently. Therefore, although the AIMS may be ideal for assessing gross motor skills in infants and toddlers with DS, it would not be appropriate as a gross motor assessment after they have started to stand and walk without assistance.

Bayley Scales of Infant and Toddler Development, 3rd edition

The Bayley-III is in its third edition, most recently updated and standardized in 2006 using a random normative sample of children from one month through 42 months of age (N = 1700).[31] For this latest edition, an equal number of male and female children were recruited, as well as children from various economic, geographic, racial, and ethnic backgrounds, representing a proportionate sample from the United States 2000 Census.[31] The Bayley-III is designed to identify suspected developmental delay by measuring five scales: cognitive, motor, language, social-emotional, and adaptive behavior.[31] Three of these scales are administered through child-interaction (cognitive, motor, and language) and two of the scales are conducted through a parent questionnaire (social-emotional and adaptive behavior).[31] The motor scale is composed of fine motor and gross motor subtests. There are 72 items in the gross motor subtest, covering limb movement, static positioning, dynamic movement, balance, and motor planning. The fine motor subtest has 66 items. When administering all five scales, the Bayley-III takes 30 to 90 minutes to complete. The examiner gives the child a score of zero (does not complete) or one (completes) for each item. Total raw scores are calculated for each subtest. Scaled scores

are then derived from the subtest total raw score. These scores are then summed to produce a composite score, which can be converted to a percentile rank for each of the five scales.[31]

The Bayley-III was assessed through a literature review, expert consult, examiner observation, and interpretation of the test to establish content validity.[31] Inter-item consistency reliability was reported to be good to excellent for the normative sample for the receptive communication, expressive communication, cognitive, fine motor, and gross motor subsets (Cronbach's $\alpha = 0.86$ to 0.91).[31] When tested in special clinical populations (children with diagnoses such as CP, developmental delay, prematurity, and DS), reliability was higher (Cronbach's $\alpha = 0.84$ to 0.99).[31] Interrater reliability has not been established. Cognitive, language, and motor scales had a test-retest reliability that ranged from 0.67 to 0.94 in a sample of 197 children between two and 42 months old.[31] The correlation increased as the age increased.[31]

The Bayley-III has the advantage of being a comprehensive measure of development over multiple domains. However, some authors have raised concerns about the ability of the Bayley-III to identify children with developmental delay.[51,52] They question whether the psychometric properties of the Bayley-III are sufficiently strong enough to warrant its use in the clinic.[51] Additionally, the Bayley-III is a long assessment, often taking over an hour to complete. Physical therapists could administer only the motor scales portion of the Bayley-III in order to capture a child's gross motor function; however, the test is meant to be given in its entirety. Special equipment and training are required for administration of the Bayley-III. When considering the Bayley-III as an

assessment of gross motor function in young children with DS (under four years old), this outcome measure may not be the ideal choice for this study. The length of the test, questionable psychometric properties, and lack of specificity for gross motor skills are some of the disadvantages to using the Bayley-III in the target population for this study.

Gross Motor Function Measure

The GMFM is a measure of gross motor function, originally developed for children with CP, which can also be used in children with DS under six years old.[23,24] To administer the GMFM, the examiner scores a child's capabilities across five dimensions of functional movement: A) Lying and Rolling, B) Sitting, C) Crawling and Kneeling, D) Standing, and E) Walking, Running, and Jumping. Each dimension is made up of multiple items on an ordinal scale, for a total of 88 items. The child receives a score of 0 (does not initiate), 1 (initiates), 2 (partially completes), or 3 (completes). This outcome measure can be administered by direct observation by the examiner or by parent report. The GMFM can take up to 60 minutes to administer, depending on the examiner's familiarity with the test and the child's gross motor capabilities. For example, younger children who are not yet standing or walking would not be able to complete Dimension D and E, decreasing the time needed for test administration.[24]

The items in the GMFM indicate whether a child is able to perform (or partially perform) a functional activity but not the quality of the movement performed. However, because there are 88 items in the GMFM, children can show improvement incrementally. For example, in Dimension B (Sitting) item 21, the child can maintain head control in an upright position for three seconds while sitting on a mat with his or her thorax supported

by a therapist. In the next item, the child is able to maintain the same position, but with head control for 10 seconds. The GMFM manual gives specific instructions as to how the child would score a 1 (initiates) or a 2 (partially completes) for each item.[24]

In a study to evaluate the GMFM for children with DS, two articles reported on the psychometrics of the GMFM in a sample of 123 children with DS, ages 1.7 months to 72 months of age.[7,9] The children were assessed twice over a six-month period on both the GMFM and the Bayley II.[7,9] They found evidence that the GMFM was responsive to change over time in children with DS, with a mean change in GMFM scores of 12 points.[7,9] In these studies, the GMFM was more responsive than the Bayley II.[7,9] To examine reliability, the authors used a sub sample of 22 children with DS, ages five months to 67 months. The GMFM has strong interrater reliability (ICC = 0.96 to 0.98) and test-retest reliability (ICC = 0.95 to 0.96).[7,9]

Conclusion: Measuring Gross Motor Function

Thus far, GMFM has been extensively studied in children with DS and seems to be a good choice for measuring gross motor function over time in the target population. For example, AIMS cannot be used to measure gross motor function once the child attains walking. Although no training or special equipment is needed for the GMFM, using the manual is necessary for those unfamiliar with this test. Another advantage of this assessment is that parent report can be used for items not observed by the examiner.[9] The length of the test (up to 60 minutes) is one of its drawbacks. Based on the available evidence, the GMFM appears to be the most feasible outcome measure for quantifying

gross motor function in the target population of infants and toddlers with DS for this study.

INTERVENTIONS FOR INFANTS AND TODDLERS WITH DS

Children with DS need access to physical therapy early to improve the quality and variability of their movement, as well as to accelerate the acquisition of gross motor skills. A search of the literature reveals evidence exists for a variety of different early physical therapy interventions for this population. This section reviews direct interventions for infants and toddlers with DS performed by physical therapists, including: strength training, hippotherapy, sensorimotor intervention, and massage.

Lower Extremity Strength Training

Many early intervention programs focus on strengthening as a basis for developing gross motor skills. However, few studies have evaluated the benefits of strengthening as a physical therapy intervention for infants and toddlers with DS. One study by Santos et al[53] analyzed the effect of adding lower extremity (LE) weights on the frequency of kicks in three and four-month-old infants with DS. The authors speculate that kicking in infants is a precursor to more complex motor skills.[53] They hypothesized that an increase in LE strength and coordination will produce a more developed movement pattern that can develop into more complex tasks such as crawling and walking.[53] In their study, the authors recruited five infants with DS and five typically developing infants, ages three to four months.[53] The infants were positioned supine and stabilized at their shoulders with a touch pad positioned near their feet.[53] When the infant kicked the pad, he or she was rewarded with a visual and auditory stimulus via activation

of a melody and moving mobile above the infant's head.[53] Infants went through a training phase in which the examiner placed the infant's feet on the touch pad and activated the mobile. Frequency of kicks were analyzed during three conditions: baseline (free kicking without restrictions), weight (addition of ankle weight equal to one-third of the lower limb weight based upon body weight), and post-weight (same as baseline).[53] Each condition lasted one minute, with a 30 second rest between conditions. Testing lasted approximately 5 minutes for each infant. Data was analyzed using a multivariate analysis of variance (MANOVA) using group, age, and condition as the independent variables.[53] Dependent variables were frequency of kicks, frequency of foot contact on touch pad, and frequency of success in moving touch pad to make the mobile move.[53] The authors found that infants with DS had significantly lower frequencies than the typically developing infants for all variables between all groups ($p < 0.05$).[53] Additionally, both groups significantly improved from baseline to post-weight in the frequency of foot contact and the frequency of success ($p < 0.001$).[53] They concluded that the added weight was an important stimulus for both groups and was especially important for the infants with DS because they showed improved motor control and muscular strength required for acquiring new motor milestones.[53] The small sample size was a limitation of this study. Also, it is unclear how the infants performed over time. A longitudinal study that also analyzed gross motor skills would be helpful in interpreting the results. A future study might compare frequency of kicks in two groups of infants with DS with and without added ankle weight. Although the results are interesting, it is difficult to draw conclusions about improvement in overall gross motor skills.

Hippotherapy

Hippotherapy is a therapeutic intervention performed by physical, occupational, and speech therapists that uses a horse's multidimensional movement to improve motor skills in the rider. In a typical hippotherapy session, a child sits on a horse with a therapist walking beside the horse and facilitates the movement of the child. Additional assistants are present to manage the horse (front walker and side walker) so that the physical therapist can concentrate on the child's treatment. Champagne and Dugas[54] documented a case study using hippotherapy in two young children with DS. The purpose of their study was to assess how forward horse motion influenced gross motor function in the two participants.[54] The two children were 28 and 37 months old and had medical consent to participate in hippotherapy through their physicians.[54] X-ray screening of atlantoaxial instability was negative for both children.[54] The children each participated in one 30-minute hippotherapy session per week for 11 weeks, led by an occupational therapist.[54] Both children performed three different positions in each session (sitting forward, sitting sideways, and sitting backward) while wearing a helmet and safety belt when riding. Gross motor ability was assessed through administration of the GMFM by two independent raters before and after the 11-week hippotherapy intervention. Data was analyzed using a *t*-test, revealing a significant increase in the mean GMFM total scores from pretest to posttest for child 1 ($t = 3.058$, $df = 4$, $p < 0.05$).[54]

Although the treatment effect for Child 2 did not meet the criteria for significance, there was an increase in overall GMFM total scores ($t = 2.662$, $df = 4$, $p = 0.056$).[54] The authors speculate that they did not see a statistically significant difference in scores for

Child 2 because he was close to a ceiling effect in dimensions A and B of the GMFM.[54] Both children showed the most improvement in dimension E (running, walking, and jumping) of the GMFM.[54] Because of the small sample size, caution should be used in generalizing this case study to the greater DS population. The results of this case study are promising; however, a single-case design may have provided more valuable information on GMFM changes over time with intervention. Although hippotherapy may improve gross motor skills in young children with DS, it is not an intervention that is easily accessible to all children. Hippotherapy is more costly than traditional physical therapy, requiring extra space, additional personnel, and maintenance of the horses. The cost and labor limits the number of clinics that offer hippotherapy as an intervention.

Sensorimotor Interventions

Sensorimotor interventions promote task specific gross motor function by providing a child a combination of sensory and motor experiences. LaForne Fiss et al[55] investigated whether children with DS receiving individualized intervention (physical and occupational therapy) would improve in their gross motor skill acquisition with the addition of sensorimotor group participation than those who only received the individualized intervention. They recruited 10 children with DS, five in the treatment group and five in the control group, ages 13 to 29 months. Both groups of children continued to receive their regular weekly, individualized physical and occupational therapy sessions based on their current plan of care. The treatment group also attended 10 weekly sensorimotor group sessions outside of their normal course of therapies. The

sensorimotor groups were led by one physical therapist and one occupational therapist with parents present and participating in the sessions. Group sessions lasted for 1 hour.[55]

Participants in both the control group and the treatment group were assessed using the GMFM and Goal Attainment Scaling (GAS), which uses individualized objectives (goals) set by a therapist to assess progress based on a child's baseline status and expected outcome. Testing occurred pre-intervention, post-intervention, and at a 5-week follow up visit. The Mann-Whitney *U* test was used to compare GMFM and GAS scores between the two groups. The treatment group demonstrated a greater average improvement on the GMFM (between 1.4% to 14.8%) and improvement on the GAS.[55]

The authors found a significant difference between the two groups from pretest to posttest for both the GMFM and the GAS, with a significance level of $p < 0.01$.[55] The authors justified their use of a non-conventional *p* value for significance because of their exploratory study design using a small sample size.[56,57] The effect size between the two groups for change in GMFM scores was large ($d = 1.116$), as was the effect size for change in GAS scores ($d = 1.075$).[55] No significant difference was found in the outcome measures from pretest to follow-up or posttest to follow-up.[55] The authors concluded that participation in sensorimotor groups along with individualized intervention can improve motor skill acquisition in young children with DS more than individualized treatment alone.[55] These findings should be interpreted with caution because of the small sample size and inconsistency of the GAS. The "individualized intervention" that all children participated in included a wide range of physical and/or occupational therapy of varying frequencies performed by different therapists with varying levels of experience.

Massage

Therapeutic massage is a passive technique that involves gentle rubbing or kneading of soft tissue to promote healing. Hernandez-Reif et al[58] investigated whether massage could improve motor function and muscle tone in infants and toddlers with DS. The researchers recruited 21 children with DS with a mean age of 24.5 (*SD* = 9.5) months.[58] The children were randomly assigned to either the control group, in which children were held and read to for 30 minutes twice weekly (*n* = 12), or the experimental group, in which children received a 30-minute massage twice weekly (*n* = 11).[58] All children continued with their regular physical and occupational therapy sessions per their individual care plans.[58] Pretesting and posttesting occurred on the first day and last day of the study. Outcome measures included the Developmental Profile for Infants and Young Children (DPIYC) and the Arms, Legs and Trunk muscle tone score (ALT), a new preliminary scale developed by two of the authors.[58]

The DPIYC is a checklist that provides developmental age appropriate norms across many areas of development (fine motor, gross motor, self-care, social/emotional, language, and cognition). This scale was developed in the 1970s but has not been updated recently.[58,59] It was chosen by the authors out of convenience and familiarity.[58] The scale has been correlated with an earlier version of the Bayley III (*r* = 0.62 to 0.96), and interrater reliability for the DPIYC is 82%.[59,60] The ALT was designed by two of the authors during the pilot phase of the study in an attempt to objectively measure fluctuations in tone in the arms, legs, and trunk.[58] The authors report that the ALT has good reliability (Cronbach $\alpha = 0.90$).[58]

The study lasted 8 weeks. During the massage sessions, children were placed supine and prone and were not stimulated for active movement. The infant massage technique was performed on the LEs, stomach, face, hands, arms, and back. Children in the control group received individual attention from a massage therapist who held the child and read a book. A one-way analysis of variance was conducted to compare both groups at baseline revealing no significant differences between the groups for the DPIYC. Change scores on the DPIYC were calculated by subtracting pretest scores from posttest score, which were then analyzed using one-tailed *t*-tests. The children in the experimental group had significantly greater change scores than the control group in fine motor functioning ($t(19) = 1.80$, $p < 0.05$) and gross motor functioning ($t(19) = 1.74$, $p < 0.05$). Mann-Whitney *U*-tests were conducted on the change scores on the ALT. There was a significant improvement in arm muscle tone ($U = 8.5$, $p \leq 0.05$) and LE muscle tone ($U = 8.5$, $p \leq 0.05$).[58]

One of the biggest limitations in this study was the selection of the outcome measures. Psychometric properties for the DPIYC and the ALT have not been extensively studied. These outcome measures are not commonly used in research for children with DS and the improvement in change in scores should be interpreted with caution. Increased scores may also be attributed to natural maturation effects. The use of a passive technique such as massage to improve gross motor function is questionable. The literature does not support passive manual techniques to improve active function in children.

Modified Ride-On Cars

A ride-on-car (ROC) is a commercially-available power mobility device designed for typically developing children, ages one to five years of age. Recently, ROCs have been modified for use by children with neuromotor disabilities or developmental delays to increase their independent mobility and improve their quality of life. Programs such as Go Baby Go (University of Delaware, Newark, DE) lead workshops to teach therapists how to make accessible mobility for their clients using a ROC and modifying it with common materials such as Velcro, pool noodles, adaptive switches, and plastic pipe. These modified ROCs can be used by young children (children less than three years old) with neuromotor disabilities who would otherwise be dependent for mobility, thus potentially increasing their participation in everyday activities.

Huang and Chen[61] examined whether ROC training is more effective than traditional therapy on mobility and socialization in young children (ages one to three years) with neuromotor disabilities. Children with diagnoses such as CP, DS, Developmental Delay, and "Other" were recruited and assigned to either the ROC intervention group (n = 10) or the control group (n = 10).[61] All children participated in their regular physical, occupational, and speech therapy programs throughout the study. Children in the ROC intervention group received supervised ROC training with an occupational therapist for two hours a day, twice per week for nine weeks. The same occupational therapist performed all the treatment sessions for all children in the ROC intervention group. Children in the control group did not receive additional intervention and attended therapies for one to two hours a day, twice per week for nine weeks. The

ROC intervention was performed at a hospital using public spaces. Each child received two 35-minute ROC driving sessions and two 25-minute natural play sessions during each treatment. The ROC driving sessions concentrated on building cause-and-effect concepts, goal-oriented driving, and using hands for exploration and play.[61]

Mobility and socialization were assessed using the Pediatric Evaluation of Disability Inventory (PEDI). The PEDI is an outcome measure that assesses a child's self-care, mobility, and social function using scales of functional skills, caregiver assistance, and modifications.[62] Construct validity has been established for the PEDI, as well as intrarater reliability (ICC = 0.95 to 0.99) and interrater reliability (ICC = 0.64 to 0.74).[63] A separate, blinded occupational therapist administered the PEDI on the children before and after the 9-week intervention.[61] The parents also completed the Parenting Stress Index (PSI), a questionnaire that measures perceived stress in parents. Paired and independent *t*-tests were used to compare the mean differences within and between groups.[61]

There were no significant differences in demographics between the ROC intervention and control groups. There was no significant difference of PEDI scores or PSI scores between the two groups at pretest. At the posttest assessment, the ROC intervention group had a significant improvement in the PEDI mobility domain ($p < 0.001$), PEDI social function domain ($p = 0.002$), and PSI ($p = 0.01$).[61] The control group showed significant improvement in the PEDI social function domain ($p < 0.001$) but no significant change in the PEDI mobility domain ($p = 0.10$) or PSI ($p = 0.26$).[61] There were no significant differences between the two groups at posttest.[61]

Like other studies for young children with neuromotor disabilities, this study lacks power. The authors found that to achieve a power of 0.8 with an effect size of 0.55, a sample of 54 children with neuromotor disabilities would have been needed.[61] The study did not measure the long-term effects of the ROC intervention. The ROC intervention was limited to supervised clinic-use only, but more improvements in outcome measures may have been evident if the children and caregivers were given the opportunity to practice with the ROC at home. A drawback to the use of ROCs in treatment interventions is the cost and labor involved in modifying the ROC. The cost of a child's ROC is around $250 before modifications. Although this is much less expensive than traditional power mobility, the cost can be prohibitive for some families. Each child requires different modifications, which can take a few hours to complete.

HOME PROGRAMS FOR INFANTS AND TODDLERS WITH DS

For infants and toddlers with neuromotor disabilities receiving early intervention therapies, Beverly D. Ulrich[15] advocates for an "increase in the structured engagement of caregivers, guided by therapists, in administering well-defined activity intervention programs focused on the development of specific functional skills." She argues that therapist-supervised home programs have the potential to optimize development of neuromotor control in a financially feasible way.[15] The home environment plays an important role in the development of motor skills in infants and toddlers with DS.[49] Infants and toddlers with DS may benefit from structured, supervised home programs to improve their trunk control and gross motor skills. This section will explore the available

evidence for physical therapy home program interventions in infants and toddlers with DS.

Home Massage

The use of massage to improve gross motor function was discussed earlier in the context of a trained professional administering the intervention. Silva et al[64] published a small randomized controlled study on the effects of daily qigong massage administered by parents on motor skills and sensory regulation in young children (under four years old) with CP and DS. Participants included nine children with DS and 16 children with spastic CP with a mean age of 2.44 (*SD* = 1.46) years.[64] Children were randomly assigned to the experimental group (A) or the control group (B). After a five-month waiting period in which the Control Group B underwent a pretest and posttest, the children in Group B also received the 5-month intervention. All children continued with their regular physical and occupational therapy sessions. Both groups underwent a pretest and posttest using the PDMS-2 to assess gross motor skills, and the Sense and Self-Regulation Checklist (SSC) to assess sensory responses. The SSC is a parent questionnaire developed by two of the authors to measure hyposensitivity and hypersensitivity of the five senses to common stimuli in children under six years of age.[65] The tool has evidence of internal consistency (Cronbach's $\alpha = 0.87$) but has not been broadly researched.[65] Scores on the SSC range from 0 to 75.[65] As discussed previously, the PDMS-2 is a norm-referenced instrument used to measure fine and gross motor skills in children from birth through age five. The Gross Motor Scale of the PDMS-2 tests three domains: Stationary (30 items), Locomotion (89 items), and Object Manipulation (24 items).[48]

To administer the intervention, parents attended a 3-hour training session prior to the start of the study. The intervention phase lasted 5 months, during which parents performed a qigong massage protocol daily on their children. Each week parents and their children attended a 30-minute support program in which a qigong massage trainer gave guidance to the parents. Children received both the parent version of the qigong massage and the trainer version during these half-hour sessions. Parents were instructed to perform the qigong massage protocol daily for at least 15 minutes.[64]

To check for group differences, analyses of covariance (ANCOVAs) and MANCOVAs were used with group as the dependent variable and age as the covariate for the pretest scores. There were no statistical differences between the groups before the intervention started for the SSC, (F[1, 39] = 0.004, p = 0.952) or for the PDMS-2 (F[3, 37] = 1.141, p = 0.345).[64] Children in the treatment group showed positive significant improvements on the PDMS-2 for the Locomotion subtest (t = 3.77, p = 0.001) and the Object Manipulation subtest (t = 3.17, p = 0.004).[64] There was no significant difference on the PDMS-2 Stationary subtest or on the SSC for the treatment group. The control group did not demonstrate a statistically significant change for any of the outcome measures. There was no treatment effect for sensory impairment (F[1, 36] = 1.28, p = 0.265); however, there was a significant overall treatment effect for motor development (F[3, 33] = 3.119, p = 0.039).[64] The authors concluded that qigong massage is promising as a home intervention to improve motor skills in young children with CP and DS.[64]

This study is a good example of a caregiver home program that is supervised and guided by a professional. However, the study design has some limitations. The

heterogeneity of the sample makes drawing conclusions on the efficacy of this intervention difficult in infants and toddlers with DS. The sample included children with hypertonicity and hypotonicity with diagnoses of DS and CP. Additionally, the length of the study (5 months) may have been too long to account for maturation effects in young children. Several families dropped out of the study or did not continue to attend the training sessions. The use of the PDMS-2 was a limitation because it is norm-referenced tool without strong psychometrics for children with CP or DS. The GMFM would have been a better choice for assessing the gross motor skills in this sample since it is valid and reliable for children with CP or DS. A different sensory impairment measure may have been a better choice than the SSC, which has not been extensively studied.

Tummy Time

Allowing infants time to play in prone, "tummy time," promotes gross motor skill acquisition.[66,67] Recently, Erin E. Wentz[68] explored the importance of initiating tummy time early in infants with DS. The author recruited 19 infants with DS from birth to 20 weeks old. Infants were divided into an early start group (n = 10), who started the tummy time intervention before 11 weeks of age, and a late start group (n = 9), who started the intervention at 11 weeks of age or later. To include a control group, the author used historical data that was collected on the motor development of infants with DS who did not have a formal tummy time intervention (n = 9). Participants in the early start and late start groups were assessed each month for 12 months. Data from the control group was collected monthly for the first four months and then at 11 months of age. Motor development was assessed using the motor domain (both gross and fine motor) of the

Bayley III. The tummy time intervention was initiated at the baseline visit. Parents in both intervention groups were instructed to perform 90 minutes of supervised tummy time each day and were given a list of activities with pictures that they could choose from. Families also kept a log of daily tummy time and were told to stop actively engaging in tummy time once the infant could transition to and from a sitting position.[68]

Data in this study was analyzed using a repeated-measures ANOVA model. To find overall marginal group differences, the author used a marginal linear model with months as the continuous covariate. She then reanalyzed the data treating time as a categorical variable for the first four months of intervention because a nonlinear trend of group motor development was found over this period. The groups were similar in demographics, except for the mean age in days at study entry (which was the criteria used for group assignment). The early start group demonstrated the highest motor development at each assessment, but the slope declined more steeply (-1.70, $p < 0.001$) than the slope for the late start group (-0.84, $p < 0.001$).[68] The two slopes were significantly different (0.862, $p < 0.001$). The slope for the control group declined significantly more rapidly than slopes of either intervention group. The difference in slope from the control group was (-1.62, $p < 0.001$) for the early intervention group and (-2.48, $p < 0.001$) for the later intervention group.[68] A significant difference between the early start and late start groups was found in months two ($p = 0.001$) and three ($p = 0.004$).[68] There was also a significant difference between the early start group and the control group in months two ($p = 0.023$), three ($p = 0.004$), and four ($p = 0.003$).[68] There was no significant difference between the late start group and the control group at any time. A large effect ($d \geq 0.8$) on motor

development was found at the majority of time points for the early start group.[68] Based on these results, initiating a tummy time intervention before 11 weeks of age appears beneficial for impacting motor development in infants with DS. Additionally, motor development improves the most early in the implementation period. In later months, the benefit of early tummy time diminishes.[68]

This study had several limitations, most notably the small sample size. The author, who performed the assessments for both intervention groups, was not blinded to group assignment. A different physical therapist administered the assessments on the control group. Whether the author gave guidance on the intervention throughout the study as Ulrich suggests was not clear.[15] As discussed earlier, the Bayley III as an outcome measure for motor development may not be the ideal choice for this population. The Bayley III is recommended to identify infants with developmental delays against a normative sample. Although early initiation of a time intervention may be beneficial, more research is needed to clarify if this intervention increases gross motor acquisition over a longer period.

Home Use of Modified Ride-On Cars

The efficacy for ROC use in children with neuromotor disabilities has been mostly been studied in a home environment; however, a search of the literature reveals only one study on home ROC use for children with DS. In a case study of a 16-month-old girl with DS, a family was given a modified ROC for three months, closely supervised and educated weekly by one of the study's investigators.[69] The child and her family were filmed and interviewed before, during, and after the three-month modified ROC

intervention. Weekly education sessions consisted of making sure the modified ROC was safe, teaching parents fun activities to perform with the ROC, encouraging daily home use of the ROC, and filming the child in the ROC. Videos of the child's natural play and ROC play sessions were coded and parents completed a daily activity log, as well as a questionnaire. The PEDI was also administered at baseline, pre-intervention, and post-intervention.[69]

At baseline, the child required assistance and cues to activate the ROC. During the intervention, her independent mobility through use of the ROC increased and she maintained this increase during the retention period. Additionally, the child's total independent driving time increased from baseline (minimum of zero minutes) to intervention (up to six minutes) and declined only slightly during the retention period (between two and four minutes). The child demonstrated an increase in visual attention to the switch from baseline to intervention, but it declined from intervention to retention period. The child's PEDI scores decreased from baseline to pre-intervention, then increased from pre-intervention to post-intervention. The child's family reported that driving the ROC increased her socialization as she drove while on evening family walks in the neighborhood. Other children in the neighborhood learned her name and began approaching her to play.[69]

In this case study, the researchers concluded that daily modified ROC use appears to be feasible, fun, and can increase mobility in the community.[69] Generalizing these results to a broader DS population is difficult because of the single case design. This study also lacked objective outcome measures of gross motor skills; therefore, whether

the child improved her gross motor function is unknown. The study is rich in descriptive detail but lacks statistical significance as well as valid and reliable outcome measures. A larger study with a control group would help determine if ROCs can impact motor development in children with DS.

Orthotics

Children with DS are often prescribed orthoses, either foot orthoses (FOs) or supra-malleolar orthoses (SMOs), to correct for pes planus, which is a common foot deformity associated with DS. Several studies have investigated the efficacy of using orthoses in young children with DS.[70-72] Selby-Silverstein et al[70] explored whether FOs immediately affected the gait of children with DS who had excessively pronated feet. Children with DS between the ages of three and six years old (n = 16) and typically developing children of the same ages (n = 10) participated in a repeated-measures design study.[70] The children with DS were included if they had excessive foot pronation. They were seen for three visits. During the first visit, the children with DS were casted for FOs and measured for anthropometric data. On the second visit, which took place one to four weeks later, gait measurements were collected on the children with DS wearing sneakers only. At this visit, children were also fitted with their FOs and the parents were given a wear schedule to accommodate the children to their new orthoses. One week later, after they had been able to wear the FOs for 5 hours per day on 4 consecutive days, the children with DS returned for a third visit in which gait measurements were collected with sneakers and FOs. Using force plates and motion analysis, the measurements collected included ankle moment, pronation-supination index, foot length contact index,

transverse plane foot angles, and gait velocity. These measurements were also collected on age-matched typically developing children wearing sneakers.[70]

Data was analyzed by comparing the children with DS walking in sneakers with and without FOs using univariate two-way ANCOVA with subject and group as independent variables and velocity as the covariate. Children with DS were compared to typically developing children using a univariate two-way ANCOVA with age and velocity as covariates, trial and group as independent variables, and the measurements from the force plates and motion analysis as dependent variables. Average linear stance velocity was also tested using a two-way ANCOVA with age as a covariate. Trial-to-trial variability was analyzed by finding the coefficient of variation, then compared with a Wilcoxon Matched-Pairs Test for the children with DS with and without FO use, and the Mann-Whitney *U* test for children with DS compared to typically developing children.[70]

Heel eversion was decreased when the children with DS wore FOs in standing ($p < 0.001$).[70] During gait, the transverse plane foot angle decreased with use of the FOs ($p < 0.001$). When the children with DS wore the FOs with sneakers, trial-to-trial variability of pronation-supination index, foot length contact, transverse plane foot angle, and walking speed, all significantly decreased compared to wearing sneakers alone ($p < 0.001$). These findings suggest that FOs had an immediate positive effect on foot alignment in children with DS with excessive pronation, but a negative effect on gait speed.[70]

Most of the measurements in this study reflect the body structure and function level of the ICF model. This study had a strong methodology and adds to the body of

knowledge about whether FOs are beneficial for children with DS. The authors conclude that prescribing FOs may prevent foot pronation deformities in children with DS. More outcome measures at the functional level of the ICF model would be helpful for physical therapists who want to promote meaningful changes in functional activities and participation in their patients.

Functional outcomes of using orthoses in children with DS were investigated by Tamminga et al.[71] In a single-subject, alternating treatment design, the authors investigated the effects of two types of SMOs on gross motor skills in toddlers with DS who were not yet independent walkers. Two children with DS, ages 24 months and 19 months, were recruited to participate in this study. Over a period of six months, the children's gross motor skills were evaluated weekly using dimensions D (Standing) and E (Walking, Running, and Jumping) of the GMFM. The alternating treatment design (A1, B1, A2, B2) included two baseline phases, A1 and A2, which lasted four weeks each, a first orthotic intervention phase, B1, which lasted eight weeks, and a second orthotic intervention phase, which also lasted eight weeks. Two different SMOs were used during the orthotic intervention phases, the SureStep SMO (Midwest Orthotic and Technology Center, Inc, South Bend, Indiana) and the Cascade DAFO #4 Free Plantarflexion Orthosis (Cascade DAFO, Inc, Fern-dale, Washington).[71] One child was randomly assigned the SureStep orthosis for the first orthotic intervention phase and the other child used the DAFO #4. In the second orthotic intervention phase, the children used the orthosis they had not yet worn. During the intervention phases, participants were encouraged to wear

the orthosis for at least six hours daily and parents were instructed to keep a daily log of orthosis use.[71]

Data was analyzed using the two-standard deviation band method to identify significant differences between the scores of the baseline phases and those of the intervention phases.[71] Both children showed a significant improvement in performance in GMFM dimensions D and E when using the SureStep orthosis. Participant 1 also significantly improved with the DAFO #4, but Participant 2 declined in GMFM scores using this orthosis. Furthermore, Participant 1 demonstrated a greater improvement with the SureStep than with the DAFO #4. Both children maintained their newly acquired gross motor skills during the second baseline phase. The authors concluded that both children benefited from orthoses use by acquiring more gross motor skills during orthoses use than would be expected normally.[71]

The study has an interesting design and sound methodology for a single case design, but generalizations to other children with DS cannot be made due to the small sample size. The authors discussed how Participant 2 preferred not to wear the DAFO#4, and her wearing time with this orthosis was less than it was with the SureStep orthosis. The difference in wear times may have accounted for the difference in gross motor skill acquisition for this child. This study examined the use of orthotics in pre-ambulatory children with DS. A different single-subject design, such as a concurrent multiple-baseline design across participants would help inform clinicians of the optimal time to start orthosis use in young children with DS.[73]

Treadmill Training

There is an abundance of evidence to support treadmill training for infants with DS. The earliest evidence of this came from a study by Ulrich et al[74] that examined whether 11-month-old infants with DS (n = 7) who were not independent walkers could demonstrate the ability to produce alternating steps when supported on a motorized treadmill. Infants were held upright with support under their arms by an experimenter, with the infant's feet resting on the treadmill belt. Eight trials were administered, each lasting 30 seconds. The first and last trials were treated as baseline trials with the treadmill stationary. During trials two through seven, the treadmill speed in meters per second (m/s) was set to a slow (0.1m/s), moderate (0.15m/s), or fast (0.2m/s) speed, which was randomized so that the infants all did each of the speed trials twice. Infants had a rest period of two minutes between trials four and five. Infants were videotaped and steps were coded as alternating, single (one leg stepped and the other had non-stepping movements), parallel (both legs stepped simultaneously), double (step-to pattern), or non-stepping leg movements.[74]

The authors analyzed their data using a repeated measures MANOVA to adjust for correlations among trials. Analysis of the videos showed that one infant did not perform any steps during the trials but had a comorbidity of seizure activity and was more developmentally delayed than the other infants. The remaining six infants demonstrated an average of 40.2 alternating steps when the treadmill was moving across the six trials.[74] When the treadmill was stationary, none of the infants took steps.[74] There was a significant main effect of treadmill speed (Wilks' lambda = 0.413, F[3,15] = 7.12,

$p = 0.003$).[74] Post hoc analysis revealed a significant difference in number of steps taken between the stationary treadmill and the moving treadmill, but not significant difference in number of steps between the three speeds of the moving treadmill. Infants produced a much higher number of alternating steps than of any other type of step or leg movement.[74]

The results of this study indicate that infants with DS have the ability to take alternating steps when supported on a treadmill before they are able to stand or walk independently.[74] In fact, follow up interviews indicated that the infants did not take independent steps until an average of 13.3 months after the testing was done.[74] This initial research led to many more treadmill studies in infants with DS, but the authors did not assess carryover of the treadmill intervention.

In 1995, Ulrich et al[75] investigated the change over time in the ability of infants with DS to produce alternating steps when supported on a treadmill. Participants with DS ($n = 9$), ages eight to 11 months old participated in monthly test sessions, which took place in the infants' homes. The infants continued with their regular therapies and no additional interventions were administered. The test session protocol was the same as the 1992 Ulrich et al study.[74] Outcome measure testing continued each month until the infant was able to produce consistent alternating steps during three consecutive testing sessions. In addition to videos, the authors also assessed anthropometric variables and gross motor skills using an earlier version of the Bayley III.[75]

An abstract by Ulrich et al[2] published in 2001 explored whether practice stepping on a motorized treadmill would accelerate the onset of walking in infants with DS. The

researchers randomly assigned 30 infants with DS to either a control group or an experimental group. All participants continued with their regular physical therapy. The infants in the experimental infants participated in treadmill training for 8 minutes per day, 5 days per week on a specially made small treadmill that was kept in the home. Parents were trained on how to support the infant over the treadmill. The infants in both groups were tested on their gross motor skills every two weeks using an earlier version of the Bayley III. The participants entered the study when they could sit unsupported for 30 seconds. The testing concluded when the infant could walk independently.[2]

The length of time from independent sitting to pulling to stand, walking with help, and independent walking was compared between the two groups. Infants in the experimental group walked with help significantly sooner (73.8 days) than infants in the control group.[2] The experimental group also walked independently significantly faster (101 days) than the control group.[2] There was no significant difference between the groups in the amount of time from independent sitting to pulling to stand. The authors conclude that a treadmill training home program administered by parents and supported by therapists can accelerate the rate at which infants with DS learn to walk. This pilot study led to further research to improve the protocol.[2]

In 2007, Wu et al[17] investigated two different treadmill protocols for infants with DS (average age approximately ten months old). In their study, participants were randomly assigned to a lower-intensity-generalized (LG) group (n = 15) or a high-intensity-individualized (HI) group (n = 15). A control group (n = 15) was from the previous Ulrich et al[2] study in which infants with DS did not receive a treadmill

intervention. The treadmill intervention was similar to previous research in which parents were provided a small motorized treadmill and performed the intervention at home with training from the researchers. Parents held the infants around their trunk and under their arms, off-loading some of their body weight while the treadmill moved their feet backward causing the infants to produce a stepping pattern. Participants in the LG group received treadmill training at a constant speed of 0.18m/s for six minutes per day, five days per week. The HI group also received treadmill training 5 days per week, but with an individualized protocol based on the step frequency produced by each infant. For the HI group, treadmill speed and training duration were individually progressed, and small ankle weights were also attached to the infants' feet during training. Participants in all groups entered the study when they were able to produce six steps per minute with support on a treadmill (about 10 months old) and the study concluded when they could take three independent steps over ground without help. Outcome measures included walking onset (the ability to take three independent steps over ground without help), and gait parameters measured at 1 month and 3 month follow-up.[17] Gait parameters included average velocity, stride length, step width, stride time, stance time, and dynamic base.[17]

The mean age at walking onset was 19.2 months for the HI group, 21.4 months for the LG group, and 23.9 months for the control group.[17] The HI group was significantly younger than the control group for walking onset ($p = 0.011$).[17] A MANCOVA was conducted for the gait parameters. The three groups differed significantly in overall gait patterns between one month and three months post walking onset (Wilk's lambda = 0.447, $F[12,50] = 2.07$, $p = 0.037$).[17] Post-hoc analysis showed

that stride length significantly differed between the three groups (F[2,35] = 3.76, p = 0.033), with the HI group producing a significantly longer stride length than the control group (p = 0.03).[17] There were no significant differences in gait parameters between the LG and HI groups.[17]

This study indicates that an individualized approach with increasing intensity may be more beneficial for accelerating walking onset in infants and toddlers with DS. The authors did not measure overall gross motor development in the participants, which may have been helpful for determining if treadmill training affects other gross motor skills. This study did not find a significant difference in the age of the participants for onset of walking between the LG group and the control group; however, the Ulrich et al[2] study did find a difference when comparing the control group to an experimental group with a similar protocol as the LG group. The difference was that the experimental group in the Ulrich et al[2] study performed treadmill training for eight minutes per day, whereas the LG group of the Wu et al[17] study performed the treadmill training for six minutes per day. This 30% increase in practice per day may account for significant effects found in the previous study. The results of both studies support the theory that increased practice and repetition may lead to improved outcomes.

In another study using the same cohorts of HI and LG groups, Wu et al[76] used long-term follow-up to explore how newly walking toddlers with DS adopted clearance strategies to negotiate an obstacle in their travel path. The authors were able to complete a 1-year follow-up on 26 infants (13 infants in each group with an average age of about 10 months) to include four visits: immediately after the treadmill intervention when able

to take eight continuous steps independently and at three, six, and 12 months later. They were videotaped, and gait parameters were recorded using the GAITRite mat (CIR systems, Havertown, PA). At each session, obstacles of various heights were placed on the floor in the infant's path, with four trails collected for each obstacle height during each follow-up visit.[76]

The authors analyzed the videos using categories of obstacle negotiation to include *refusal*, *crawl*, *fall*, and *walk over*. Gait parameters of velocity, cadence, step length, and step width were calculated using the GAITRite system. Repeated measures ANOVA with post-hoc analysis were used to evaluate the results. The HI group produced a significantly higher percentage of *walk* strategy and a lower percentage of *crawl* strategy than the LG group at the three-month follow up visit ($p < 0.05$) but both groups *walked* significantly more than *crawled* over the object for the six and 12-month follow up visits ($p < 0.05$).[76] Both groups showed consistent anticipatory locomotor adjustments, such as decreased velocity, cadence, and step length, when approaching the obstacle. The authors concluded that the HI group was able to walk over the obstacle earlier than the LG group because of retention effects from their treadmill training protocol.[76]

A study by Angulo-Barruso et al[32] used the same cohorts of HI and LG infants with DS (average age of 10 months), measuring their physical activity. Activity monitor accelerometers (Actiwatch, Respironics/Mini Mitter, Bend, OR) were placed on the infants' trunks above the right iliac crest using medical tape and around their right ankles for 24 hours every other month during the intervention phase. The activity monitors were also placed on the infants in the same locations for 24 hours at the four follow-up visits

(independent walking at least eight steps, and at three months, six months, and 12 months later).[32]

The 24-hour activity data was analyzed and categorized as moderate-to-vigorous activity (Highact) or sedentary-to-light activity (Lowact) using criterion values or thresholds for each individual infant. Data was divided into two phases: activity during the treadmill intervention, and post-intervention follow-up. Results indicated that infants in the HI group had higher levels of Highact than infants in the LG group during the intervention phase. As the intervention continued, the HI group had an increase in Highact in the final sessions, but Highact stayed about the same in the LG group throughout the intervention. Additionally, infants in the LG group had higher levels of Lowact than infants in the HI group. Highact tended to increase over the post-intervention follow-up phase visits, but the HI group retained significantly more Highact and less Lowact than the LG group throughout the post-intervention follow-up.[32]

This study lacked a control group for comparison, therefore whether the LG group demonstrated more Highact than would an infant who did not receive treadmill training is unknown. The results indicate that a higher-intensity individualized approach to treadmill training has a positive effect on physical activity in infants with DS. Additionally, this increase in physical activity appears to be retained over time.

To explore the relationship of early physical activity and the acquisition of independent walking in infants with DS, Lloyd et al[77] performed a longitudinal study using the same cohort of infants (average age of 10 months old) with DS performing HI and LG treadmill training and wearing activity monitors.[32] The authors placed the activity

monitors on the infants for 24 hours every other week from the start of the study until the children were able to talk independently for at least seven bimonthly sessions.[32,77] The Highact and Lowact intensity definitions from the previous study were used in this study as well.[32,77]

Six linear regression models were used to analyze whether early activity is related to walking onset in infants with DS using the first three time points of physical activity (at an average of ten, 12, and 14 months) as predictors for age of walking onset.[77] Age of walking was the dependent variable and the six independent variables included trunk total activity, trunk Highact, trunk Lowact, LE total activity, LE Highact, and LE Lowact. Regression analysis was also performed using the same independent variables at each time point as predictors of onset of independent walking. Height, weight, and Bayley raw motor score at each time point were also included in the regression model. Results indicated that increased total LE activity was a significant predictor of earlier onset of walking at an average age of 12 months ($p = 0.04$) and 14 months ($p = 0.02$).[77] Further regression analysis revealed that LE Highact explained a significant amount of the variance ($R^2 = 0.349$, $p = 0.22$) for the onset of independent walking.[77] None of the other five independent variables were significant predictors. Additional regression analysis revealed that increased LE Highact at an average age of 12 and 14 months was a good predictor of walking onset in children with DS, regardless of intervention group (HI or LG).[77]

The infants in this study were all participating in treadmill training at home. No control group was included. Therefore, the results of the study cannot be generalized to

all infants with DS. However, the results indicate that in infants with DS, more activity in the LEs at an average of 12 and 14 months is related to earlier onset of independent walking. The authors recommend promoting early LE activity in infants with DS.[77]

Ulrich et al[16] published another study investigating the effects of HI and LG treadmill training with a similar approach as previously discussed; however, in this study, the authors used the original protocol of 8 minutes per day for 5 days per week with a treadmill speed of 0.15m/s for the LG group.[16,17] The HI group had individualized training with gradually increasing time per day and speed of treadmill, as well as a gradual increase in ankle weights while training. Infants were randomly assigned to HI (n = 16) or LG (n = 14) groups, with six infants lost to attrition. The researchers tested the infants on an earlier version of the Bayley III every other week to test for gross motor function. During this biweekly visit, the researchers also video recorded the caregiver and child during treadmill training.[16]

The videotapes of the sessions were coded for frequency of alternating steps per minute, and the average of alternating steps per minute over a two-month time span was calculated. A *t*-test was conducted using the frequency of steps per minute from the first visit to determine if there was a difference between the two groups at study onset. Additionally, a repeated measure ANOVA was used to compare alternating step frequency over time using five time points across two months for each child. The Mann-Whitney *U* test was conducted to test for group differences in age of onset for each motor milestone. The groups were not significantly different in frequency of alternating steps taken at study entry. The repeated measures ANOVA revealed that the number of

alternating steps per minute at study entry to onset of independent walking was significant for time ($p < 0.0001$) and interaction effects ($p < 0.05$) for the HI group.[16] Infants in the HI group acquired gross motor skills at an earlier age than infants in the LG group, but only two items on the Bayley outcome measure achieved statistical significance ($p \leq 0.05$).[16]

The results support the use of treadmill training as a home-based intervention for infants with DS. Both the HI group and the LG group showed an increase in frequency of alternating steps from the beginning of the study until the onset of independent walking. The HI group attained gross motor skills earlier than the LG group. Although this study replicates and expands on previous research, the use of the earlier version of the Bayley III may have been a limitation to this study. The Bayley was intended to identify gross motor skill delay in typically developing infants and has not been tested for reliability or validity in infants with DS. The GMFM would have been a more accurate outcome measure for this population in assessing gross motor skills. Like the previous LG and HI study, too many variables may have been manipulated in the HI group. The authors increased the treadmill speed, time spent during daily training, and ankle weights in the HI group participants.[16] Whether manipulating just one of these variables may have produced similar results, or whether it is necessary to manipulate all three of these variables to obtain similar results, is unknown.

Wu et al[78] performed a follow-up evaluation of the children from the Ulrich et al[16] study. Children with DS in both the HI group and LG group returned to the clinic immediately after the intervention ended, and at three, six, and 12 months later. Four

children were lost to attrition; therefore, 26 participants completed the study. Kinematic patterns of the hip, knee, and ankle joints were measured to determine if there was a difference between the two interventions. The authors compared the timing and magnitude of peak extension and flexion at the hip, knee, and ankle joints, as well as abduction and adduction at the hip joint.[78]

To analyze the data, the authors used two-way repeated measures ANOVAs with peak timing and magnitude at each joint as the dependent variables. Independent variables were two groups and four visits. Overall, the authors found that both the HI group and the LG group significantly advanced in the development of joint kinematics during the follow-up period, with significant effect of visit for all joints. The HI group had a significant effect for group for two joints, with an increased timing of peak ankle plantar flexion before toe-off and increased duration of forward thigh swing after toe-off. The authors concluded that the HI group demonstrated an accelerated development of joint kinematic patterns compared to the LG group one year after walking onset.[78]

Although this study provides valuable information about development of joint kinematics in children with DS, clinical application is unclear. The evidence suggests that children in the LG group develop the same joint kinematics eventually, but there was no comparison of the two treadmill groups to a control group. This study indicates that an individualized, more intense home treadmill program is overall more beneficial for infants with DS. When implementing a home program, physical therapists should continue to monitor and update the program for their patients.

In an extension of previous treadmill training research, Looper and Ulrich[79] incorporated orthosis use into the lower-intensity treadmill training protocol to investigate the impact on gross motor skill acquisition compared to treadmill training alone. In an entirely separate cohort, infants with DS who were able to pull themselves into a standing position independently but unable to take steps and who had no prior history of orthosis use were included in the study. Children's ages ranged from one to just over two years old at study onset. They were randomly assigned to a control group (n = 12) or an experimental group (n = 10). Five children dropped out of the control group, so the final analysis included seven children in the control group and ten in the experimental group. The children who were in the experimental group were fitted for SMOs and were instructed to wear them for 8 hours per day at least five days per week throughout the study period. GMFM measurements were taken monthly from the start of the study until the end of one month of independent walking. Lower-intensity treadmill training was performed at home, as described by Ulrich et al,[2] and was stopped once the children could take three independent steps over ground.[79]

Data analysis included independent *t*-tests. The authors found that the experimental group had a higher overall GMFM score at study entry, so this was included as a covariate in the mixed linear models. Both groups significantly improved GMFM scores over time ($p < 0.001$) but there was no significant group difference in overall GMFM score. Children were tested one month after walking onset at the study's conclusion. The control group scored significantly higher on the GMFM than the experimental group ($p = 0.01$) with a large effect size ($d = 1.55$).[79] Group by time

interactions demonstrated that the predicted developmental trajectories for dimension C and D of the GMFM displayed a rapid increase in scores for the control group, followed by a levelling out. The experimental group showed a more linear improvement.[79]

The results indicate that SMO use with treadmill training does not significantly improve gross motor skills more than treadmill training alone. Established treadmill protocols in infants with DS start the treadmill training as soon as the infant is able to sit up independently. This study did not start the intervention until the infants were able to pull to stand independently, which occurred at an average age of 20.5 months.[79] The study would have been strengthened by the addition of an orthosis-only group that did not receive treadmill training. Although rigidity and thickness of orthoses can vary, the authors did not control for the type of SMO used. Another limitation of the study was that the GMFM rater was not blinded to group assignment, which may have biased the results.

DYNAMIC STANDING DEVICE

A dynamic standing device, the Upsee, shows potential as a tool for home programs that can be used by parents of infants and toddlers with DS. However, this device has not yet been studied in this population. The few studies that have been published on the efficacy of the Upsee shows promise that upright mobility through dynamic standing may impact gross motor function in children with neuromotor disabilities. All of the available literature on the efficacy of the Upsee is in the form of a case study.

Ardolino and colleagues[18] investigated home use of the Upsee in two children with neuromotor disabilities who exhibited hypotonicity of the trunk. Child one was 24

months old with a diagnosis of agenesis of the corpus collosum and chromosome three deletion. Child two was 21 months old with a diagnosis of CP, GMFCS level V.[18] The parents of the children were fitted for an Upsee to use at home. They were encouraged to use the Upsee four days per week progressing up to 20 minutes per day for a period of three months, keeping a daily log of time and activities performed. The parents were educated on dynamic weight bearing to encourage their children to be active through their legs and trunk while using the device. The children were tested on their trunk control using the SATCo and gross motor function using the GMFM before and after the intervention. Child one improved with his SATCo score, but child two did not show a change in trunk control. Both children improved in their GMFM scores beyond what would be expected with natural progression.[18,80] The authors suggest that dynamic standing through use of the Upsee may be beneficial for improving gross motor skills and trunk control in children with neuromotor disorders who exhibit trunk hypotonia.[18]

Another case study by Casey et al[19] examined a more intense home program using the Upsee. The child in this case was a 4-year-old boy with Pitt Hopkins syndrome.[19] Caregivers at a local day care used the Upsee 5 days per week for 1 hour each day for a period of three months. The child was encouraged to be active while using the Upsee including head control, active weight bearing, communication, and social interaction with peers. GAS was evaluated prior to the intervention and every four weeks until the end of the intervention (at four, eight, and 12 weeks). Follow-up testing occurred 3 months after the completion of the intervention (24 weeks). For the GAS, the parents and caregivers were involved in setting realistic, individual goals in three separate domains: physical

activity, health and wellness, and social interaction. The score for the GAS ranged from -2 (much less than expected outcome) to +2 (much better than expected outcome, with a score of 0 as the expected outcome. The child improved to +1 or +2 in all three of his goals over the three month intervention period, but returned to baseline during the follow-up period.[19] Although gains were not maintained over time, the case study highlighted comments by parents and caregivers that the child was happy, smiling, and participating more when using the Upsee. They also noted that the child improved in socialization and physical activity that was not reflected in the GAS. This case study illustrates that upright mobility can improve socialization in a child with participation restrictions.[19]

Barria et al[81] describe a case series in which four children with CP classified as GMFCS levels IV and V participated in clinical use of the Upsee 3 days per week for 20 minutes each day, for 15 sessions. The protocol for using the Upsee was not described, so whether the child was actively using the device or whether the therapist was passively walking with the child in the device is unclear. The researchers measured heart rate, LE passive range of motion at all joints, and LE spasticity. They took these measurements before and after each session. Results were analyzed via *t*-test to compare the mean differences before and after each session. There were no significant changes in spasticity or range of motion. There was a significant increase in mean heart rate after the Upsee sessions in all four children, $p \leq 0.01$.[81] The authors conclude that use of the Upsee may improve cardiovascular health in children with severe CP, but it does not appear to have an impact on range of motion or spasticity.[81]

Fergus[82] describes a case in which home use of the Upsee combined with kinesio-taping was performed for 24 weeks in a 31-month-old child with CP classified as GMFCS level III. The child attended weekly physical therapy sessions for 16 weeks and was given a home program of kinesio-tape and Upsee. The child's mother was encouraged to use the Upsee daily, progressing from 10 minutes per day up to 45 minutes per day. Kinesio-tape was applied to the child's feet to prevent excessive inversion. The mother was educated on normal gait characteristics to apply while using the Upsee with her child. These included arm swing, normal base of support, and symmetrical step length. Outcome measure included the GMFM, Modified Pediatric Activity Log (mPMAL), and gait characteristics (coded by two raters based on videos of the child). The mPMAL is a tool for assessing how often, how well, and how willing the child walks under various conditions. Psychometrics of the mPAL have not yet been examined. Testing occurred during a preintervention and postintervention session. The child demonstrated a decrease in gait deviations from pretest to posttest when the child was ambulating with a walker, requiring no assistance on even surfaces. She also had an increase in GMFM and mPMAL scores. The mother reported that use of the Upsee was enjoyable for both her and the child. The researcher concluded that home use of the Upsee with kinesio-taping might improve gait characteristics and overall function.[82]

Taken together, these case studies demonstrate that use of the Upsee may be beneficial in children with neuromotor disabilities. None of the children in these studies had a diagnosis of DS. All four of these case studies or series have the limitation of small sample size and no control group. Recruitment and retention can be a problem when

researching the effectiveness of any intervention in pediatric participants. Case studies present valuable information, but they do not provide the rigor of a randomized controlled trial. One solution to consider is single subject research design in which participants act as their own control.[21,22,73]

CONCLUSION

In infants and toddlers with DS, there is a paucity of research investigating measurements of trunk control and the relationship of trunk control to gross motor function. The SATCo shows promise as an outcome measure for trunk control in this population. The GMFM is the gold standard for measuring gross motor function in children with DS. Because children with DS develop their gross motor skills at a slower rate than typically developing children, early physical therapy intervention is critical. Supervised home programs that focus on attaining gross motor skills and trunk control through upright mobility have the potential to increase the acquisition of gross motor function in infants and toddlers with DS. Treadmill training appears to be beneficial in children with DS. However, use of a dynamic standing device, the Upsee, may be a more cost-effective solution to upright mobility with a difference of approximately $1000. The effectiveness of the Upsee has not been studied in children with DS.

The purpose of this dissertation is to investigate trunk control in infants and toddlers with DS. The first study will investigate the reliability of the SATCo in infants and toddlers with DS. The second study will examine the validity of the SATCo with the GMFM and determine whether age and SATCo score predicts GMFM score in infants and toddlers with DS. The third study will explore the impact of a dynamic standing

device (Upsee) on trunk control and motor skills in infants and toddlers with DS. Overall, this dissertation will provide physical therapists with a better understanding of trunk control in infants and toddlers with DS.

CHAPTER III

RELIABILITY OF THE SEGMENTAL ASSESSMENT OF TRUNK CONTROL (SATCo) IN INFANTS AND TODDLERS WITH DOWN SYNDROME

INTRODUCTION AND PURPOSE

DS occurs in approximately 12.6 per 10,000 live births in the United States and is a common cause of developmental disability.[1] During early development, children with DS perform gross motor milestones at a slower rate than their typically developing peers. Most typically developing infants can sit without support at approximately six months of age and walk independently by 12 to 15 months of age; however, infants with DS show gross motor skill delay, attaining independent sitting at around 11 months and independent walking at approximately 24 months.[1-4] Trunk control is a precursor to upright gross motor skills.[5] Many infants with DS display trunk hypotonicity with decreased trunk strength and postural control.[83] An outcome measure to assess trunk control in infants and toddlers with DS is needed for evaluative purposes and to assess the effectiveness of physical therapy interventions.

Standardized tools specifically for infants and toddlers with DS are limited. Physical therapists should employ effective, reliable tools to measure the incremental changes that occur in infants and toddlers with DS at various levels of the World Health Organization's ICF.[6,84] The GMFM has been shown to be a reliable and valid measure of motor skills at the activity level of the ICF for children with DS.[7-9] However, there is a paucity of research on outcome measures of trunk control at the body structure and

function level of the ICF for infants and toddlers with DS. The SATCo shows promise as a quick and easy tool that can be employed by pediatric physical therapists in any setting to measure changes in trunk control in infants and toddlers with DS.[10]

Initial psychometrics indicate the SATCo is a good tool for measuring discrete levels of trunk control in different populations [10] Reliability and validity of the SATCo was originally determined by rating eight typically developing children and 24 children with neuromotor disability.[10] Twenty-one of the 24 children with neuromotor disability were diagnosed with cerebral palsy in Gross Motor Function Classification Scale (GMFCS) levels I through V; however, none of the children in the sample had a diagnosis of DS.[10] Intrarater reliability for the SATCo was excellent (ICC = 0.98) across all data sets and all aspects of control; and interrater reliability was good (ICC = 0.8 to 0.84).[10] Concurrent validity has been established with the GMFM Dimension B (r = 0.73 to 0.83) and with the sitting section of Alberta Infant Motor Scale (r = 0.86 to 0.88).[10] The study established that the SATCo is a reliable scale of trunk control for typically developing children and those with neuromotor disability.

Hansen et al[11] examined the reliability of the SATCo in children with CP across all GMFCS levels for intrarater inter-day, interrater inter-day and live versus video reliability (ICC $\geq$ 0.9); and Cardoso de Sa et al[12] studied the interrater reliability of the SATCo for children with Duchenne muscular dystrophy using the kappa statistic (κ = 0.90, 95% CI = 0.83, 1.00). Both articles found the SATCo to be a reliable outcome measure for these populations.[11,12] To date, the psychometric properties of the SATCo have not been analyzed in children with DS.

The purpose of this study is to determine the interrater, intrarater, and live versus video reliability of the SATCo in infants and toddlers with DS.

METHODS

Study Design

A repeated measures methodological reliability study on a single group was conducted. The study was approved by the Institutional Review Boards of Texas Woman's University and the University of St. Augustine for Health Sciences. Written informed consent was obtained from the parents of all the participants prior to testing.

Participants

To obtain a kappa statistic of 0.8 at 90% power, a sample size of 17 participants is required to detect statistical significance at $p \leq 0.05$ for reliability studies.[20] Therefore, a sample of 18 children with DS were recruited from local parent support groups, early intervention programs, and outpatient clinics. To participate in the study, participants had to be between the ages of six to 24 months, have a diagnosis of DS, and speak and understand English. Participants were excluded from the study if they had a diagnosis unrelated to DS that limited gross motor movement, or medical restrictions that contraindicated movement or handling. The age range for the participants was selected to capture children who were still developing sitting balance, as well as those who had mastered the skill.

Instrument

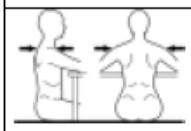
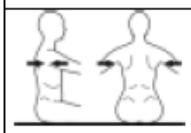
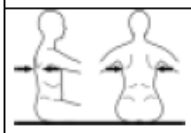
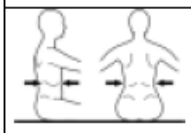
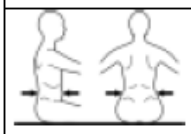
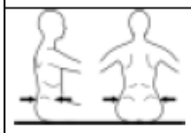
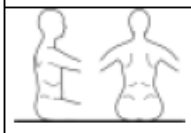
The SATCo was designed and refined by Butler et al[10] as a dichotomous scale to assess discrete levels of trunk control in children with neuromotor disabilities. To

complete the assessment, the examiner assists the child into sitting on a low bench with his or her feet on the floor. The child's pelvis is secured in a neutral position. This can be done either with a positioning strap or with the assistance of a second person. From behind the child, the examiner gives manual support laterally at specific landmarks around the child's trunk. Starting at the shoulders, the examiner moves segmentally down the child's trunk through seven levels.[10] At each level, the child is tested on static trunk control (ability to hold head in neutral for five seconds), active trunk control (ability to turn head left and right), and reactive trunk control (ability to maintain control with perturbations at the sternum, thoracic spine, and at each acromion). The child's arms cannot be used to maintain sitting balance or upright posture. The first level of the SATCo, in which the examiner's hands support the child at the shoulders, only tests static and active trunk control. As the examiner's hands move down the trunk, levels two through seven of the SATCo test static, active, and reactive trunk control. A higher level score indicates the examiner is giving less support and the child is displaying more trunk control. The child receives a score of present (✓), absent (-), or not tested (NT) for each item tested. Credit is given for static control if the child can maintain neutral posture above the level of support given by the examiner. Active control is present if the child can maintain neutral posture during head movement. The child is given credit for reactive control if the trunk posture is maintained or quickly regained during an external perturbation.[10]

For the purposes of this study, children were given one point for each item that control was "present" for a possible total item score up to 20. Figure 1 shows the SATCo

scoring form and levels. As an example, if the child was able to maintain static, active, and reactive control at the shoulder girdle, axillae, and inferior scapula, but could not maintain control for any items from the lower ribs down, the child would receive an item score of eight. The child's level of trunk control is the highest level at which he or she can maintain control in all items for that level. A child who demonstrates only head control receives a SATCo level score of one. In the example above, the child who got an item score of eight would receive a SATCo level score of three, as this was the highest level he or she was able to maintain static, active, and reactive control. Higher levels indicate better trunk control, indicating less support needed from the examiner. Level seven is the highest, indicating the child can maintain all aspects of trunk control with no manual support.[10]

Figure 3.1. SATCo Score Sheet[10]

Identification #: Tester Name: Date:	**Level of Manual Support** Pelvic/thigh strap used except as indicated	**Functional Level** Arms and hands in air except as indicated	**Static**	**Active**	**Reactive**	**Comments**
			Maintain vertical neutral position of head and trunk above manual support level			
			minimum of 5 seconds	while turning head with arms lifted	Maintain/quickly regain following brisk nudge	
	Shoulder girdle Testers hand position may vary from horizontal	**Head control** Arms may be supported throughout			**NOT Tested for Head Control**	Level 1
	Axillae	**Upper Thoracic Control**				Level 2
	Inferior scapula	**Mid Thoracic Control**				Level 3
	Over lower ribs	**Lower thoracic Control**				Level 4
	Below ribs	**Upper lumbar Control**				Level 5
	Pelvis	**Lower lumbar Control**				Level 6
	No support given and pelvic/thigh straps removed	**Full trunk control**				Level 7
Fixed spinal deformity? Yes_____No_____Comments_____ Limitation of Cervical Rotation Left_____Right_____Comments_____						

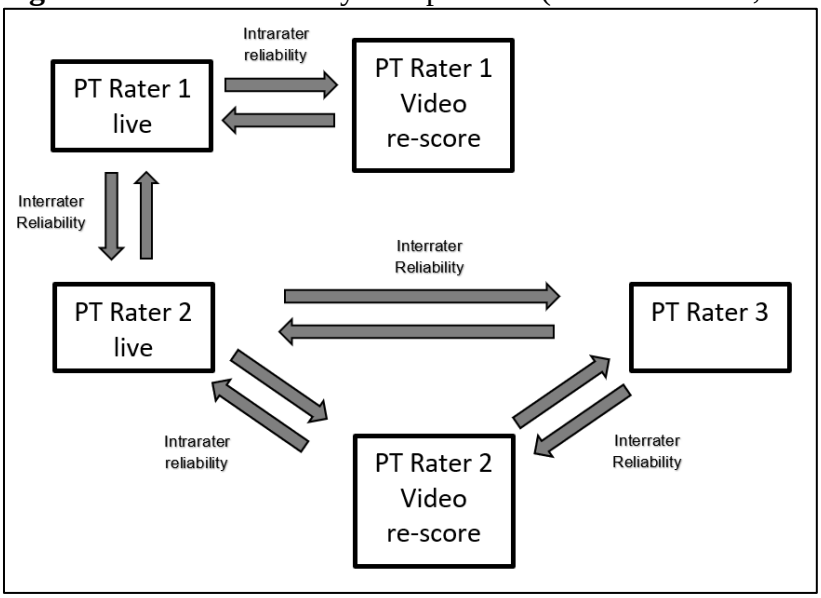
Procedure

Three experienced physical therapist raters (PT raters) with at least five years working in pediatric physical therapy participated in data collection. Prior to recruiting child participants, the PT raters were given the SATCo protocol, score sheet, and

supporting literature to review. The PT raters attended a training session with the principal investigator to ensure consistency of scoring. At the training session, the principal investigator explained the outcome measure and the PT raters watched a video demonstration of the SATCo. The principal investigator and the PT raters discussed common testing errors and possible compensatory strategies that children with DS might employ. The PT raters demonstrated their competence with the SATCo by performing and scoring the outcome measure on a live volunteer child with DS.

Each child participant was tested on the SATCo by two different PT raters (PT Rater 1 and PT Rater 2). Testing occurred on one day with a 30-minute separation between SATCo testing sessions. Each PT rater was randomly assigned to testing during the first or second session. To ensure consistency of testing, the participants were tested in the same environment, using the same research assistants, equipment, and toys. Research assistants provided visual stimulus with a colorful toy to encourage the child to turn his or her head during the active control portion of the test and gentle nudges during the reactive control portion of the test. A research assistant also assisted in securing the child's pelvis during the test. Each PT rater tested the child on the SATCo through all seven levels (as long as the child was safe), which lasted about five to 10 minutes. Children were video recorded from the front and the side during each testing session. After a period of at least two weeks, PT Raters 1 and 2 re-scored their own SATCo testing sessions by watching themselves test each participant in the video recordings. PT Rater 3 did not perform live testing but watched and scored the videos for PT Rater 2. Figure 3.2 shows the reliability comparisons for this study.

Figure 3.2. Five Reliability Comparisons (Three Interrater, Two Intrarater)



Data Analysis

For data analysis, each item in the SATCo was given one point if control was considered "present." The columns were summed to reveal a score for static control (maximum score = 7), active control (maximum score = 7), and reactive control (maximum score = 6). The total SATCo item score was also obtained, from zero to 20, and the level of trunk control was identified, from zero to seven.

Five comparisons were made among the three PT raters. Interrater reliability was assessed by comparing the live independent ratings of PT Raters 1 and 2, live rating of PT Rater 2 versus video rating of PT Rater 3, and video ratings of PT Raters 2 and 3. To assess intrarater reliability, video recordings were re-scored by Raters 1 and 2 at least two weeks after the testing session to minimize recall bias.

Data was entered into a spreadsheet and analyzed using IBM SPSS Statistics 25 software.[§] Because of the ordinal nature of the SATCo, Cohen's Kappa was used to assess reliability for each category (static, active, and reactive), overall total score, and SATCo level of trunk control. A *p*-value < 0.05 and 95% confidence intervals (CI) not containing the null value of zero were used to indicate significance. Cohen's Kappa reliability results were interpreted as no agreement (< 0.2), minimal agreement (0.2 to 0.4), weak agreement (> 0.4 to 0.6), moderate agreement (> 0.6 to 0.8) strong agreement (> 0.8 to 0.9), and almost perfect agreement (> 0.9).[85] To be consistent with previous studies of the SATCo[10,11,86,87], reliability was also calculated using Intraclass Correlation Coefficient (ICC) (2,1) estimates with 95% CI based on a single measure, absolute consistency, two-way random effects model. ICC reliability results were interpreted as poor (< 0.5), moderate (0.5 to 0.75), good (> 0.75 to 0.9), and excellent (> 0.9).[37] Bland-Altman plots were generated for each reliability comparison. Because the ICC is a more robust analysis of reliability, and to be consistent with previous studies of the SATCo, reliability results using the ICC will be discussed in more depth.

RESULTS

Eighteen children with DS (six female and 12 male) between the ages of six months to 23 months, with a mean age of 13.67 months (*SD* = 5.31) participated in the study. Demographics for the participants are in Table 3.1. Trunk control was assessed using the SATCo by three PT raters in live and/or video-recorded sessions. The three PT

[§] IBM Corporation, Armonk, New York

raters were all female with an average age of 41 years (*SD* = 7.94), with experience in pediatric physical therapy clinical practice for an average of 11.33 years (*SD* = 7.09).

Table 3.1. Participant Demographics

Participant Number	Gender	Race/ Ethnicity	Age (months)
1	F	Caucasian	6
2	M	Latino/a	18
3	F	Black/ African American	6
4	M	Caucasian	8
5	M	Caucasian	10
6	M	Caucasian	15
7	F	Caucasian	17
8	M	Caucasian	11
9	M	Caucasian	23
10	M	Latino/a	19
11	M	Caucasian	17
12	M	Black/ African American	11
13	M	Caucasian	15
14	M	Caucasian	18
15	M	Caucasian	8
16	F	Latino/a	18
17	F	Latino/a	19
18	F	Caucasian	7

The interrater and intrarater reliability results are displayed using Cohen's Kappa in Table 3.2. The data for reactive trunk control for Rater 1 live versus Rater 1 video intrarater reliability had a confidence interval that indicated it was not significant (κ = 0.27, 95% CI = -0.01, 0.54). All other data points were significant as indicated by 95% confidence intervals not crossing zero. The strongest Cohen's Kappa agreement between raters was for the category of static trunk control with all $\kappa \geq 0.68$. Agreement was weakest for category of SATCo level (κ = 0.24 to 0.59). Overall, interrater reliability

varied from minimal to moderate (κ = 0.33 to 0.68) and intrarater reliability was minimal to strong (κ = 0.24 to 0.87).

Table 3.2. Reliability of the SATCo Using Cohen's Kappa

	Static κ [95%CI]	**Active** κ [95%CI]	**Reactive** κ [95%CI]	**Total Score** κ [95%CI]	**SATCo Level** κ [95%CI]
Interrater Reliability (Rater 1 vs Rater 2)	0.68 [0.43,0.92]	0.55 [0.28,0.82]	0.33 [0.05,0.62]	0. 36 [0.11,0.60]	0.42 [0.14,0.69]
Interrater Reliability (Rater 2 live vs Rater 3 video)	0.68 [0.43,0.92]	0.42 [0.14,0.69]	0.67 [0.41,0.92]	0.55 [0.28,0.82]	0.47 [0.22,0.72]
Interrater Reliability (Rater 2 video vs Rater 3 video)	0.74 [0.51,0.97]	0.48 [0.21,0.76]	0.68 [0.43,0.82]	0.48 [0.21,0.76]	0.47 [0.22,0.72]
Intrarater Reliability (Rater 1 live vs Rater 1 video)	0.74 [0.51,0.97]	0.55 [0.28,0.82]	0.27 [-0.01,0.54]	0.29 [0.02,0.56]	0.24 [0.02,0.46]
Intrarater Reliability (Rater 2 live vs Rater 2 video)	0.87 [0.70,1.00]	0.74 [0.51,0.97]	0.87 [0.69,1.00]	0.68 [0.43,0.92]	0. 59 [0.35,0.83]

ICC (2,1) results are presented in Table 3.3. Interrater reliability was moderate to good among all three raters with ICC values between 0.5 and 0.9 ($p \leq 0.013$). The highest reliability scores were obtained when PT raters re-scored their own videos taken from live testing sessions, with good to excellent intrarater reliability of the SATCo ($p < 0.001$). Among raters, the interrater reliability between Rater 1 and Rater 2 reflected the lowest overall scores (ICC (2,1) ≤ 0.686). Highest scores were found in the intrarater reliability of Rater 1 (ICC (2,1) ≥ 0.806). When comparing live rating versus video recording, the interrater reliability of the SATCo remained moderate for most categories but improved to good reliability for static control and overall total score. Across all raters, the category of static trunk control (ICC (2,1) = 0.647 to 0.922) and total SATCo score

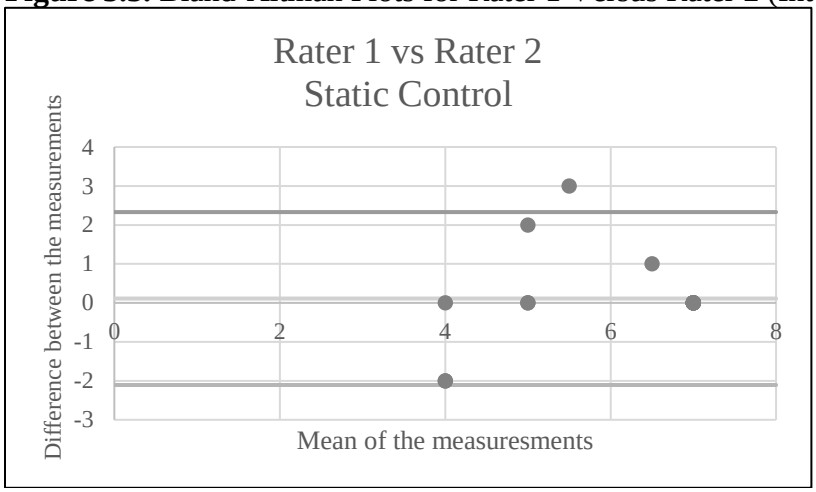
(ICC (2,1) = 0.661 to 0.941) showed the strongest reliability. The lowest reliability was reactive trunk control (ICC (2,1) = 0.508 to 0.846).

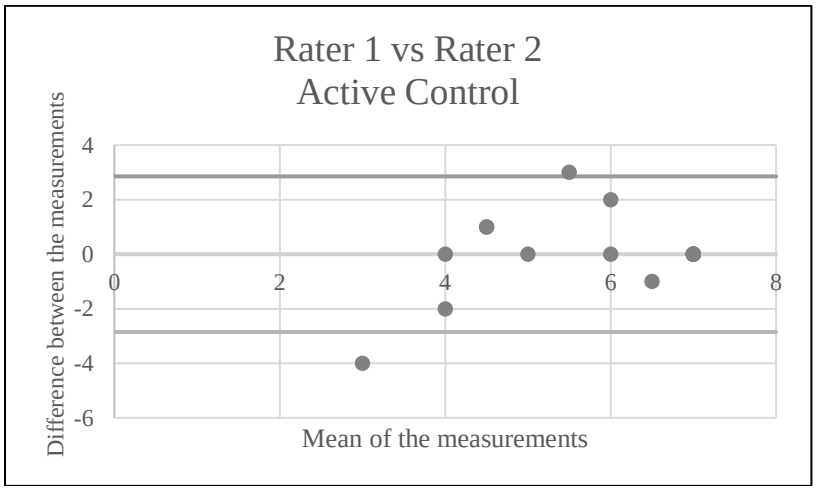
Table 3.3. Reliability of the SATCo Using ICC (2,1)

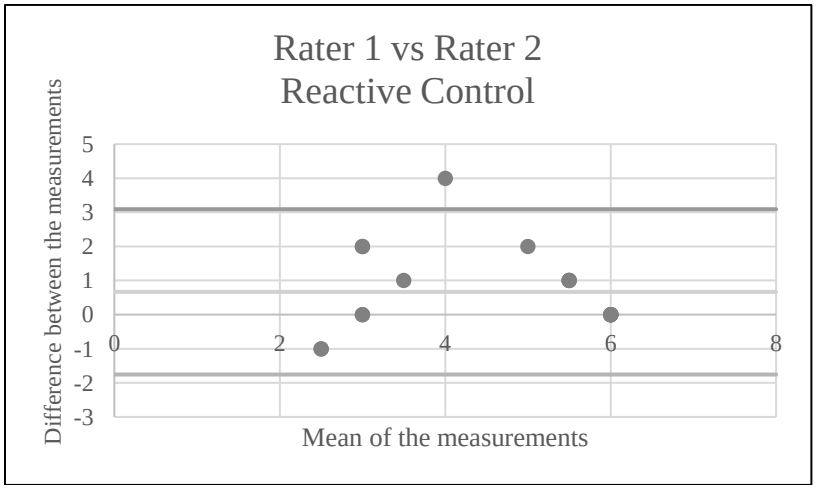
	Static ICC (2,1) [95%CI]	**Active** ICC (2,1) [95%CI]	**Reactive** ICC (2,1) [95%CI]	**Total Score** ICC (2,1) [95%CI]	**SATCo Level** ICC (2,1) [95%CI]
Interrater Reliability (Rater 1 vs Rater 2)	0.647 [0.272,0.852] $p = 0.001$	0.544 [0.118,0.801] $p = 0.008$	0.686 [0.336,0.870] $p = 0.001$	0.661 [0.294,0.858] $p = 0.001$	0.615 [0.221,0.836] $p = 0.003$
Interrater Reliability (Rater 2 live vs Rater 3 video)	0.852 [0.647,0.942] $p < 0.001$	0.679 [0.324,0.867] $p = 0.001$	0.549 [0.124,0.803] $p = 0.008$	0.784 [0.511,0.913] $p < 0.001$	0.568 [0.152,0.813] $p = 0.006$
Interrater Reliability (Rater 2 video vs Rater 3 video)	0.859 [0.662,0.945] $p < 0.001$	0.678 [0.332,0.866] $p = 0.001$	0.508 [0.068,0.782] $p = 0.013$	0.747 [0.441,0.897] $p < 0.001$	0.524 [0.090,0.791] $p = 0.011$
Intrarater Reliability (Rater 1 live vs Rater 1 video)	0.852 [0.648,0.0.942] $p < 0.001$	0.830 [0.602,0.933] $p < 0.001$	0.806 [0.554,0.923] $p < 0.001$	0.859 [0.662,0.945] $p < 0.001$	0.806 [0.554,0.923] $p < 0.001$
Intrarater Reliability (Rater 2 live vs Rater 2 video)	0.922 [0.803,0.970] $p < 0.001$	0.772 [0.488,0.908] $p < 0.001$	0.846 [0.635,0.939] $p < 0.001$	0.941 [0.850,0.978] $p < 0.001$	0.867 [0.679,0.948] $p < 0.001$

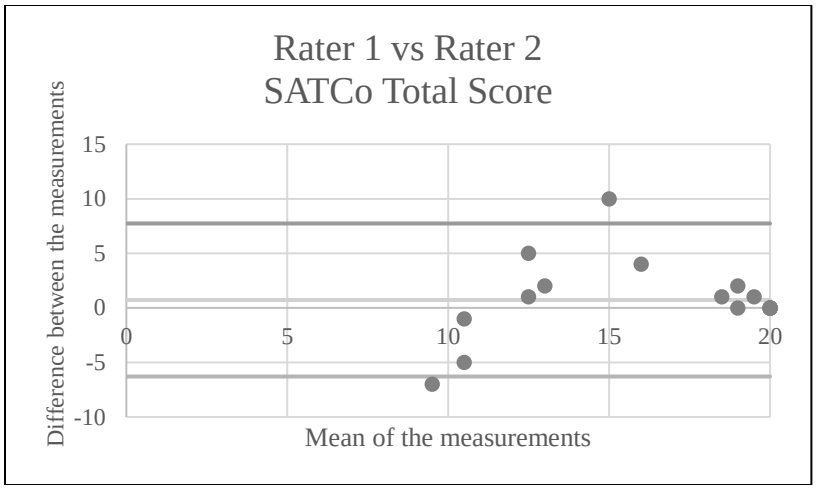
Bland-Altman plots for all comparisons are presented in Figures 3.3 through 3.7. Together with ICC (2,1) scores, the Bland-Altman plots corroborate the moderate-to-good interrater reliability among raters who separately scored the same video recording. The plots show that when the mean total score was 14 out of 20, there was a larger difference between raters for both interrater reliability of live rating versus video recording and video scoring for two separate PT raters. PT Rater 2 had some higher intrarater reliability ICC (2,1) scores than PT Rater 1, but the Bland-Altman plots for intrarater reliability of both PT raters did not demonstrate large differences between the raters. The general trend of the Bland-Altman plots is consistent with the outcome of the ICC (2,1) values among raters across all comparisons of reliability.

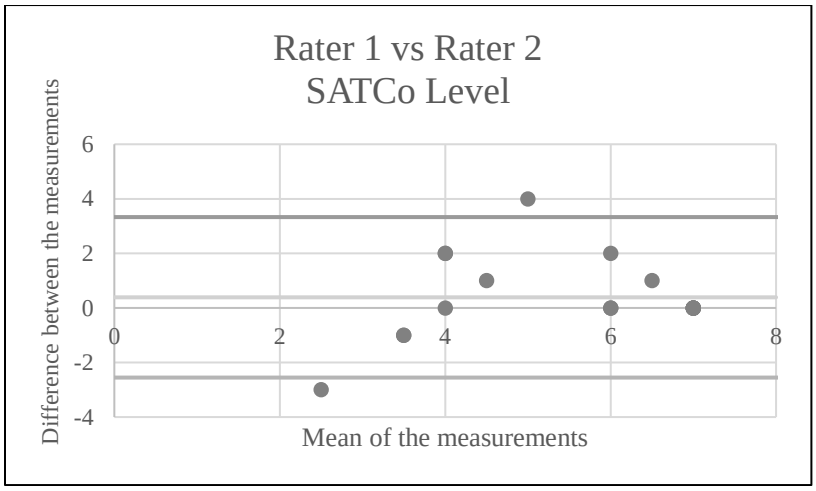
Figure 3.3. Bland-Altman Plots for Rater 1 Versus Rater 2 (Interrater Reliability)





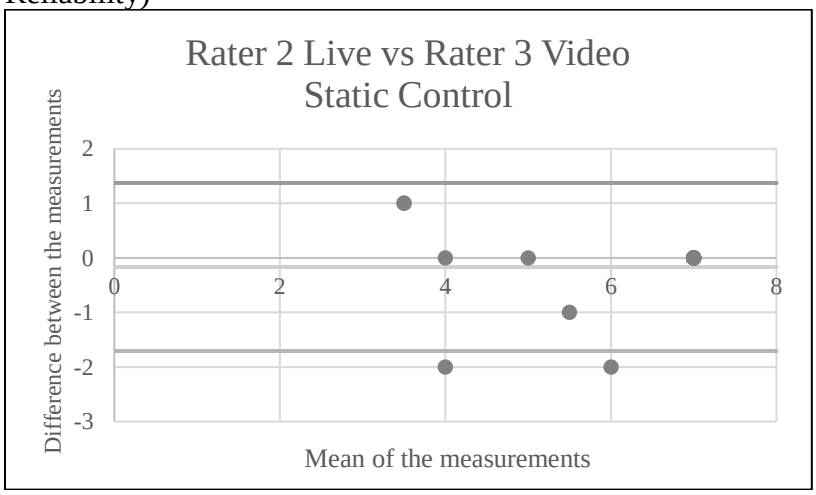


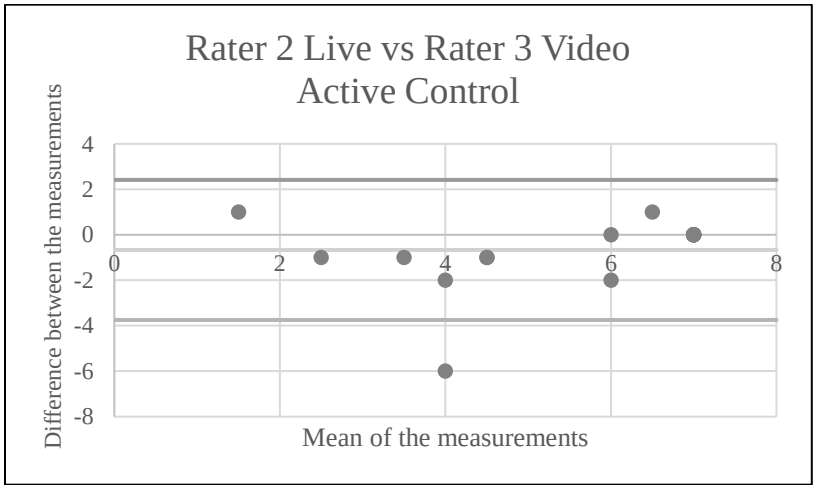


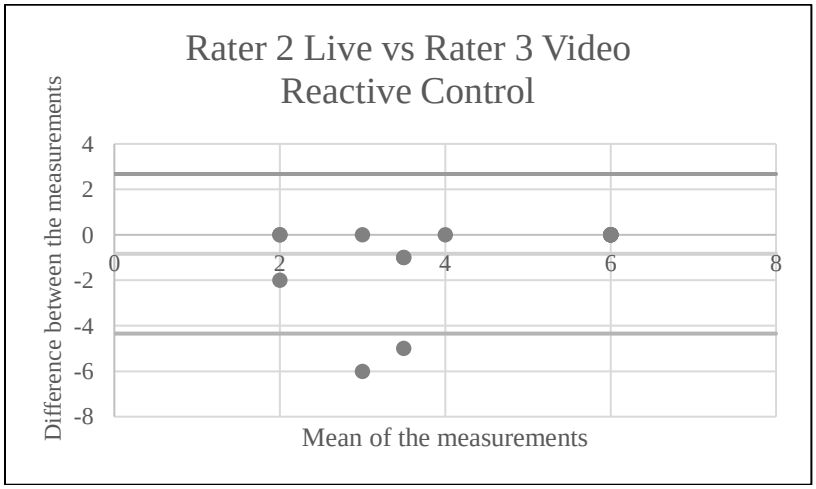


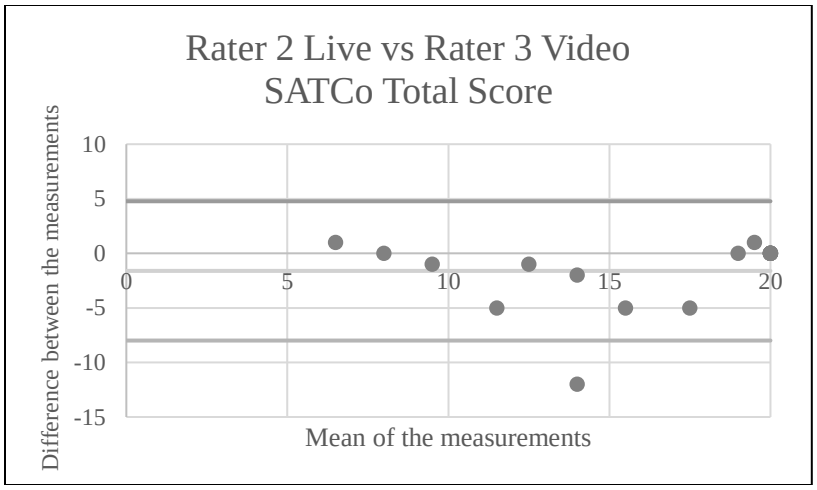
Difference
Mean
Lower
Upper

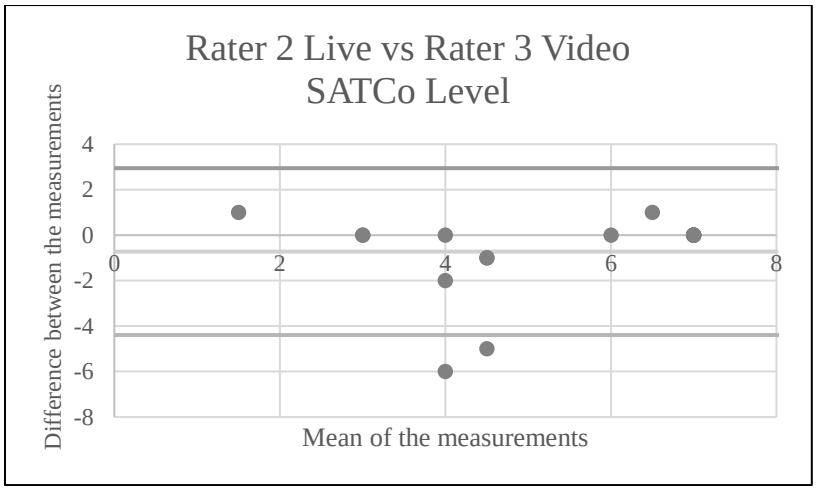
Figure 3.4. Bland-Altman Plots for Rater 2 Live Versus Rater 3 Video (Interrater Reliability)





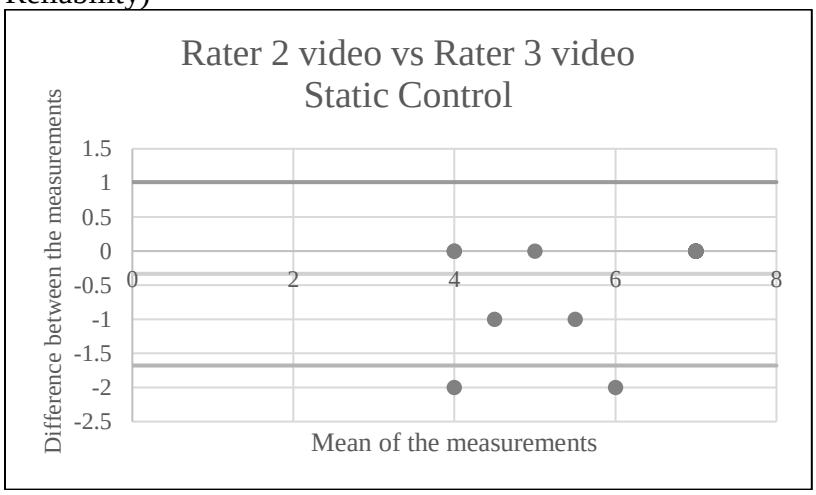


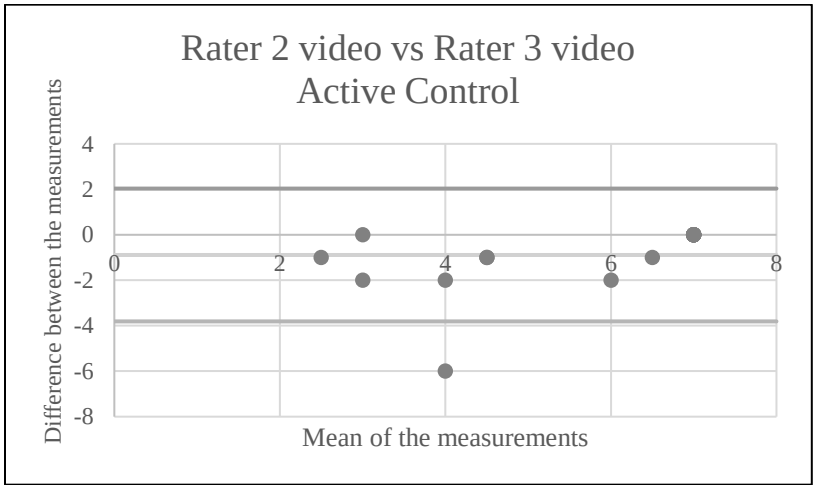


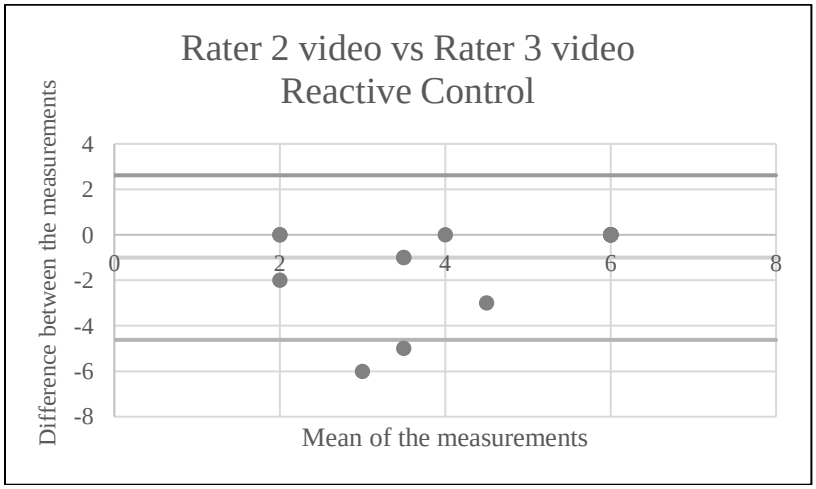


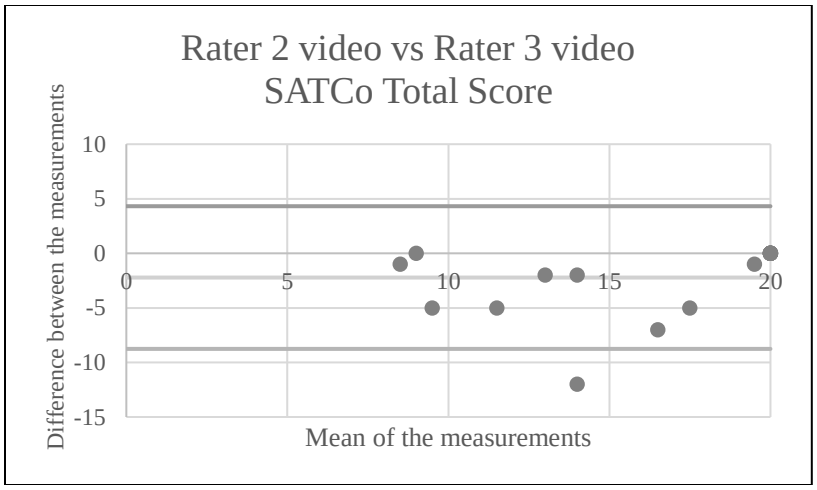
Difference
Mean
Lower
Upper

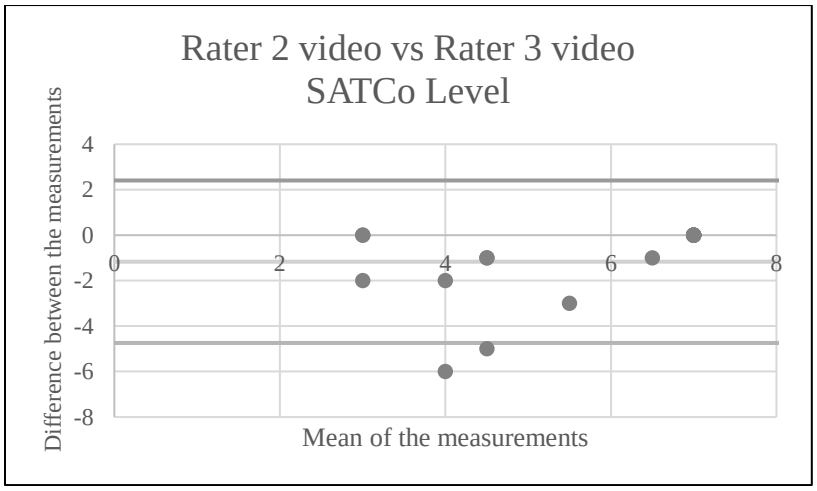
Figure 3.5. Bland-Altman Plots for Rater 2 Video Versus Rater 3 Video (Interrater Reliability)





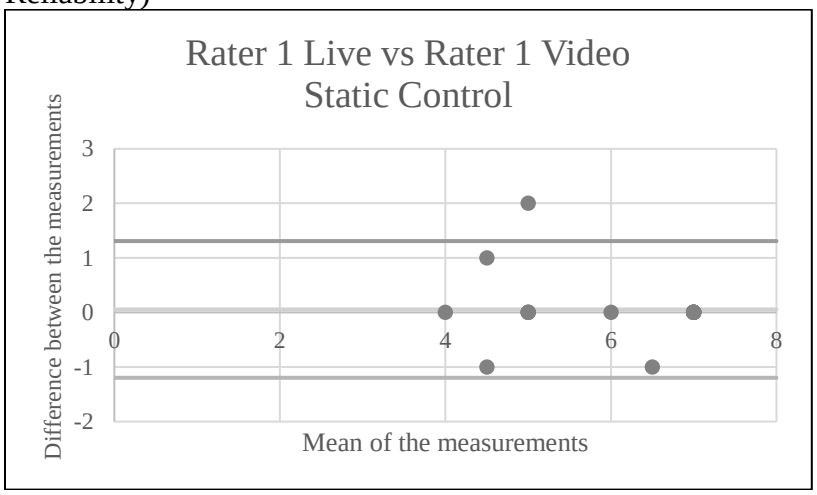


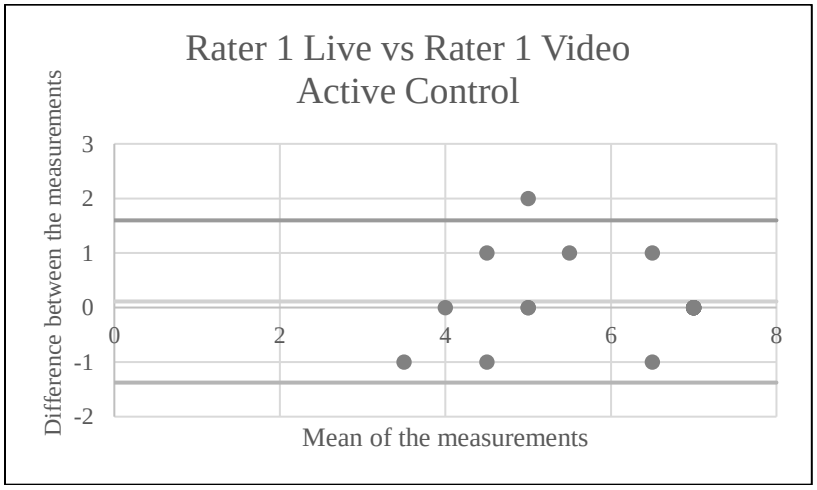


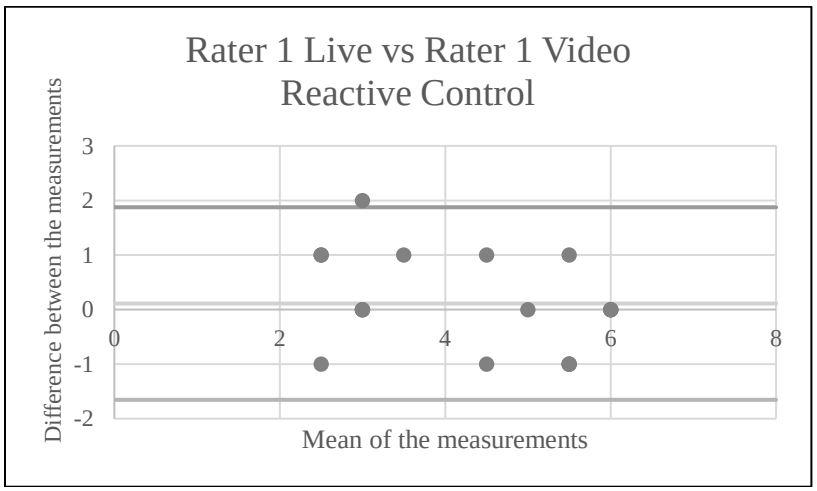


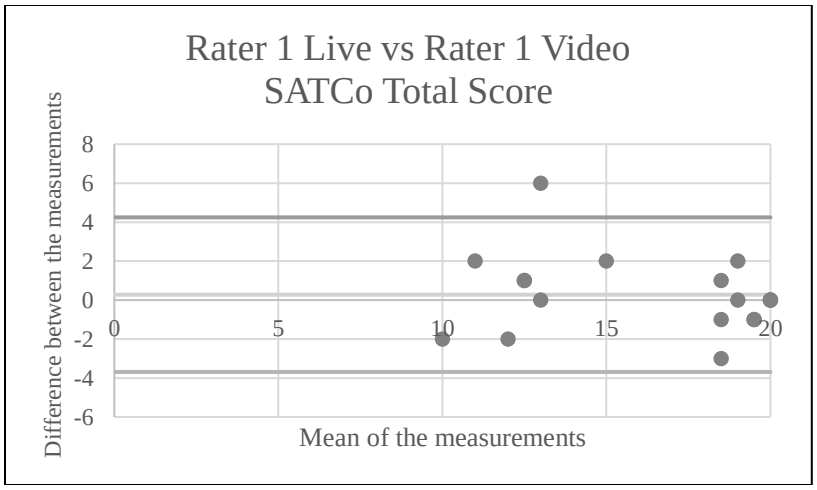
Difference
Mean
Lower
Upper

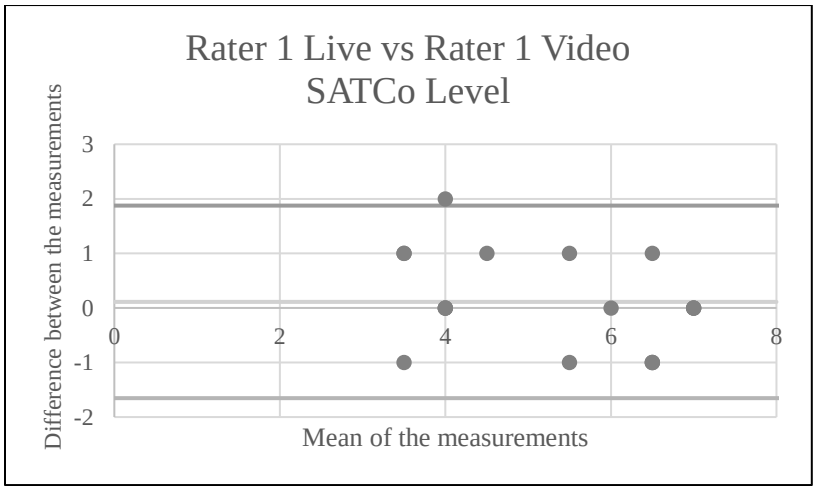
Figure 3.6. Bland-Altman Plots for Rater 1 Live Versus Rater 1 Video (Intrarater Reliability)








Rater 1 Live vs Rater 1 Video
SATCo Total Score
Difference between the measurements
8
6
4
2
0
-2
-4
-6
0
5
10
15
20
Mean of the measurements


Rater 1 Live vs Rater 1 Video
SATCo Level
Difference between the measurements
3
2
1
0
-1
-2
0
2
4
6
8
Mean of the measurements

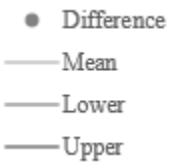
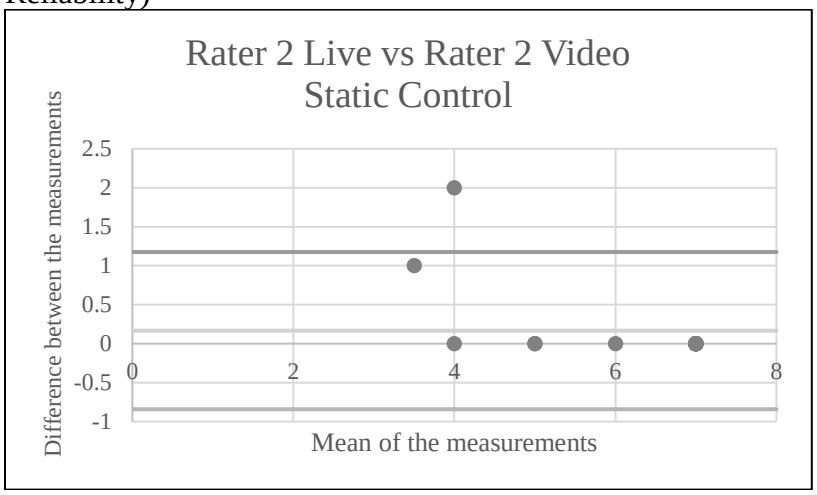
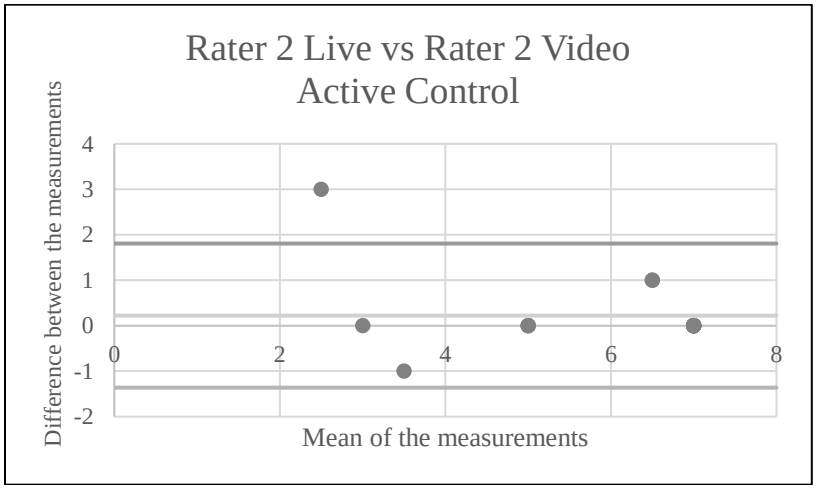
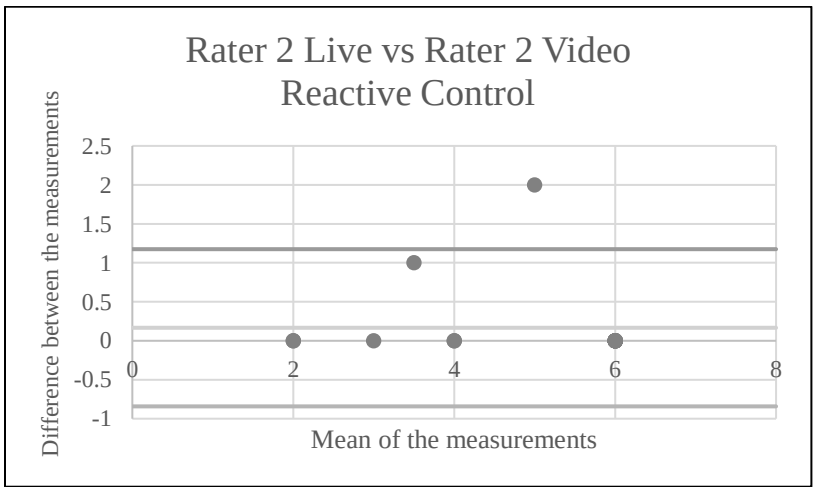
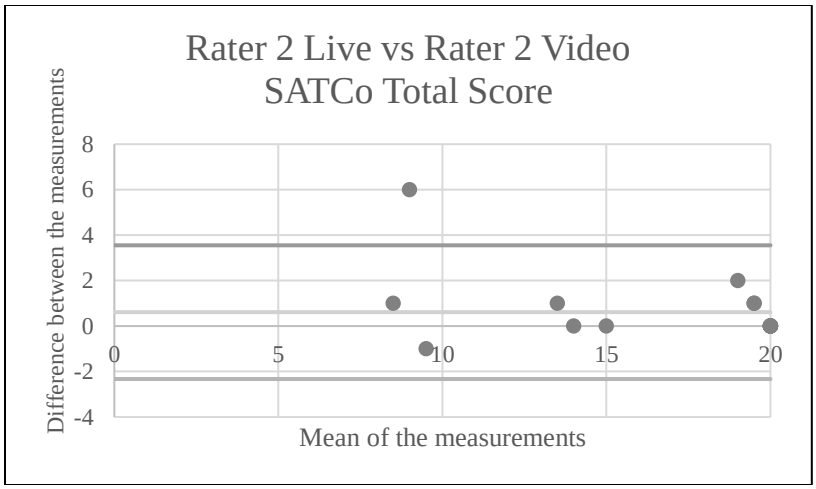

Difference
Mean
Lower
Upper

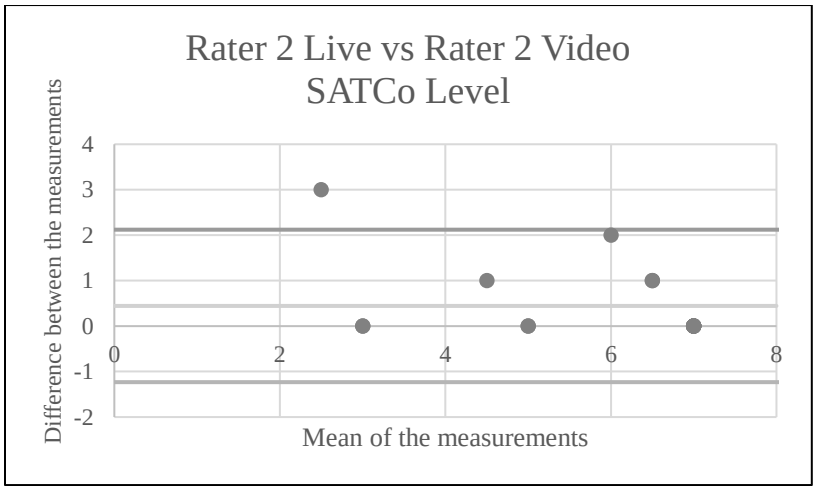
Figure 3.7. Bland-Altman Plots for Rater 2 Live Versus Rater 2 Video (Intrarater Reliability)

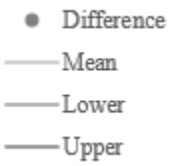












DISCUSSION

The SATCo is a measure of segmental trunk control for children with neuromotor disabilities. This study expands upon previous psychometric studies of the SATCo and informs clinicians about the reliability of this tool for infants and toddlers with DS.[10-13,42,86] Among PT raters scoring live testing sessions, the interrater reliability of the

SATCo was moderate, as demonstrated by both the ICC (2,1) scores and the Bland-Altman plots. Clinically, the SATCo appears to be a useful tool for children with DS with good to excellent intrarater reliability and moderate to good interrater reliability among PT raters. The SATCo may prove more useful when used by the same clinician to monitor and document incremental changes in trunk control in infants and toddlers with DS. However, responsiveness of the SATCo in this population has not yet been established.

Agreement among raters was highest for the older children who demonstrated full trunk control, suggesting a ceiling effect for children with DS who are older than 16 months or who demonstrate the ability to sit upright without assistance. The SATCo takes less than ten minutes to administer, but it does require the assistance of at least one extra person.

PT raters were all trained during one session using one video example and one live demonstration example. They were also provided with the supporting literature and written administration instructions. However, the primary investigator observed some variability between the raters in the testing sessions. Rater 1 appeared to be more lenient with scoring, erring on the side of giving children a score of "present" more often. Rater 3, who only watched videos and did not participate in any of the live testing sessions appeared to have a stricter, more narrow view of trunk control that was "present." Butler et al[10] found good to excellent reliability among all categories of the SATCo. The raters in Butler et al's study all had previous experience with development or administration of the SATCo, which may account for their higher reliability results.[10] The PT raters in our

study, although experienced clinicians who all attended training on the SATCo, had no previous exposure to the SATCo.

When the participants had difficulty maintaining trunk control, they often used subtle compensations such as hand support on top of the leg or clasping their hands together. PT raters experienced difficulty in observing these compensations while sitting behind the wiggling participants. Pin et al[86] suggest having a trained assistant vigilantly monitor and report these small compensations during the testing session.

Study Strengths

All three raters for this study were blinded to each other's scores and their own previous scores. The primary author (MF), who did not perform any of the scoring for this study, analyzed the data. Children across the inclusion age range from six to 24 months were included in the sample, representing a diverse range of gross motor development. This sample allowed the research team to administer the SATCo to children with DS who were just beginning to develop trunk control and to those who had mastered full upright sitting balance. All testing occurred in the same location with the same materials and assistance to ensure consistency of testing.

Study Limitations

A more rigorous training session with several live child demonstrations and more hands-on practice could have decreased the variability in scoring among the PT raters for control that was "present" or "absent" in infants and toddlers with DS. The addition of more than one training session may also have improved testing consistency among the raters.

Scores for the live testing sessions were obtained immediately after administration. The developers of the SATCo suggest using a video recording to review and assist with scoring.[10] Good to excellent reliability was found by several authors when video recordings were used score the SATCo.[10,11,86] Perhaps allowing the PT raters to view their video and score the SATCo within one day of administration would have improved overall interrater reliability of the tool.

The number of participants for this study was obtained a-priori but is still a relatively small sample. Caution should be used in generalizing the results for the wider population of infants and toddlers with DS. Future studies should focus on further investigating the psychometric properties of the SATCo in infants and toddlers with DS. For example, clinicians would benefit from information on the responsiveness of the SATCo and the predictive ability of the SATCo for functional mobility in children with DS.

CONCLUSION

Three separate PT raters who had no prior experience with the SATCo were able to administer and score this outcome measure in infants and toddlers with DS. This study contributes to the literature on the psychometric properties of the SATCo and supports its use to measure trunk control in infants and toddlers with DS between the ages of six to 24 months.

CHAPTER IV

ASSESSING TRUNK CONTROL IN INFANTS AND TODDLERS WITH DOWN SYNDROME: VALIDITY AND PREDICTIVE ABILITY OF THE SEGMENTAL ASSESSMENT OF TRUNK CONTROL (SATCo)

INTRODUCTION AND PURPOSE

DS is a genetic condition that occurs in approximately 1.26 per 1,000 live births in the United States.[1] Infants and toddlers with DS display deficits in gross motor skills and postural control.[28,88,89] Although children with DS follow the same predictable sequence of motor development as their typically developing peers, they require twice as much time to acquire basic motor skills.[88] Postural control at the trunk is a precursor to the development of upright gross motor skills;[5] however, the association between trunk control and gross motor function in children with DS is not clearly understood. To date, few articles have investigated this association in children with DS.

Most of the research exploring postural control and gross motor function has been performed in children with cerebral palsy. Mendoza et al[90] found a relationship between sitting ability and the capacity to walk in children with cerebral palsy. The authors used the Level of Sitting Scale, which was originally developed to assess seated posture and motor control for the evaluation of seating systems in children with neuromotor disabilities. They compared the Level of Sitting Scale to walking ability (as measured by the Gross Motor Function Classification System) and found a significant correlation.[35,90]

Although the results are interesting, walking ability does not capture the full range of gross motor function in children.

Curtis et al[13] explored the relationship between trunk control and gross motor function in children with cerebral palsy. The authors used the SATCo, which assesses static, active, and reactive control at seven discrete levels of the trunk.[10,13] They found that both SATCo level and age were significant predictors of gross motor function as measured by the GMFM. The article supports the hypothesis that trunk control influences gross motor function in children with cerebral palsy. Additionally, Butler et al[10] found a significant correlation between the SATCo and the sitting dimension of the GMFM in children with neuromotor disabilities. These studies both demonstrate an apparent relationship between gross motor function and trunk control, but none of the research included children with DS. For infants and toddlers with DS, the relationship between trunk control and gross motor function has not yet been investigated.

Investigating the relationship between trunk control and gross motor function can inform clinicians toward effective interventions for their clients. Physical therapists should employ effective outcome measures to document impairments, demonstrate progress toward goals, and justify the need for physical therapy services. The SATCo shows potential as an outcome measure of segmental trunk control that may also be a predictor of gross motor function in infants and toddlers with DS.

The purpose of this study is to examine the concurrent validity of the SATCo with the GMFM and determine whether a model of staggered entry with age and SATCo score predicts GMFM score in infants and toddlers with DS.

METHODS

Study Design

A methodological study on a single group of children with DS was conducted. Dual Institutional Review Board approval was received from both Texas Woman's University and the University of St. Augustine for Health Sciences. All parents signed a written consent form prior to starting the testing.

Participants

A priori power analysis using G*Power[**] for a Pearson correlation test with a large effect size of $\rho = 0.7$, power = 0.8 and $p = 0.05$ revealed a sample size of 11 participants was required. For linear regression with an effect size of 0.7, power = 0.8, and $p = 0.05$, a sample size of 18 participants was required. Therefore, 18 participants (6 female, 12 male) with DS were recruited from local parent support groups, early intervention programs, and outpatient clinics within 50 miles of Austin, Texas. The average age of the participants was 13.67 months old ($SD = 5.31$).

The study was limited to children with a diagnosis of DS, between the ages of six to 24 months, who spoke and understood English. The age range was chosen to cover children who were developing sitting balance, as well as those who could maintain sitting independently. Participants were excluded from the study if they had a diagnosis

[**] G*Power Version 3.1.9.2, Franz Faul, Universitat Kiel, Germany

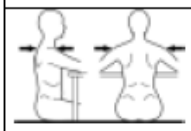
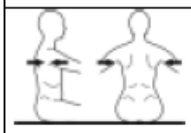
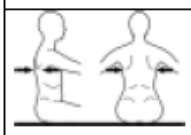
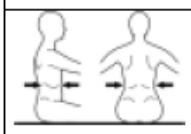
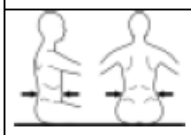
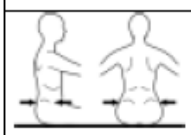
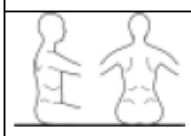
unrelated to DS that limited gross motor movement, or medical restrictions that contraindicated movement or handling.

Instruments

The SATCo is an outcome measure used to assess discrete levels of trunk control in children with motor disabilities.[10] To perform this assessment, the child is seated on a bench with his or her feet on a stable surface. The child's pelvis is held in a neutral position, either with a strapping system or manually by a second person. The tester gives manual support at specific landmarks on the trunk, starting at the shoulder girdle and moving segmentally down the trunk. There are seven levels of control. Static trunk control (seven items), active trunk control (seven items), and reactive trunk control (six items) at each level are recorded, for a total of 20 items. The tester gives the child a score of present (✓), absent (-), or not tested (NT) for each item. This is demonstrated by the child's ability to maintain a neutral posture for static control, maintain neutral posture during head turns for active control, and maintain or quickly regain neutral posture during perturbations for reactive control. Levels increase as the examiner's hands move down the trunk. The highest level at which a child is able to maintain control in all items is recorded as the child's level of trunk control. Higher levels indicate better trunk control with less support needed from the tester. The highest score, level seven, is given when no support is needed, and the pelvis strap or support is removed.[10] For this study, children were given one point for each item in which trunk control was marked "present," resulting in a score range of zero to 20 for the items, and zero to seven for the highest level of control. For example, a child who was able to maintain static and active control

at the shoulder girdle and axillae but was unable to maintain reactive control at the axillae (and unable to maintain control at any other level) would receive an item score of four and a SATCo level score of one. See Figure 4.1 for the SATCo score sheet and levels.

Figure 4.1. SATCo Score Sheet with Levels[10]

Identification #: Tester Name: Date:	**Level of Manual Support** Pelvic/thigh strap used except as indicated	**Functional Level** Arms and hands in air except as indicated	**Static**	**Active**	**Reactive**	**Comments**
			Maintain vertical neutral position of head and trunk above manual support level			
			minimum of 5 seconds	while turning head with arms lifted	Maintain/quickly regain following brisk nudge	
	Shoulder girdle Testers hand position may vary from horizontal	**Head control** Arms may be supported throughout			**NOT Tested for Head Control**	Level 1
	Axillae	**Upper Thoracic Control**				Level 2
	Inferior scapula	**Mid Thoracic Control**				Level 3
	Over lower ribs	**Lower thoracic Control**				Level 4
	Below ribs	**Upper lumbar Control**				Level 5
	Pelvis	**Lower lumbar Control**				Level 6
	No support given and pelvic/thigh straps removed	**Full trunk control**				Level 7
Fixed spinal deformity? Yes_____No_____Comments_____ Limitation of Cervical Rotation Left_____Right_____Comments_____						

Intrarater reliability for the SATCo has been shown to be excellent (ICC = 0.98) across all aspects of control; and interrater reliability was good (ICC = 0.8 to 0.84) in typically developing children and those with neuromotor disabilities.[10] The SATCo shows concurrent validity with the GMFM Dimension B (r = 0.73 to 0.83), and with the sitting section of Alberta Infant Motor Scale (r = 0.86 to 0.88).[10]

The GMFM was developed to measure gross motor function in children with cerebral palsy and can be used for children with DS under six years old.[7,23,24] The examiner scores a child's capabilities across five dimensions of functional movement: A) Lying and Rolling, B) Sitting, C) Crawling and Kneeling, D) Standing, and E) Walking, Running, and Jumping. Each dimension is made up of several items (88 total items) with a total possible score of 264. For each item, the child receives a score of 0 (does not initiate), 1 (initiates), 2 (partially completes), or 3 (completes). The GMFM can be administered by direct observation of the child supplemented with parent report.[8,9] For children with DS, the GMFM has strong interrater reliability (ICC = 0.96 to 0.98) and test-retest reliability (ICC = 0.95 to 0.96), with evidence of responsiveness and validity with the Motor Scale of the Bayley Scales of Infant Development.[9,27]

Procedure

The SATCo and GMFM were administered to all participants by a pediatric physical therapist with 19 years of clinical experience who was familiar with the outcome measures. Both tests were performed during one session. Two trained assistants helped with the administration of the SATCo. One assistant secured the child's pelvis and the other assistant provided gentle nudges and visual stimulus (shaking a rattle) for head

turns. The pediatric physical therapist administered the GMFM with the modifications recommended by Russell et al[7] and Gemus et al[9] for children with DS. These included the use of parent report if the child refused to perform an item and the use of verbal cues or demonstration.[9]

Data Analysis

Data was analyzed using IBM SPSS Statistics 25 software.[††] To examine the concurrent validity of the SATCo with the GMFM, Spearman's rho was calculated for each category of the SATCo, total SATCo score, SATCo level, dimension B (sitting) of the GMFM, and total GMFM score. A p-value < 0.05 was used to indicate significance. Spearman's rho correlation results were interpreted as: little or no relationship (0 to 0.25), fair relationship (> 0.25 to 0.50), moderate to good relationship (> 0.50 to 0.75), and good to excellent relationship (> 0.75).[91]

To explore whether age and SATCo scores have a predictive effect on GMFM in infants and toddlers with DS, a linear model was used:

$$\mathrm{GMFM}_i = b_0 + b_1\mathrm{SATCo}_i + b_2\mathrm{Age}_i$$

Age was included in the model because gross motor growth curves from the GMFM have been shown to be related to age in children with DS.[8] To fully explore the relationships, several models of linear regression were analyzed using blocked or hierarchical entry.[92]

[††] IBM Corporation, Armonk, New York

RESULTS

Six female and 12 male children with DS participated in the study. The race and ethnicity of the participants included children who identified as Black/ African American (n = 2), Latino/a (n = 4) and Caucasian (n = 12). All participants were tested on the SATCo and the GMFM by the same experienced pediatric physical therapist during one session. Descriptive statistics for the variables tested are found in Table 4.1.

Spearman's rho correlations revealed a good to excellent significant relationship ($r_s > 0.75$, $p < 0.001$) for every category of the SATCo (static, active, and reactive control), SATCo total score (composite of all categories), and SATCo level versus GMFM Dimension B score and GMFM total score. Dimension B of the GMFM represents the child's ability to maintain static and dynamic sitting. Correlation values are presented in Table 4.2.

Table 4.1. Descriptive Statistics for the Variables

Variable	**Mean (*SD*)**	**Min**	**Max**
Age (months)	13.67 (5.31)	6	23
GMFM Total Score	98.83 (45.88)	36	167
GMFM Dimension B (Sitting) score	34.44 (20.75)	7	67
SATCo Static Score	5.72 (1.53)	4	7
SATCo Active Score	5.83 (1.30)	4	7
SATCo Reactive Score	4.22 (1.59)	2	6
SATCo Total Score	16.06 (4.01)	10	20
SATCo Level	5.22 (1.59)	3	7

Table 4.2. Spearman's Rho Correlations (r_s)

	SATCo Static Score	**SATCo Active Score**	**SATCo Reactive Score**	**SATCo Total Score**	**SATCo Level**
GMFM Dimension B Score	0.781	0.803	0.834	0.821	0.834
GMFM Total Score	0.788	0.832	0.821	0.829	0.821

*all values significant at $p < 0.001$

Results of all linear regression models are summarized in Table 4.3. A hierarchical model was used to determine if SATCo level has a predictive effect on GMFM total score beyond what age predicts. In this model, age accounted for 63% of the variation in GMFM total score and SATCo total score accounted for an additional 17%. To further explore the data, another hierarchical model was used with SATCo total score as the primary predictor and age as a secondary predictor. This model showed that SATCo total score accounted for 71% of the variation in GMFM total score and age accounted for an additional 9%. Both models revealed a significant regression equation ($F[2,15] = 30.45$, $p < 0.001$). The raw coefficients for the predictive equation for both models were as follows: $GMFM_i = -53.22 + (6.40 \times \text{SATCo total score}) + (3.61 \times \text{Age})$.

Table 4.3. GMFM Prediction Models

Variable	*B* (SE)	*p*	*F*	R	R^2	Adjusted R^2
Block Entry Models with One Predictor on GMFM Total Score						
Age	6.87 (1.31)	<0.001	27.53	0.79	0.63	0.61
SATCo Total Score	9.63 (1.55)	<0.001	38.64	0.84	0.71	0.69
Block Entry Model with Two Predictors on GMFM Total Score						
Model 1			30.45	0.90	0.80	0.77
Age	3.61 (1.34)	0.017				
SATCo Total Score	6.40 (1.78)	0.003				
Hierarchical Models with Two Predictors on GMFM Total Score						
Model 1			27.53	0.80	0.63	0.61
Age	6.87 (1.31)	<0.001				
Model 2			30.45	0.90	0.80	0.78
Age	3.61 (1.34)	0.017				
SATCo Total Score	6.34 (1.78)	0.003				
*R^2 Change = 0.17						
Model 1			38.64	0.84	0.71	0.69
SATCo Total Score	9.63 (1.55)	<0.001				
Model 2			30.45	0.90	0.80	0.78
SATCo Total Score	6.34 (1.78)	0.003				
Age	3.61 (1.34)	0.017				
*R^2 Change = 0.09						
Block Entry Models with One Predictor on GMFM Dimension B						
Age	3.19 (0.56)	<0.001	31.89	0.82	0.67	0.65
SATCo Total Score	4.47 (0.66)	<0.001	46.59	0.86	0.74	0.73
Block Entry Models with One Predictor on SATCo Total Score						
GMFM Total Score	0.07 (0.01)	<0.001	38.64	0.84	0.71	0.70
GMFM Dimension B	0.17 (0.02)	<0.001	46.59	0.86	0.74	0.73
Age	0.51 (0.014)	0.002	13.39	0.68	0.46	0.42

Models using block entry of the single predictors of age or SATCo level were consistent with the hierarchical models, demonstrating 63% and 71% of the variance in GMFM total scores, respectively. When analyzing the opposite model, the single predictor of GMFM total score also predicted 71% of the variance in SATCo total score.

Block entry of the single predictors of age (R = 0.82, R^2 = 0.67, F[1,16] = 31.89, p < 0.001) and SATCo total score (R = 0.86, R^2 = 0.74, F[1,16] = 46.599, p < 0.001) had a significant predictive effect on dimension B (Sitting) of the GMFM. Results of the linear regression models are shown in Table 4.3.

DISCUSSION

In previous published research, the relationship of trunk control and gross motor function in infants and toddlers with DS has not been fully explored. This study provides evidence that segmental trunk control, as measured by the SATCo, shows a strong correlation to gross motor function, as measured by the GMFM. Furthermore, there is an excellent correlation between trunk control and the dimension B (sitting) of the GMFM. These results confirm the concurrent validity of the SATCo for infants and toddlers with DS between the ages of six to 24 months.

The SATCo and the GMFM are measurements at two different levels of the World Health Organization's ICF model.[6,84] The SATCo measures trunk control at the body structure and function level and the GMFM measures gross motor function at the functional activities level. The SATCo is a quick and easy assessment to perform, taking a fraction of the time that it takes to perform the full GMFM. Given that the GMFM takes around 40 minutes to administer and the SATCo takes less than 10 minutes, this information may be useful for clinicians who do not have the time to perform the full GMFM during their session. Clinicians may want to consider that the SATCo requires a second person if a strapping system is not used to secure the child's pelvis.

The results of this study showed a predictive effect of trunk control on gross motor function for infants and toddlers with DS. All linear regression models showed that the SATCo was a significant predictor of GMFM total score. This evidence indicates trunk control is an important factor for determining gross motor function in infants and toddlers with DS.

This study builds upon similar work done by Curtis et al[13] in children with cerebral palsy. In their study, age did not have a predictive effect on GMFM total score, which is not surprising for the cerebral palsy population because they usually do not follow the normal trajectory of development of gross motor skills as they age.[93] However, children with DS show a predictable, but delayed, trajectory of gross motor skills. Our results indicate that as a single predictor, age, accounted for a large amount of the variance in GMFM total score. Regardless of whether hierarchical or blocked entry was used for the regression models, both SATCo and age remained significant predictors for children with DS who were less than 24 months of age.

This study had a strong research design with a diverse sample of participants of various ages and ethnicities. However, this study also has several limitations. Given the narrow age range for inclusion, recruitment of participants was difficult. Therefore, the sample size for regression analysis was relatively small. A larger sample would make the results more generalizable to the general population of children with DS. Also, the SATCo has a ceiling effect and we suspect that as children with DS get older, the SATCo will no longer predict GMFM total scores because children will likely demonstrate full trunk control well before they have mastered all the items on the GMFM. Therefore, the

results of this study cannot be generalized for children above our inclusion age cut-off of 24 months.

CONCLUSION

Trunk control appears to play a central role in the gross motor function of infants and toddlers with DS. This study confirms the concurrent validity of the SATCo and supports its use to measure trunk control in infants and toddlers with DS who are between the ages of six to 24 months.

CHAPTER V

IMPACT OF A DYNAMIC STANDING HOME PROGRAM FOR INFANTS AND TODDLERS WITH DOWN SYNDROME:

A SINGLE-CASE EXPERIMENTAL DESIGN STUDY

INTRODUCTION AND PURPOSE

DS is a genetic condition that occurs in about 1 in every 800 live births in the United States.[1] Children with DS experience a predictable sequence of development, but they are delayed in their gross motor skills compared to their typically developing peers.[28,88,89] The health care costs for children with DS are estimated to be about 12 times higher than for typically developing children, emphasizing the necessity for early intervention.[14] In addition, financially feasible and effective home programs to supplement physical therapy are needed to optimize the development of neuromotor control.[15]

Home programs that promote upright mobility through treadmill training have proven effective for accelerating the acquisition of motor skills in infants (ages eight to 12 months) with DS.[2,16] Numerous studies have determined that in-home use of treadmills for infants (ages eight to 12 months) with DS is feasible.[2,16,17,32,79] Furthermore, infants (ages eight to 12 months) with DS who participate in a treadmill training home program walk earlier than they normally would when compared to infants of similar age with DS who do not participate in treadmill training.[2,16,17] The literature provides strong evidence that early implementation of treadmill training can positively

impact the gross motor skill acquisition in infants with DS.[2,16,17] However, limitations of a treadmill home program include the bulky size of the equipment and its cost, which can be upwards of $1600. An alternative form of upright mobility that is more affordable and less cumbersome is needed.

The Upsee[‡‡] shows promise as a dynamic standing device that can be used by a parent and child to promote upright mobility. The device uses a child trunk harness connected to an adult hip belt with a shared set of sandals worn by both the adult and child. Children must be able to bear weight through their lower extremities (LEs) and be shorter than the adult's umbilicus when standing in order to use the device.

A case series by Ardolino et al[18] demonstrated that home use of the Upsee improved trunk control and gross motor skills in two children (ages 21 and 24 months old) with trunk hypotonia who could bear weight through their LEs, but were unable to stand unassisted. The authors used the SATCo and the GMFM as outcome measures. Casey et al[19] documented the feasibility of the Upsee to promote physical activity (as measured by Goal Attainment Scaling) and social engagement (as measured by the Mobility Ability Participation Assessment) in a four-year-old child with Pitt Hopkins syndrome. The child was able to maintain standing balance for five seconds, had difficulty maintaining head control for five seconds, and required maximum assistance to walk. After using the Upsee, he could maintain standing balance more than 30 seconds, maintain head control more than 30 seconds, and walk with moderate assistance. Another case study discovered improved GMFM score in a 31-month-old child with cerebral

[‡‡] Firefly by Leckey, Lisburn, Northern Ireland

palsy classified as Gross Motor Function Classification System level III after using the Upsee in combination with kinesiotaping.[82] The evidence from these cases suggest that this dynamic standing device may lead to improved gross motor skills, but more research is needed to explore the use of this device in children with DS.[18,19,82] The research also indicates that trunk control, which is an important factor for mobility, may also be impacted.[5] The effectiveness of dynamic standing through use of the Upsee has not been studied in infants and toddlers with DS.

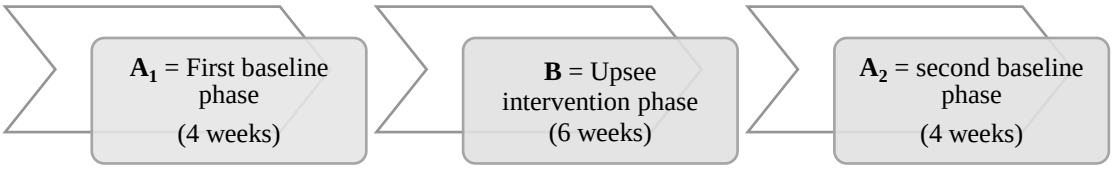
The purpose this single-case experimental designed study was to explore the impact of a dynamic standing device (Upsee) on trunk control and motor skills in infants and toddlers with DS (ages nine to 30 months).

METHODS

Study Design

This withdrawal/reversal single-case experimental design study lasted 14 weeks with two conditions: two baseline phases (A_1 and A_2) lasting four weeks each, and a six-week dynamic standing phase (B). During the baseline phases A_1 and A_2, participants continued with their usual therapy with no additional home program. The six-week intervention phase, B, employed a home program dynamic standing intervention. Figure 5.1 shows a graphic of the research design. The independent variables were the presence or absence of a dynamic standing home program. Dependent variables included an assessment of trunk control over time as measured by the Segmental Assessment of Trunk Control (SATCo), and assessment of gross motor function over time as measured by the Gross Motor Function Measure (GMFM).

Figure 5.1. Graphic of A_1BA_2 Withdrawal/Reversal Single-Case Experimental Design



Dual institutional review board approval was received from both Texas Woman's University and the University of St. Augustine for Health Sciences. All parents signed a written consent form prior to starting the testing.

Participants

Six children with DS (one female, five male) were recruited from local clinics and parent support groups in the Austin, Texas area to participate in the study. Participants were required to meet the following inclusion criteria: 1) diagnosis of DS, 2) between nine and 30 months of age, 3) able to bear weight through lower extremities when supported, 4) delay in gross motor skills, and 5) not independently walking. Participants were excluded from the study if they had: 1) a diagnosis unrelated to DS that limited gross motor movement, 2) medical restrictions that contraindicated movement or handling, 3) severe hypertonicity in the lower extremities, 4) a history of multiple lower limb fractures, 5) severe lower extremity asymmetry including hip dislocation, or 6) an unstable medical condition requiring rapid repositioning for treatment (such as epilepsy or breathing difficulties). Additionally, the adult using the device could not be pregnant or suffering from back pain, mobility, or balance problems. The child and adult had to both be able to stand upright in the device with the child's shoulders below the level of

the adult's navel. Participants were screened on inclusion and exclusion criteria prior to study start. The average age of the child participants was 16.83 months (SD = 3.98).

Outcome Measures

The SATCo is tool that assesses discrete levels of trunk control in children with motor disabilities.[10] To complete the SATCo, the child is seated on a bench with his or her feet on a stable surface and the pelvis secured in a neutral position by using a strapping system or a second person. The tester provides manual support at specific locations on the trunk, starting at the shoulder girdle and moving segmentally down through seven levels. Static, active, and reactive trunk control at each level are recorded as present (✓), absent (-), or not tested (NT), for a total of 20 items. The highest level that a child can maintain control in each category (static, active, and reactive) is the child's level of trunk control. There are seven levels and higher levels indicate better trunk control.[10]

The SATCo has excellent intrarater reliability (ICC = 0.98) and good interrater reliability (ICC = 0.80 to 0.84). Concurrent validity has been established with the GMFM Dimension B (r = 0.73 to 0.83), and with the sitting section of Alberta Infant Motor Scale (r = 0.86 to 0.88).[10]

The GMFM is a measure of gross motor function that was originally developed for children with cerebral palsy.[94] The GMFM is also commonly used as a criterion-referenced outcome measure for children with DS under six years old.[7,23,24] The tester scores a child's capabilities across five functional movement dimensions: A) Lying and Rolling, B) Sitting, C) Crawling and Kneeling, D) Standing, and E) Walking, Running,

and Jumping. The highest possible total GMFM score is 88. In children with DS, the GMFM can be administered by direct observation of the child supplemented by parent report.[8,9] The GMFM has strong interrater reliability in children with DS (ICC = 0.96 to 0.98) and strong test-retest reliability (ICC = 0.95 to 0.96), with evidence of responsiveness and validity with the Motor Scale of the Bayley Scales of Infant Development.[7-9]

Intervention

A pediatric physical therapist with five years of experience who was blinded to the intervention administered all outcome measures. The GMFM and the SATCo were performed in the home of the participants one time per week for duration the study. Outcome measures were administered at the same time each week during both four-week baseline phases (A_1 and A_2) and the six-week intervention phase (B), for a total of 14 weeks of testing. Throughout the study, participants continued with their usual therapy sessions.

During the two baseline phases (A_1 and A_2), the children did not receive any additional intervention, but continued with their usual therapy sessions. During the intervention phase (B) a dynamic standing device, the Upsee, was used in a home program supervised by an experience pediatric physical therapist. The device consists of a child harness connected to an adult hip belt. A shared pair of sandals are worn over shoes by both the parent and child. As the parent stands, the child is encouraged to stand, weight shift, and take steps.

On the first day of the intervention phase, a pediatric physical therapist with 14 years of experience delivered the dynamic standing device (Upsee) to the parent and child. The physical therapist provided training and education to the parent on the home program and assessed the device for fit and function. When wearing the device, parents can feel decreased weight through the adult hip belt when the child is actively weight bearing and engaging in play, defined as "active" use of the device. Parents can also feel more weight added to the hip belt when the child sits back into the device taking weight off their LEs, defined as "passive" use of the device. Parents were educated on "active" versus "passive" use of the device and were able to verbalize when they felt the difference when using the device. They were given a list of activities that would promote active use of the device by the child (in standing, weight shifting, or stepping), such as playing at a low table, reaching for objects, or taking steps. If the child was passive in the device for more than a few minutes, the parents were instructed to stop use and try again later. One participant (Child 3) wore supramalleolar orthotics while using the device and all participants wore shoes. None of the other participants owned or used orthotics. Figure 5.2 shows Child 1 and his mother using the dynamic standing device.

Figure 5.2. Child 1 Using the Upsee with his Mother

Parents were instructed to use the dynamic standing device for 30 minutes per day, at least five days per week with emphasis on active use of the device, promoting active standing and/or stepping, depending on the child's level of function. The parents kept a written daily log to record duration of device use, activities performed, and subjective observations of the child during the intervention. The physical therapist

performed weekly visits, lasting approximately 30 to 45 minutes, during the intervention phase of the study to check the fit of the device, ensure compliance, collect the written log, and answer any questions about the home program. A second physical therapist performed weekly outcome measure testing on the children through all three phases of the study. The physical therapist performing the supervised home program was blinded to the child's performance on the outcome measures.

Data analysis

Trunk control changes were assessed through graphing the median SATCo scores of the participants for each phase. The mean and standard deviation of each phase was calculated for the GMFM. Data was analyzed using visual analysis of graphed data points with the 2-standard deviation (2SD) band method for the GMFM. The 2SD method shows the probable distribution of scores that would occur over time without any additional intervention. A statistically significant change in performance on the outcome measures was indicated if two consecutive data points in a phase fell outside the 2SD band.[25,27] Additionally, between-phase comparisons were made to determine if positive changes that were immediate, readily discernible, and maintained over time occurred to indicate improvement.

To further explore whether there was a significant difference in GMFM scores between each phase of the study, the average of each phase was calculated and a Friedman's Analysis of Variance (ANOVA) was conducted.

RESULTS

All six of the participants completed the 14-week trial without missing any testing sessions. Participant demographics are presented in Table 5.1.

Table 5.1. Participant Demographics

Child Number	Gender	Race/ Ethnicity	Age (mo)	Sitting Functional Ability	Standing Functional Ability	Ambulation Ability
1	M	Latino	18mo	Independent with static and dynamic sitting	Able to pull to stand and maintain standing with UEs supported	Able to take 2-3 steps with bilateral hand-held assistance
2	M	Caucasian	15mo	Able to maintain propped sitting, requires assistance for upright sitting	Able to bear weight through LEs in standing with moderate support at trunk for balance	Unable to ambulate with or without assistance
3	M	Caucasian	15mo	Independent with static and dynamic sitting	Able to bear weight through LEs in standing with minimal support at trunk for balance	Unable to ambulate with or without assistance
4	M	Caucasian	24mo	Independent with static and dynamic sitting	Able to pull to stand and maintain standing with UEs supported	Able to take 2-3 steps with bilateral hand-held assistance
5	F	Caucasian	11mo	Independent with static sitting, loss of balance with dynamic sitting	Able to bear weight through LEs in standing with moderate support at trunk for balance	Unable to ambulate with or without assistance
6	M	Caucasian	18mo	Independent with static and dynamic sitting	Able to pull to stand and maintain standing with UEs supported	Unable to ambulate with or without assistance

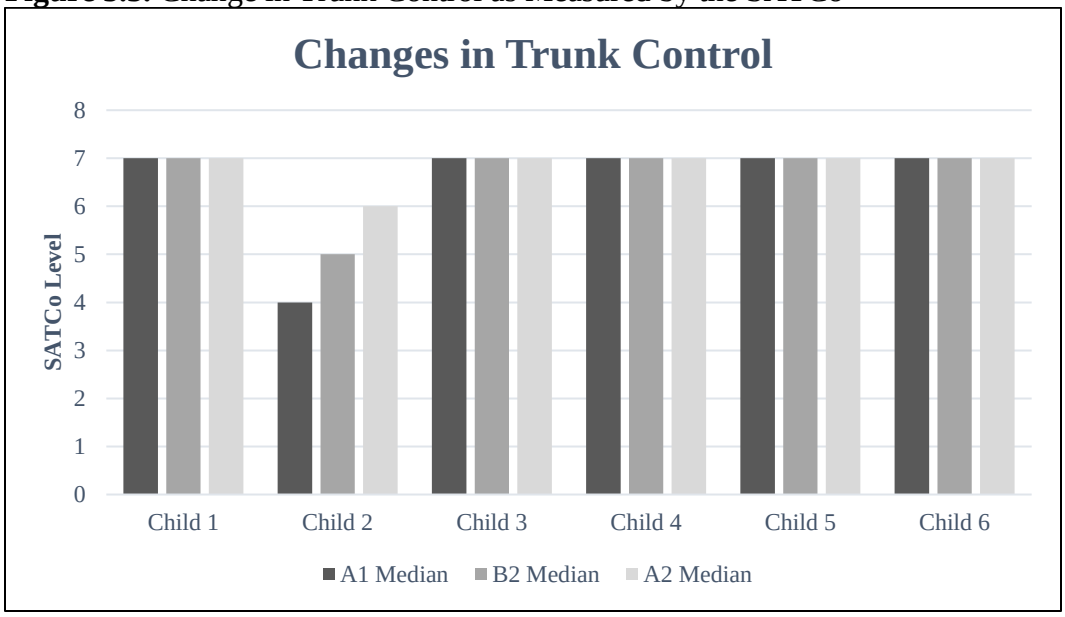
The target total amount of active time in the device was 30 minutes per day, five days per week, for six weeks (900 minutes). The parents of the participants were consistent in completing the daily log, but the total time spent using the device varied from 659 minutes (Child 2) to 1444 minutes (Child 1), with an average of 920.67 minutes (SD = 251.39). A summary of device use by each child can be found in Table 5.2.

Table 5.2. Home Program Use of the Dynamic Standing Device from Parent Log

Child Number	**Average # of days per week using the device**	**Average time spent in device per daily session (minutes)**	**Total time in device over the entire intervention (minutes)**
1	7	41.25	1444
2	5.17	21.26	659
3	5.17	26.06	808
4	5.83	27.85	975
5	5	27.83	835
6	4.5	29.74	803

Children were tested weekly on the SATCo. Four of the children reached the maximum level of the SATCo before the start of the study. Child 1 obtained the maximum SATCo score during the baseline phase (A_1) after one week in the study. The median SATCo level scores for each phase are presented for all six children in Figure 5.3. Visual inspection of the data reveals that trunk control did not change significantly with use of the dynamic standing device. All SATCo level values can be seen in Table 5.3.

Figure 5.3. Change in Trunk Control as Measured by the SATCo



*SATCo = Segmental Assessment of Trunk Control

Table 5.3. SATCo Level Scores for Each Child by Week

Week 1	Child 1	Child 2	Child 3	Child 4	Child 5	Child 6
1 (A_1)	6	5	7	7	7	7
2 (A_1)	7	4	7	7	7	7
3 (A_1)	7	2	7	7	7	7
4 (A_1)	7	4	7	7	7	7
5 (B)	7	5	7	7	7	7
6 (B)	7	4	7	7	7	7
7 (B)	7	5	7	7	7	7
8 (B)	7	5	7	7	7	7
9 (B)	7	6	7	7	7	7
10 (B)	7	6	7	7	7	7
11 (A_2)	7	6	7	7	7	7
12 (A_2)	7	6	7	7	7	7
13 (A_2)	7	6	7	7	7	7
14 (A_2)	7	7	7	7	7	7

GMFM raw total scores for the participants are presented in table 5.4. Graphed data of GMFM scores for each participant are presented in Figures 5.4 through 5.9. The graphs of GMFM scores over time show a significant improvement in gross motor skills

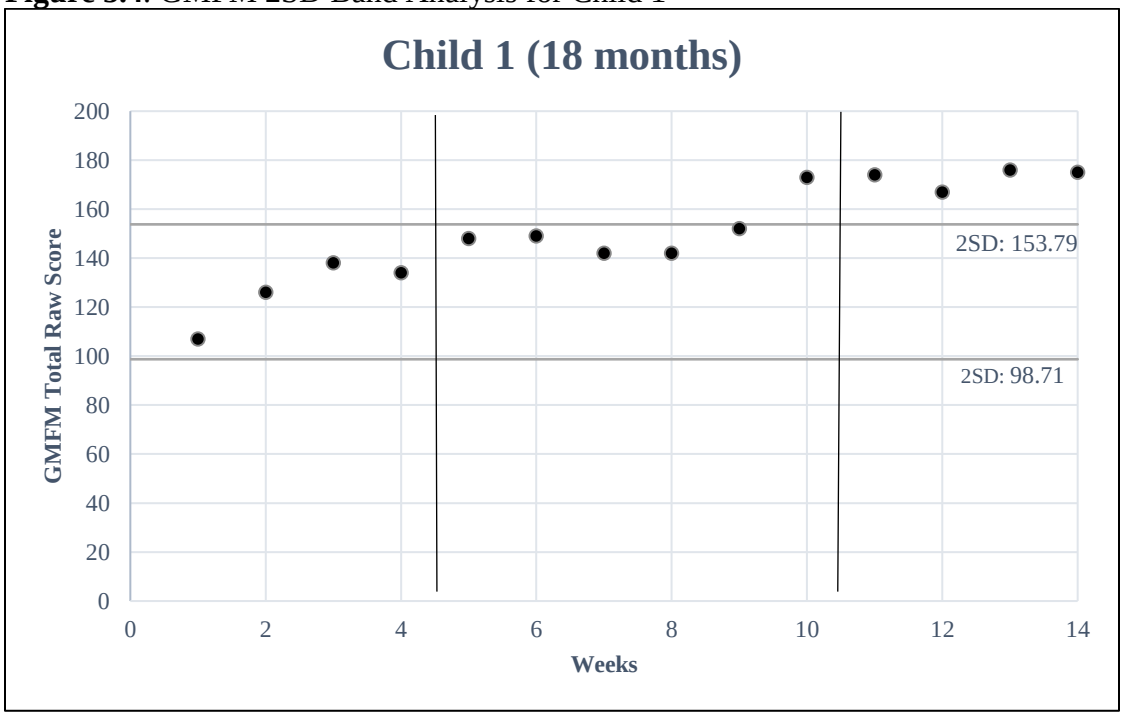
for Child 2, 3, and 5 during the intervention phase (B) compared to the baseline phase (A_1). This is indicated by at least two data points above the 2SD band. The improvement in GMFM scores for these three participants was maintained for the duration of the study. Child 1, 4, and 6 did not show significant improvements in gross motor skills from phase A1 to phase B. All six children had at least two data points above the 2SD band during the second baseline phase (A_2). The parents and therapists involved in the study reported no adverse effects.

Table 5.4. GMFM Raw Total Scores

Participant	**Phase A_1** 4 trials (Mean, SD, 2SD)	**Phase B** 6 trials (Mean, SD, 2SD)	**Phase A_2** 4 trials (Mean, SD, 2SD)
Child 1	126.25 13.77 27.54	151.00 11.49 22.98	173.00* 4.08 8.16
Child 2	47.25 4.03 8.06	58.50* 7.18 14.35	75.00* 7.39 14.79
Child 3	100.25 7.50 15.00	119.83* 10.15 20.29	150.00* 8.49 16.97
Child 4	108.50 7.19 14.38	111.00 5.17 10.35	128.25* 10.24 20.49
Child 5	76.00 6.27 12.54	93.17* 8.08 16.17	107.50* 0.58 1.15
Child 6	146.50 6.35 12.70	150.00 5.87 11.73	162.25* 3.20 6.40

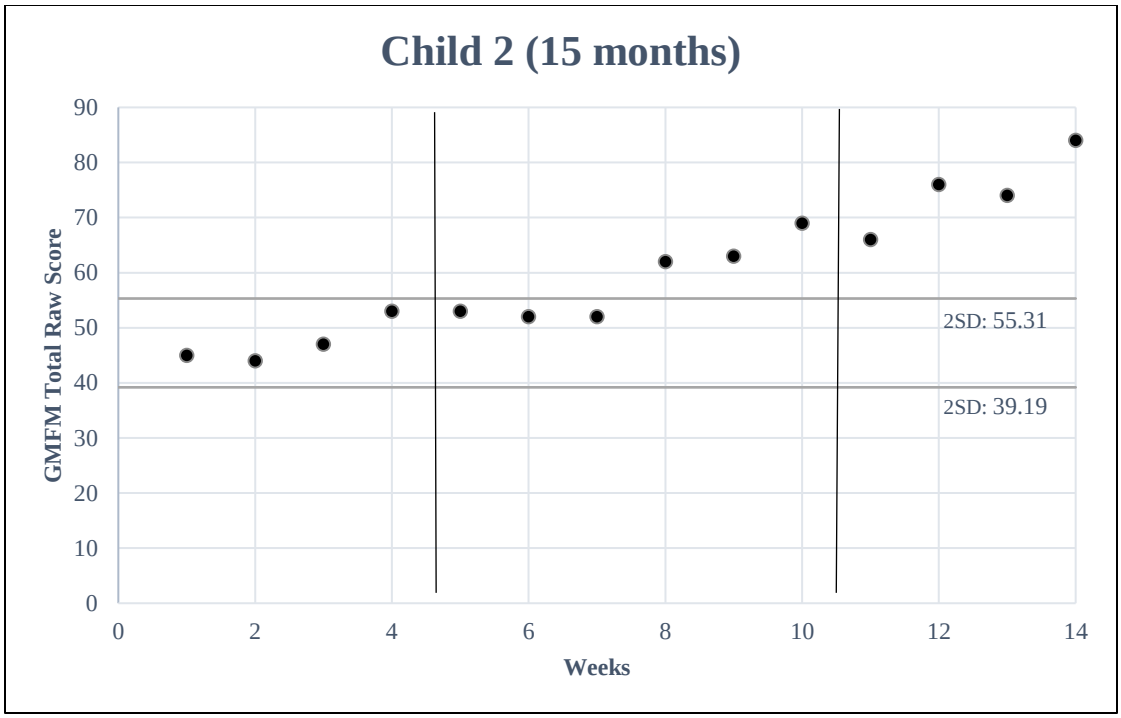
GMFM = Gross Motor Function Measure, SD = Standard Deviation, 2SD = Two Standard Deviations
*Indicates significance above 2SD

Figure 5.4. GMFM 2SD Band Analysis for Child 1



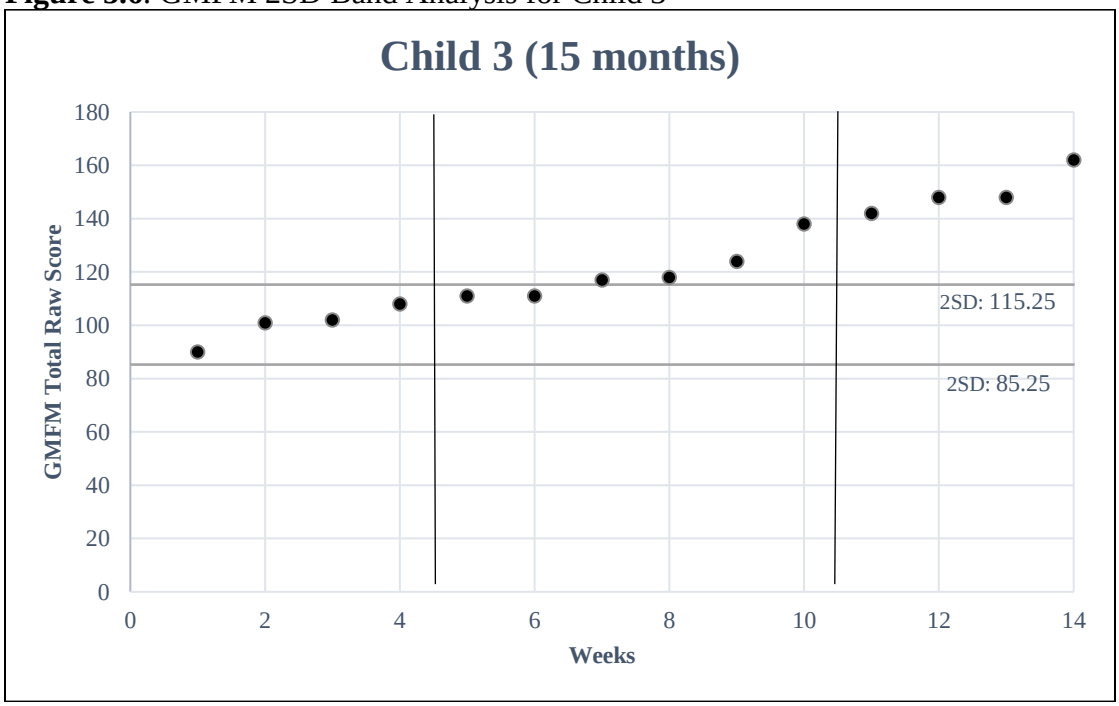
*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Figure 5.5. GMFM 2SD Band Analysis for Child 2



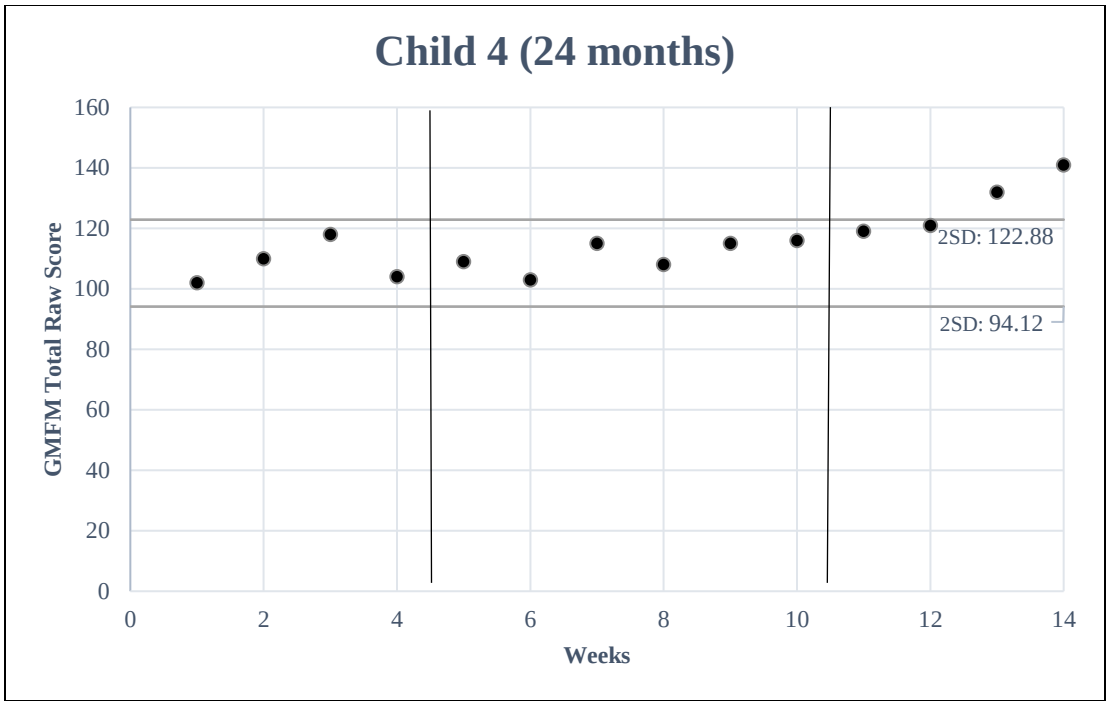
*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Figure 5.6. GMFM 2SD Band Analysis for Child 3



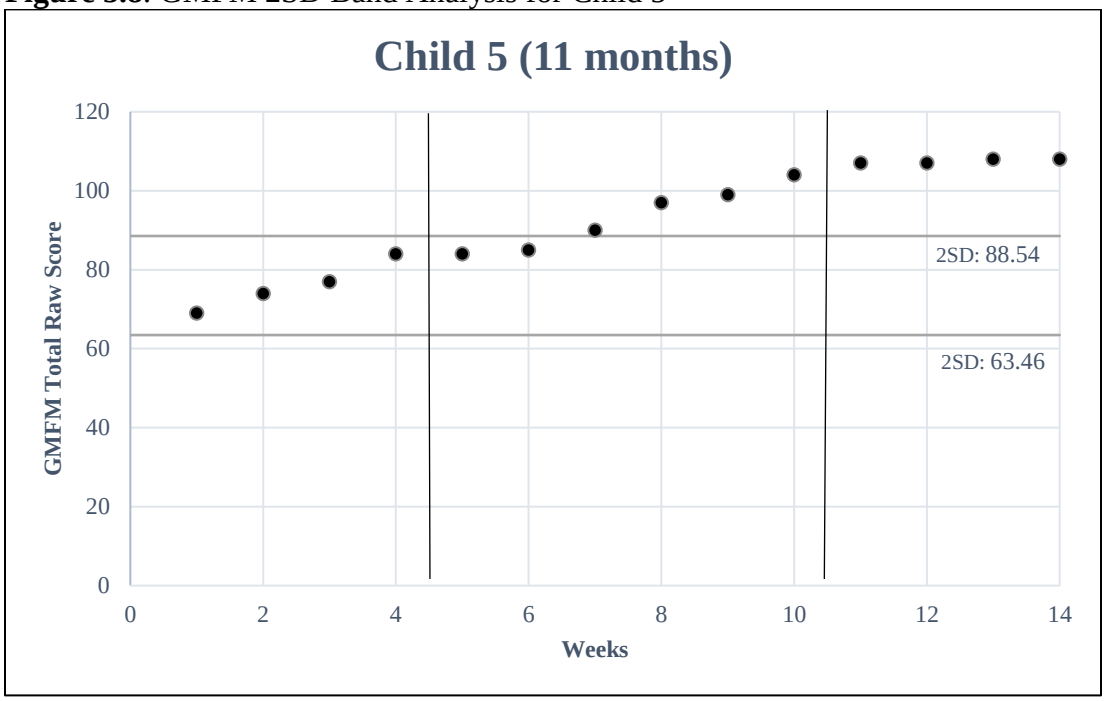
*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Figure 5.7. GMFM 2SD Band Analysis for Child 4



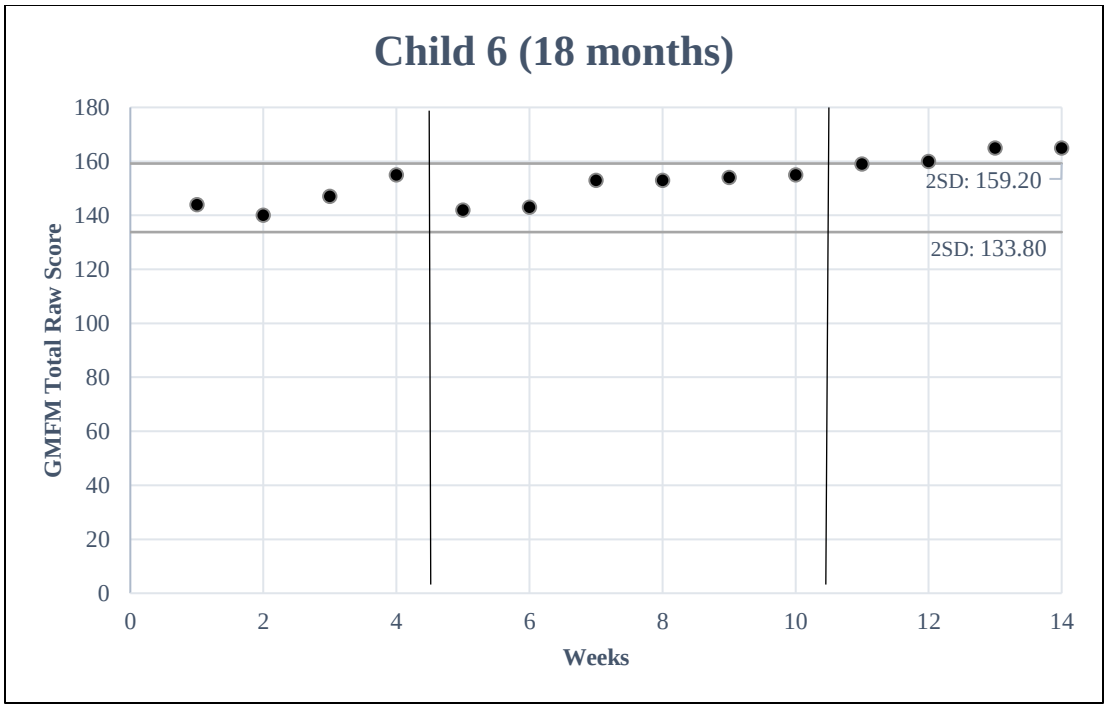
*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Figure 5.8. GMFM 2SD Band Analysis for Child 5



*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Figure 5.9. GMFM 2SD Band Analysis for Child 6



*GMFM = Gross Motor Function Measure, 2SD = Two Standard Deviations

Friedman's ANOVA results revealed a significant change from the start of the study, A_1 to the end of the study, A_2 ($\chi^2(2) = 12$, $p = 0.002$). Follow-up pairwise comparisons revealed no significant change from A_1 to B (T = -1, $p = 0.25$) or from B to A_1 (T = -1, $p = 0.25$). However, there was a significant change from A_1 to A_2 (T = -2, $p = 0.002$).

DISCUSSION

The results of this study suggest that a dynamic standing device, the Upsee, is a feasible home program intervention for children with DS ages 11 to 24 months. Overall, the average GMFM scores improved from A_1 to A_2. Three of the children (Child 2, 3, and 5) who participated in the study showed significant gross motor improvements when using the device. This change was maintained over time. These three children were the

youngest of the participants (ages 11 to 15 months) and none had completed the recommended total time for the home program (900 minutes). Of the three children who did not show a significant improvement from baseline to intervention phase, two of the families (Child 1, age 18 months, and Child 4, age 24 months) performed more than the recommended amount of time using the device. Prior to starting the study, these two children were both able to pull up to standing from a sitting position.

Child 2 did not demonstrate full trunk control as measured by the SATCo until week 14, but he was able to maintain independent sitting when placed by week 9. His change in trunk control cannot be solely attributed to the use of the Upsee because his gradual attainment of trunk control may have been due to natural maturation. Four of the children demonstrated full trunk control before the start of the study and one gained full trunk control during the first baseline phase; therefore, no conclusions about the impact of the device on trunk control in this population can be drawn.

By the end of the study, all children improved in their gross motor skills beyond what would be expected as indicated by the 2SD band. All parents reported that they saw a benefit to using the Upsee, and unprompted, two families (Child 2 and Child 6) said they would continue to use it after the study concluded. The parent of Child 3 said her child was "trying to pull to stand more without the Upsee" during the last week of the intervention phase. During the fourth week of the intervention phase, the parent of Child 5 reported that she noticed an improvement in her movements and could feel her child pushing and standing straight up for longer periods of time while using the device. She

also commented that her child was “putting more weight in her legs when not using the Upsee.” None of the children were independently walking by the end of the study.

There were no adverse effects from using the device. A majority of families (Child 2, 3, 5, and 6) reported that allocating 30 minutes per day to perform the activities was difficult and they were unable to complete the recommended time commitment. Parents of the youngest participants (Child 2 and Child 5) also reported that their children were sometimes fussy before the 30 minutes was over. These parents reported they would divide the time into shorter segments or not complete the full time. Half of the participants (Child 2, 3, and 5) were sick for a short time during the intervention phase of the study.

The dosage for the dynamic standing home program was taken from clinical experience and previous studies,[18,19,82] with parental input, age of the children, and functional level taken into consideration. The six-week intervention phase was chosen to imply that changes seen were due to the intervention and not from natural maturation effects. The most positive changes were seen in three children who were unable to pull to stand prior to the start of the study and completed less than the recommended dosage using the device. This suggests that less time using the Upsee for children not yet able to pull to stand may be as effective in improving gross motor outcomes.

The children who appeared to benefit most from the dynamic standing device were those who were less than 18 months old with an initial GMFM total score of equal to or less than 90. Functionally, these children were unable to pull up to standing and varied in their ability to maintain sitting posture. Because children must also be able to

bear weight through their legs to use the dynamic standing device, this evidence suggests a small window in which the Upsee may be most effective.

This study has several strengths. Recruiting infants and toddlers with DS between the ages of nine to 30 months old presented a challenge for a traditional randomized controlled study; therefore, a single case experimental design was employed in which six participants served as their own control. This design used purposeful manipulation of the independent variables across three phases, with weekly measurement of dependent variables, making it a rigorous alternative to a large clinical trial.[21,22] Additionally, the physical therapist performing the supervised home program for this study was blinded to the results of the weekly testing sessions.

One limitation in this study was the small sample size, making the results difficult to generalize to a larger population of children with DS. Additionally, the participants may have benefitted from a longer intervention period because most showed improvements toward the end of the intervention phase. Perhaps they would have shown greater gains from more weeks using the dynamic standing device. Consistency in implementation of the home program was also an issue. There was wide variability in the amount of time each family spent using the dynamic standing device and how many days per week they used the device. Because of this variability, drawing conclusions on optimal dosage for this device is difficult.

Future studies should focus on the optimal age and gross motor skill level to use the dynamic standing device, as well as the optimal dosage for the device. A comparison of other home standing programs with the Upsee dynamic standing home program would

also be useful. A multiple baseline single-case experimental design that systematically applies the intervention in a sequential manner with a staggered introduction across participants would be a good alternative to a randomized controlled trial if recruitment is an issue.

CONCLUSION

The Upsee is a dynamic standing device that can be implemented by physical therapists into a supervised home program for children with DS (ages 11 to 24 months) who are not yet walking but are able to bear weight through their LEs. The impact of this device on trunk control in this population is not known, but it appears to improve gross motor skills in some children with DS between the ages of 11 to 24 months.

CHAPTER VI

CONCLUSION

STATEMENT OF THE PROBLEM

DS, which occurs in approximately 12.6 per 10,000 live births in the United States,[1] is associated with gross motor skill delay.[1-4] Trunk control is a precursor to upright gross motor skills; therefore, assessing trunk control in infants and toddlers with DS (ages six to 24 months) is necessary when applying interventions for improving the acquisition of gross motor skills. The GMFM has proven to be a reliable and valid measure of gross motor skills for children with DS.[7-9] The SATCo shows promise as a simple measure that can be employed by a physical therapist in any setting to measure segmental changes in trunk control.[10] To date, the psychometric properties of the SATCo have not been studied in children with DS.

In addition to assessing trunk control in children with DS, intervention strategies for improving trunk control and gross motor skills in this population should be investigated. The Upsee[§§] shows promise as a dynamic standing device that can be used at home by the parent and child to promote upright mobility. This device uses a child harness connected to an adult hip belt with shared sandals for promoting upright activities in young children. The literature suggests that this dynamic standing device may lead to

[§§] Firefly by Leckey, Lisburn, Northern Ireland

improved trunk control and gross motor skills,[5] but its impact has not been studied in infants and toddlers with DS (ages six to 24 months).

REVIEW OF METHODOLOGY

Two methodological studies explored the psychometric properties of the SATCo in infants and toddlers with DS ages six to 24 months. Reliability, concurrent validity, and the predictive ability of the SATCo on the GMFM were examined. A withdrawal/reversal single-case experimental design study investigated the impact of a home program using a dynamic standing device on gross motor skills and trunk control in infants and toddlers with DS ages 11 to 24 months.

SUMMARY OF FINDINGS

The ICC (2,1) results showed the SATCo had moderate to good interrater reliability and good to excellent intrarater reliability in infants and toddlers with DS ages six to 24 months. Concurrent validity of the SATCo was established through good to excellent significant correlations with the GMFM. Additionally, SATCo total score was a significant predictor of GMFM total score.

The dynamic standing home program using the Upsee did not significantly change SATCo scores in infants and toddlers with DS ages 11 to 24 months, most of whom received the maximum score on the SATCo prior to starting the intervention. Half of the participants significantly improved in their GMFM scores, but the other half of the participants showed no significant gross motor skill changes. None of the children had adverse effects from the device.

CLINICAL RELEVANCE

This study suggests that the SATCo is a clinically useful tool that physical therapists can employ to measure trunk control in infants and toddlers with DS ages six to 24 months. The results suggest that the SATCo is very reliable when used by the same clinician to reassess trunk control, but reliability decreases when two different clinicians use the SATCo to measure trunk control in the same child. The SATCo is faster to perform than the GMFM, taking about one fourth of the time to administer, but does not provide as much functional detail as the GMFM. Because the SATCo correlates well with the GMFM and is a significant predictor of GMFM total score, physical therapists may choose to employ the SATCo when time is an issue. The SATCo may also be useful when screening infants and toddlers ages six to 24 months with DS for gross motor skill delay. Clinicians may choose to insert the child's SATCo scores and age into the predictive equation and get an idea of the child's overall GMFM score; however, this equation should be used with caution. The equation should not be used in children who have attained full trunk control or are able to walk independently.

The SATCo appears to be useful in infants and toddlers with DS who are less than 24 months of age or who are not yet able to maintain static sitting balance. The tool appears to have a ceiling effect; therefore, caution should be used when employing the SATCo to measure trunk control or predict gross motor abilities in older children with DS or those who have established upright dynamic sitting balance.

When used as a dynamic standing device, the Upsee can be used by physical therapists in a supervised home program for infants and toddlers with DS ages 11 to 24

months who are not yet walking. The impact of this home program on trunk control in this population is not known, but it appears to improve gross motor skills in some infants and toddlers with DS ages 11 to 24 months without adverse effects on gross motor development. The results of this research indicate that for children with DS who are able to bear weight through their lower extremities but are not yet walking, use of the Upsee in a dynamic standing home program is beneficial. The dynamic standing device also appears to be most useful as a home program in children with DS who are less than 18 months old with a total GMFM score of less than 90. The impact of the Upsee on SATCo scores could not be measured because of the ceiling effect of the SATCo. Parents report they enjoyed using the Upsee. A longer home program, perhaps using the Upsee until the child is able to take three independent steps, may be more beneficial for improving gross motor skills.

IMPLICATIONS FOR THE FUTURE

Prior to this research, SATCo psychometrics were unknown in this population. More research is needed to support the initial findings from these studies and investigate further the psychometrics of the SATCo in infants and toddlers with DS. For example, clinicians would benefit from information on the responsiveness of the SATCo and the predictive ability of the SATCo for functional mobility in children with DS. Because the SATCo has a ceiling effect, as children with DS get older, the SATCo will no longer predict GMFM total scores. Research is needed to explore the ceiling effect of the SATCo in this population, including cut-off ages.

Additionally, future studies in infants and toddlers with DS should focus on the optimal age or gross motor skill level to use Upsee, as well as the optimal dosage for the dynamic standing device. Using the Upsee as a walking device (not just a dynamic standing device) warrants exploration as well. Future research should explore other viable home programs that supplement physical therapy to improve the trunk control and gross motor skills in this population.

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