The effect of treadmill training and supramalleolar orthoses on gait and upright play development in infants with Down syndrome

by

Julia Elizabeth Looper

A dissertation submitted in partial fulfillment

of the requirements for the degree of

Doctor of Philosophy

(Kinesiology)

The University of Michigan

2008

Doctoral Committee:

Professor Dale A. Ulrich, Chair

Professor Beverly D. Ulrich

Associate Professor Daniel P. Ferris

Assistant Professor Joseph E. Hornyak

Dedication

This dissertation is dedicated to Brian.

Without his love and encouragement, it would not have been possible.

Acknowledgements

First, I would like to acknowledge and thank my family. Brian, Audrey, Jake, and my parents and siblings have put up with me and helped keep me sane during the past 5 years. I would also like to acknowledge my committee members. They have all been willing to take time to meet with me and guide me on my journey to a PhD and beyond. I would especially like to thank Dale, my advisor and chair, whose feedback has been invaluable. The experiences that I have gained while in his lab have opened my eyes to the world of developmental research. Many friends have also supported me here at Michigan. I would especially like to thank my "accountability group," Beth Crane and Antoinette Domingo. They both helped keep me focused and on track (even if our meetings were not always business).

My studies at the University of Michigan were supported by a Leadership Training Grant from the U.S. Office of Special Education Programs and by scholarships from the Foundation for Physical Therapy, Inc and the Michigan Physical Therapy Association.

Table of Contents

List of Figures

List of Tables

List of Appendices

List of Abbreviations

Down syndrome (DS)

Foot orthoses (FOs)

Gross Motor Function Measure (GMFM)

International Classification of Functioning, Disability, and Health (ICF)

Pediatric Evaluation of Disability Inventory (PEDI)

Supramalleolar orthoses (SMOs)

Typical development (TD)

Treadmill training (TT)

Abstract

The effect of treadmill training and supramalleolar orthoses on gait and upright play development in infants with Down syndrome

Looper, J

This study explores whether the use of supramalleolar orthoses (SMO) in combination with treadmill training (TT) leads to improved age at walking onset, improved upright play skills, and improved gait parameters in infants with Down syndrome (DS) when compared to TT alone.

Infants entered the study when they could pull to stand but not yet cruise and were randomly assigned into the control (n=7) or experimental group (n=10). Infants in the control group received TT 5 days/wk for 8 min/day at a speed of 0.2 m/s. Infants in the experimental group wore SMOs for 8 hours/day in addition to TT. During the study, researchers visited the infants' homes monthly. At these visits, 3 minutes of treadmill stepping were videotapes and each child's motor development was updated via administration of the GMFM and PEDI. A 20-minute upright play session at an infant activity table was also videotaped during the odd numbered visits. The TT portion of the study ended when each child took 3-independent steps. At that point, infants in the control group were measured for orthoses, which they received 1-2 weeks later. One month after walking onset, each infant came to the lab for gait analysis with and without orthoses.

Results show that while the long-term orthotic use led to improved gait parameters at walking onset and improved balance in upright play, it also led to negative effects on other developmental outcomes including poorer performance on adaptive motor skills and decreased exploratory play in upright. This negative effect on overall development outweighs the positive effects of orthoses in this young population. SMO use should be postponed in children with DS until they have an established gait pattern and adaptive motor skills.

Chapter 1

Introduction

Orthoses are a commonly used, though unproven intervention in infants and young children with Down syndrome (DS). Health care professionals often use orthoses in an effort to influence the age at walking onset and to improve gait in infants and toddlers with DS. They hypothesize that the orthoses will improve gait and balance function by limiting hypermobility at the foot and ankle. Though interventions that address balance and gait abnormalities in children with DS are often used to treat impairments, they may have further reaching functional effects.

Down syndrome is the most common genetic disorder. It is a genetic disorder named after John L. Down, who initially described the syndrome in 1866 (Down, 1866). Extra chromosomal material from the 21st chromosome causes DS. Though an extra 21st chromosome (trisomy 21) occurs in 95% of DS cases, translocation or mosacism cause 5% of DS cases (Jones & Smith, 2006). Translocation occurs when a portion of the 21st chromosome breaks off and attaches to a different chromosome. In mosaic DS, only certain populations of cells contain the extra chromosome. DS occurs in 13.65 out of every 10,000 live births and more frequently for women of advanced maternal age, reaching 1 in 50 for women over 45 (CDC, 2006).

The extra chromosomal material associated with DS leads to a highly recognizable phenotype. Children with DS tend to be of small stature with hypotonia and

hyperflexible joints. They have a small nose with a low nasal bridge. Children with DS also have inner epicanthal folds. In addition to these facial features, children with DS often have a single palmer crease on their hands and a large gap between their first and second toes (Jones & Smith, 2006). Besides the outward signs of DS, children often have other impairments: 70% of children with DS have myopia, 66% percent experience some form of hearing loss, and approximately 40% express cardiac abnormalities (Jones & Smith, 2006). Because the DS phenotype is so easy to recognize, those children not diagnosed prenatally usually receive a diagnosis shortly after birth.

In addition to these phenotypic charateristics, children with DS also have common impairments. One such impairment is hypotonia. Many agree that hypotonia is found in all children with Down syndrome (Jones & Smith, 2006; American Academy of Pediatrics, 2001). It presents itself as extreme active and passive range of motion (Lydic & Steele, 1979). Though most researchers agree that hypotonia exists in children with DS, there is no such agreement on what hypotonia is and it is therefore measured in multiple ways (Lacquaniti, 2000; Morris, Vaughan, & Vaccaro, 1982; Prechtl, 2001). If we assume that hypotonia is lowered resistance to an externally applied force, then we can assume that ligamentous laxity, lower inertia of body segments, and increased tendon compliance may all contribute (Latash, Wood, & Ulrich, in press). In addition, the neuromuscular system in people with DS produces slower muscular reactions to perturbations than is typical (Schumway-Cook & Woollacott, 1985). Latash and colleagues (Latash, Wood, & Ulrich, in press) theorize that this is due to deeper levels of muscle relaxation and co-activation patterns during muscle contraction in people with DS and may result in the perception of low muscle tone on the part of the examiner.

Another common impairment in children with DS is ligamentous laxity. This causes hypermobility, increased movement around a joint, and instability at the ankle. In an unstable situation, the top of the heel bone, the calcaneus, rolls medially. The calcaneus is the posterior base of the longitudinal arch of the foot; if misaligned, the arch collapses and the foot cannot properly support the body's weight during stance (Hoffinger, 1996). When the calcaneus rolls in this manner, it is called calcaneal eversion (Parker & James, 1985). Calcaneal eversion forces the arch of the foot to collapse because the ankle must rely primarily on lax ligaments for stability instead of the boney structure of the foot and ankle. This impacts both balance and gait in children with DS (MacNeill-Shea & Mezzomo, 1985). Children with DS display poor balance (Schumway-Cook & Woollacott, 1985) and a variable gait pattern (Looper, Wu, Angulo Barroso, Ulrich, & Ulrich, 2006). Excessive calcaneal eversion could contribute to these impairments by preventing the foot from adequately supporting the body's weight. Ligamentous laxity may affect function in addition to simply causing hypermobility at the ankle.

Motor delay and decreased activity are influenced by poor ankle alignment due, in part, to hypermobility (Caselli, Cohen-Sobel, Thompson, Adler, & Gonzalez, 1991). In children with DS, hypermobility is also associated with other orthopedic impairments, including pes planus (flat foot) (Caselli et al., 1991). Hypermobile children must learn to control excessive joint movement and must use more energy to execute this control than children with tight joints. This makes continuous walking strenuous and leads to atypical gait patterns (Selby-Silverstein, Hillstrom, & Palisano, 2001), poor balance skills (Martin, 2004), and increased energy requirements at terminal stance (Cioni, Cocilovo,

Rossi, Paci, & Valle, 2001). In order to address motor delay, abnormal gait, decreased balance and decreased endurance, many therapists turn to orthoses to counter hypermobility and support the foot and ankle in a neutral position.

Gait and Balance in children with Down syndrome

Exploration is important for multiple aspects of development. In fact, the varied pattern of environmental stimulation that children obtain through exploration and play is important for optimal brain development (Rowland, 1998). The ability to move around and explore via crawling, walking, or powered mobility increases children's cognitive ability, affective behavior, and language skills (Biringen, 1995; Butler, 1985; Campos, 2000;). Playing with peers, siblings, and parents is also an important way to explore and develop social skills (National Research Council and Institute of Medicine, 2000). Walking allows children to explore the world in an upright fashion and to relate and play with peers appropriately. It is an important avenue for cognitive and social development.

Children with DS display delayed gait development. Commonly, children with DS walk by the age of 24 months, 12 months behind the average for non-disabled children (Henderson, 1986; Palisano et al., 2001; Ulrich et al., 2001). This delay appears to continue. Parker and Bronks (1980) found that 7-year olds with DS have gait patterns similar to beginning independent walkers in that they had increased stance time and lack of heel strike; non-disabled 7-year olds show a more adult-like gait pattern.

Not only do children with DS have delayed gait development, but their gait patterns also continue to be atypical throughout life. Gait parameters in the children with DS include increased stance time, early hip extension at the end of swing, forefoot initial contact, decreased plantarflexion at toe off, and out-toeing to increase lateral stability

(Parker & Bronks, 1980). In another study, Parker, Bronks, and Snyder (1986) found that 5-year olds with DS compensate for decreased walking stability by spending a greater amount of time in stance and decreasing their step length. The decreased stability is due, in part, to hypermobility in the lower extremity joints. The impairments commonly found in children with DS lead to abnormal gait patterns.

Because people with DS have hypermobile ankles, they often have ankle dysfunction, which also leads to gait abnormalities (Cioni et al., 2001) and suggests that the ankle is an important area of study and intervention in children with DS. Mature walkers with TD use their foot and Achilles tendon as a spring to store energy at initial contact and release energy at terminal stance (Fukunaga, Kubo, Kawakami, Fukashiro, Kanehisa, & Constantinos, 2001). Due to low muscle tone, people with DS may be less able to store energy. In addition, they may be less able to take advantage of the returned energy due to pes planus. This leads to increased energy expenditure at each step and may lead to decreased endurance. Cioni and colleagues (2001) report that adults with DS make initial contact with their forefoot and have decreased plantarflexion in terminal stance, and thus have decreased saggital plane range of motion during each gait cycle. They further explain that these impairments at the ankle lead to decreased energy storage at impact and decreased force production at push off. As a result, walkers with DS must do more work during each gait cycle than non-disabled walkers, due to less-than-ideal function of the ankle-foot complex.

Differences in joint mobility, muscle tone, and ligamentous laxity necessitate different options for addressing movement problems (like learning to walk) in the DS population when compared to the typical population. In addition to these organismic

constraints, the dynamic resources available to children with DS differ from those available to children with TD. A dynamic resource is a potential source of energy that is available to an individual (Holt, Saltzman, Ellis, & Butcher, 2001). Recently, differing sue of dynamic resources have been suggested as a reason for atypical gait patterns in the DS population (Kubo & Ulrich, 2006). Recent studies of preadolescents with DS show that they respond to novel tasks differently when compared to preadolescents with TD. For example, Ulrich and colleagues (Ulrich, Haehl, Buzzi, Kubo, & Holt, 2004) found that preadolescents with DS display increased stiffness and forcing when walking on a treadmill and increased forcing when walking over ground compared to age matched peers with TD. With practice, they develop responses that are more typical. This may also hold true for infants who are tackling the challenging task of learning to walk.

Postural control is a limiting factor in the development of motor skills (Haley, 1986). The relation between posture and motor milestone attainment appears both in typically developing children and in children with DS (Haley, 1986). Children with DS, however, are more limited by their ability to control their posture than non-disabled children (Haley, 1986). This may be because people with DS have difficulty modulating the forces necessary to respond to postural perturbations (Shumway-Cook & Woollacott, 1985).

Poor postural control is noticeable in the gait patterns of new walkers with DS. They display increased step width, decreased step length, increased double support time, and increased step length and width variability when compared to new walkers with TD (Looper et al., 2006; Parker et al., 1986). This pattern suggests that dynamic postural control may play a larger role in the gait dynamics of new walkers with DS when

compared to new walkers with TD. All children who are new walkers are in a constant state of falling when they attempt to walk. They must balance this constant instability with postural control in order to maintain upright and to continue walking (Bril & Breniere, 1993). Young children who do not have a mature gait pattern spend less time in the single leg support phase of gait. Similarly, children with DS also spend less time on one foot than do typically developing children and children with other forms of intellectual disability of the same age (Kokubun, Shinmyo, Ogita, Morita, & Furuta, 1997). The reduced single limb stance time in children, when compared to adults, can be interpreted as instability in children's gait (Sutherland, 1997). By spending less time on one foot while walking, children display increased double limb stance. This allows the children more time to regain their balance between steps.

In addition to single limb stance time, sway and amplitude of sway are larger in young children than in adults (Riach & Hayes, 1987). The children increase their base of support to compensate for this increased sway and to decrease their chances of falling to the ground. This occurs in quiet stance as well as during gait. As the children's experience increases, their step width decreases (Bril et al., 1993). However, older children with DS continue to display atypical postural control. They sway closer to their limits than typically developing children do in static standing (Shumway-Cook & Woollacott, 1985) and their step width continues to remain wider than typically developing children (Parker et al., 1986). Slower onset latencies, as described by Shumway-Cook and Woollacott (1985), also lead to increased sway and make a wide base of support a necessary compensation for children with Down syndrome. The use of orthoses may compensate for weakness and slow onset latencies around the ankle by

providing support and limiting the available range of motion leading to improved upright balance.

Orthoses

As mentioned earlier, ligamentous laxity leads to calcaneal eversion in many people with DS. Orthoses are external devices that are placed in the shoes to provide support to the foot and ankle. They help prevent calcaneal eversion by supporting the calcaneus in an upright position, thus improving the bony alignment of the foot and ankle, and influencing postural and gait characteristics (Orner, Turner, & Worrell, 1994). In a review of biomechanical management of children with DS, Caselli and colleagues (1991) suggest that anterior/posterior instability should be managed early with appropriate shoes, orthoses, and physical therapy.

In addition to facilitating alignment, orthoses have an impact on shock absorption and proprioceptive input. Shock absorption at the foot and ankle is easily altered. Simply adding a neoprene insert to a shoe leads to increased shock absorption and lowers the rate of overuse injuries in typical adults (Schwellnus, Jordaan, & Noakes, 1990). However, Robbins and Waked (1997) found that too much padding led to increased landing forces around the joints in athletic males. On the other hand, orthoses made from firm but flexible materials lead to improved ankle proprioception as well as improved function in adult male golfers (Stude, & Brink, 1997; Stude & Gullickson, 2000). Ankle braces also lead to improved proprioception as measured by joint angle matching (Jeroch, Hoffstetter, Bork, & Bischof, 1995) and increased stretch reflex excitability (Nishikawa, Ozaki, Mizuno, & Grabiner, 2002) in athletes who have experienced inversion ankle

sprains. Firm but flexible braces and orthsoses help the body sense how the joint is positioned in space and increase the muscles' sensitivity to differing positions.

In specific studies looking at orthotic use in children with DS, orthoses were shown to enhance ankle alignment and kinematics during gait (Selby-Silverstein et al., 2001), and improve postural stability (Martin, 2004). Selby-Silverstein and colleagues (2001) looked at the use of custom foot orthoses in children with DS and found that 3 to 6-year old children with DS show decreased ankle eversion while wearing foot orthoses in static standing. The study also demonstrated that, during walking, the use of foot orthoses resulted in decreased foot progression angle, decreased variability in this angle, a change in the initial contact site from flat foot to heel strike, and an increase in stance phase speed. Martin (2004) has also studied orthotic use in children with DS, and found that the children (age 3 years 6 months through 8 years) showed immediate and long-term improvements in balance subscale scores of standardized tests when they used a flexible supramalleolar orthoses (SMO). While these studies show that orthoses are an effective intervention in children with DS who are three and older, little published research exists on the effect of orthoses on infants and toddlers with DS.

Infants are at a critical and unique developmental time where they are learning new skills. Orthotic use at this early age may either enhance function or decrease function. If used in a young population, foot orthoses may provide a stable foot and ankle structure that could lead to earlier onset of independent walking. This increased stability may also lead to a more typical gait pattern and may initiate increased physical activity and social interactions. Conversely, the structure provided by the orthoses may negatively influence the development of the neuromotor pathways related to control of

the ankle by limiting the range and variability of practice available that are essential to the development of a new skill.

Treadmill Training

Though not much is known about effective early intervention in infants with DS, we do know that their movement differs from children with TD. Infants with DS who are as young as 3.5 months, display weakness characterized by decreased antigravity control in their lower extremities and their necks when compared to children with TD (Rast & Harris, 1985). This weakness leads to motor delay, because the infants do not have the strength to complete developmentally appropriate tasks, such as head control and frequent kicking. Because infants with DS do not kick in the same manner that typically developing infants do, they miss essential early practice of gait-like movements (Ulrich & Ulrich, 1995). This is important because kicking is kinematically similar to stepping and allows typically developing infants to practice gait-like movements before they can support their own weight (Thelen & Fisher, 1982). In addition, early practice is exploratory in nature and allows infants to learn how to effectively coordinate and control their movements (Thelen & Corbetta, 1994). Early intervention that focuses on anti-gravity kicking and gait-like movements before the child can walk, such as use of a supine kicking toy to reinforce anti-gravity movements or use of bodyweight supported treadmill training in the first year of life may help restore the effects of this lack of early practice.

Infants are born with a stepping response that, under clinical testing procedures, disappears approximately 6 weeks after birth (McGraw, 1940). However, Thelen and colleagues (1984) discovered that the stepping response does not disappear because of

increasing neural maturation, as previously believed. Rather it is inhibited by infants' increasing leg mass, making them hard to lift when held upright. In addition, infants perform a very similar movement when kicking in supine, a pattern that does not disappear (Thelen & Fisher, 1982). When 3-5 month old infants with TD are placed in supported standing on motorized treadmills, they begin to take steps and by six to seven months can consistently perform supported voluntary stepping (Thelen, 1992; Thelen & Ulrich, 1991). When infants take steps on the treadmill early in the first year, they show unilateral, parallel, double, and alternating steps (Thelen & Ulrich, 1991). As infants' treadmill stepping frequency increases, they settle into alternation as the dominate pattern (Ulrich, Jensen, & Thelen, 1991). Infants with DS display a similar response to the treadmill but do so at a later chronological age. Over time, pre-walking infants with DS increase the frequency with which they step and the proportion of alternating steps (Ulrich, Ulrich, Collier, & Cole, 1995).

Early rigorous practice of treadmill stepping leads to earlier walking onset in children with DS (Ulrich, Lloyd, Tiernan, Looper, & Angulo-Barroso, 2008; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001). In a study by Ulrich and colleagues (2001), treadmill training was used as a supplement to physical therapy. Parents held their infants on a treadmill to train stepping behavior before they were able to walk independently. The treadmill training provided infants with repeated opportunities to explore patterns of leg movements in an upright posture long before walking onset occurred. After children were able to walk independently, their spatiotemporal gait parameters were evaluated. The children who received treadmill training walked independently 101 days earlier than a control group of children with DS who were not

receiving the treadmill intervention. The average chronological age at walking onset was 23.9 months in the control group and 19.9 months in the intervention group. The intervention group also displayed improved gait parameters including step width and dynamic base at the onset of independent walking. A second training study further validated the benefits of treadmill training for infants with DS (Ulrich et al., 2008). Treadmill training appears to be a well-researched, effective way to improve age of walking onset and gait parameters in infants with DS.

International Classification of Functioning, Disability, and Health

Researchers and clinicians use outcome measures to determine if a change has taken place and to determine if an intervention is successful. Earlier attainment of independent walking is an important outcome measure, but only one of many possibilities. The International Classification of Functioning, Disability, and Health (ICF) model (WHO, 2001) is a tool designed to help clinicians characterize multiple levels of outcomes. The ICF disablement model consists of three levels of functioning: bodily functions and structures, activity, and participation. Abnormalities in the body functions and structures level results in physiological or anatomical impairments. In the case of children with DS, this includes poor ankle alignment from ligamentous laxity and weakness. The next level of this model, activity limitation, results from the inability of an individual to perform a task; this would include a child's ability to achieve motor milestones. The final level, participation limitation, results from the individual's inability to participate fully in society. This includes the ability to perform daily living skills and to interact with friends and family members. In this study, ankle alignment falls under body function and structure, motor milestones and gait pattern fall under activity, and play is in the participation category. This research will focus on the use of foot orthoses

as a means to improve functional mobility in children with DS. By providing a device that addresses impairment level problems and an intervention with the device that includes the activity and participation level, this study is able to look at multiple levels of disablement. By intervening at the first two levels with orthoses and treadmill training, I hope to see larger improvements in activity and participation than with treadmill training alone.

Purpose

While the body of literature is growing, research into specific early interventions for children with DS remains limited. An even greater gap exists concerning orthotic intervention in children with DS. This investigation seeks to determine whether activity and participation level skill development improves by combining treadmill training with orthotic use in infants with DS. More specifically, are there benefits of early orthotic use that may make it an appropriate intervention in infants and toddlers such as decreased age at walking onset, improved gait parameters at walking onset, and improved upright play skills? It is expected that: A) Infants who wear orthoses while learning to walk will walk earlier than the infants who do not wear orthoses. B) Infants wearing orthoses will spend more time in using their hands for play during a 20-minute play task than those without orthoses. C) New walkers who wore orthoses during treadmill training will display better gait compared to new walkers who did not wear orthoses. Overall, treadmill training, an activity level intervention, in combination with orthotic use, a body function and structure level intervention, will lead to improved gait development as well as an increase in participation level skills.

References

American Academy of Pediatrics, Counsel on Genetics (2001). American academy of pediatrics: health supervision for children with down syndrome. *Pediatrics*, 107(2), 442-449.

Bril, B. & Breniere, Y.(1993). Posture And Independent Locomotion In Early Childhood: Learning To Walk Or Learning Dynamic Postural Control? In G. J. P. Savelsbergh (Ed.), *The Development Of Coordination In Infancy* (Vol. 97, Pp. 337-358). Amsterdam: Elsevier Science.

Butler, C. (1986). Effects of powered mobility on self-initiated behaviors of very young children with locomotor disability. *Developmental Medicine & Child Neurology*, 28(3), 325-332.

Caselli, M. A., Cohen-Sobel, E., Thompson, J., Adler, J., & Gonzalez, L. (1991). Biomechanical Management Of Children And Adolescents With Down Syndrome. *Journal Of The American Podiatric Medical Association, 81*(3), 119-127.

CDC. (2006). Improved National Prevalence Estimates For 18 Selected Major Birth Defects - United States, 1999-2001. *Morbidity Mortality Weekly Report, 54*(51/52), 1301-1305.

Cioni, M., Cocilovo, A., Rossi, F., Paci, D., & Valle, M. S. (2001). Analysis Of Ankle Kinetics During Walking In Individuals With Down Syndrome. *Americal Journal Of Mental Retardation, 106*(5), 470-478.

Down, J. (1866). Observations On An Ethnic Classification Of Idiots. *Clinical Lecture Reports, London Hospital, 3*, 259-262.

Fukunaga, T., Kubo, K., Kawakami, Y., Fukashiro S., Kanehisa, H., & Constantinos, N. M. (2001). In Vivo Behaviour of Human Muscle Tendon During Walking. *Proceedings: Biological Sciences*. 268(1464), 229-233.

Haley, S. M. (1986). Postural Reactions In Infants With Down Syndrome. *Physical Therapy, 66*(1), 17-22.

Hoffinger, S. A. (1996). Evaluation And Management Of Pediatric Foot Deformities. *Pediatric Clinics Of North America, 43*(5), 1091-1111.

Holt, K. G., Saltzman, E., Ellis, T., & Butcher, R. (2001). Teaching Spasticity and Weakness From a Dynamic Systems Perspective – Part 1. *Neurology Report*, 25(3), 102-105.

Jones, K. L., & Smith, D. W. (2006). *Smith's Recognizable Patterns Of Human Malformation*. Philadelphia: Elsevier Saunders.

Kokubun, M., Shinmyo, T., Ogita, M., Morita, K., & Furuta, M. (1997). Comparison Of Postural Control Of Children With Down Syndrome And Those With Other Forms Of Mental Retardation. *Perceptual And Motor Skills, 84*, 499-504.

Kubo, M., & Ulrich, B. D. (2006). Early Stage Of Walking: Development Of Control In Mediolateral And Anteroposterior Directions. *Journal Of Motor Behavior, 38*(3), 229-237.

Lacquaniti, F. (1990). Quantitative Assessment of Somatic Muscle Tone. *Functional Neurology*, 5, 209-215.

Latash, M., Wood, L., & Ulrich, D. A. (in press). What is Currently Known about Hypotonia, Motor Skill Development, and Physical Activity in Down Syndrome. *Down Syndrome Research and Practice.*

Looper, J., Wu, J., Angulo Barroso, R., Ulrich, D., & Ulrich, B. D. (2006). Changes In Step Variability Of New Walkers With Typical Development And With Down Syndrome. *Journal of Motor Behavior, 38*(5), 367-372.

Lydic, J. S., & Steele, C. (1979). Assessment Of The Quality Of Sitting And Gait Patterns In Children With Down's Syndrome. *Physical Therapy, 59*(12), 1489-1494.

Macneill-Shea, S. H., & Mezzomo, J. M. (1985). Relationship Of Ankle Strength And Hypermobility To Squatting Skills Of Children With Down Syndrome. *Physical Therapy, 65*(11), 1658-1661.

Martin, K. (2004). Effects Of Supramalleolar Orthoses On Postural Stability In Children With Down Syndrome. *Developmental Medicine & Child Neurology, 46*(6), 406-411.

McGraw M. B. (1940). Neuromuscular Development of the Human Infant as Exemplified in the Achievement of Erect Locomotion. *Journal of Pediatrics,* 17, 747–771.

Morris A. F., Vaughan, S. E., Vaccaro, P. (1982). Measurements of Neuromuscular Tone and Strength in Down's Syndrome Children. *Journal of Mental Deficiency Research*, 26, 41-46.

Orner, C. E., Turner, D., & Worrell, T. (1994). Effect Of Foot Orthoses On The Balance Skills Of A Child With A Learning Disablility. *Pediatric Physical Therapy, 6*, 10-14.

Palisano, R. J., Walter, S. D., Russell, D. J., Rosenbaum, P. L., Gemus, M., Galuppi, B. E., et al. (2001). Gross Motor Function Of Children With Down Syndrome:

Creation Of Motor Growth Curves. *Archives Of Physical Medicine & Rehabilitation, 82*, 494-500.

Parker, A. W., & Bronks, R. (1980). Gait Of Children With Down Syndrome. *Archives Of Physical Medicine & Rehabilitation, 61*, 345-351.

Parker, A. W., Bronks, R., & Snyder Jr, C. W. (1986). Walking Patterns In Down's Syndrome. *Journal Of Mental Deficiency Research, 30*, 317-330.

Parker, A. W., & James, B. (1985). Age Changes In The Flexibility Of Down's Syndrome Children. *Journal Of Mental Deficiency Research, 29*, 207-218.

Prechtl, H. F. R. (2001). General Movement Assessment as a Method of Developmental Neurology: New Paradigms and Their Consequences. *Developmental Medicine and Child Neurology*, 43, 836-842.

Rast, M. M., & Harris, S. R. (1985). Motor Control In Infants With Down Syndrome. *Developmental Medicine & Child Neurology, 27*(5), 682-685.

Riach, C. L., & Hayes, K. C. (1987). Maturation Of Postural Sway In Young Children. *Developmental Medicine & Child Neurology, 29*(5), 650-658.

Robbins, S. & Waked, E. (1997). Balance and vertical impact in sports: role of shoe sole materials. *Archives Of Physical Medicine & Rehabilitation,* 78, 463-7.

Schwellnus, M. P., Jordaan, G., & Noakes, T. D. (1990). Prevention of common overuse injuries by the use of shock absorbing insoles. A prospective study. *American Journal of Sports Medicine,* 18, 636-41.

Selby-Silverstein, L., Hillstrom, H. J., & Palisano, R. J. (2001). The Effect Of Foot Orthoses On Standing Foot Posture And Gait Of Young Children With Down Syndrome. *Neurorehabilitation, 16*(3), 183-193.

Shumway-Cook, A., & Woollacott, M. H. (1985). Dynamics Of Postural Control In The Child With Down Syndrome. *Physical Therapy, 65*(9), 1315-1322.

Stude, D. E. & Brink, D. K. (1997). Effects of nine holes of simulated golf and orthotic intervention on balance and proprioception in experienced golfers. *Journal of Manipulative Physiological Therapy*, 20, 590-601.

Stude , D. E.& Gullickson, J. (2000). Effects of orthotic intervention and nine holes of simulated golf on club-head velocity in experienced golfers. *Journal of Manipulative Physiological Therapy*, 23, 168-74.

Sutherland, D. (1997). The Development Of Mature Gait. *Gait & Posture, 6*(2), 163-170.

Thelen, E. & Corbetta, D. (1994). Exploration and Selection in the Early Acquisition of Skill. *International Review of Neurobiology*, 37, 75-102.

Thelen, E., & Fisher, D. M. (1982). Newborn Stepping: An Explanation For A "Disappearing" Reflex. *Developmental Psychology, 18*(5), 760-775.

Thelen, E., Fisher, D. M., & Ridley-Johnson, R. (1984). The Relationship Between Physical Growth And A Newborn Reflex. *Infant Behavior And Development, 7*, 479-493.

Thelen, E., Ulrich, B. D. (1992) *Monographs of the Society for Research in Child Development*, 56(1).

Thelen, E. (1992). Development Of Locomotion From A Dynamic Systems Approach. In H. Forssberg & H. Hirschfeld, H. (Eds.), *Movement Disorders In Children* (Vol. 36, Pp. 169-173). Basel: Karger.

Ulrich, B. D., Haehl, V., Buzzi, U. H., Kubo, M., & Holt, K. G. (2004). Modeling Dynamic Resource Utilization In Populations With Unique Constraints: Preadolescents With And Without Down Syndrome. *Human Movement Science, 23*(2), 133-156.

Ulrich, B. D., Jensen, J. L., & Thelen, E. (1991). Stability And Variation In The Development Of Infant Stepping: Implications For Control. In A. E. Patla (Ed.), *Adaptability Of Human Gait: Implications For The Control Of Locomotion.* (Pp. 145-164): North-Holland.

Ulrich, B. D., & Ulrich, D. A. (1995). Spontaneous Leg Movements Of Infants With Down Syndrome And Nondisabled Infants. *Child Development, 66*(6), 1844-1855.

Ulrich, B. D., Ulrich, D. A., Collier, D. H., & Cole, E. L. (1995). Developmental Shifts In The Ability Of Infants With Down Syndrome To Produce Treadmill Steps. *Physical Therapy, 75*(1), 14-23.

Ulrich, D. A., Lloyd, M. C., Tiernan, C. W., Looper, J. E., & Angulo-Barroso, R. M. (2008). Effects Of Intensity Of Treadmill Training On Developmental Outcomes And Stepping In Infants With Down Syndrome: A Randomized Trial. *Physical Therapy, 88*(1).

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill Training Of Infants With Down Syndrome: Evidence-Based Developmental Outcomes. *Pediatrics, 108*(5), E84.

Whitt-Glover, M. C., O'neill, K. L., & Stettler, N. (2006). Physical Activity Patterns In Children With And Without Down Syndrome. *Pediatric Rehabilitation, 9*(2), 158-164.

WHO. (2001). *International Classification Of Functioning, Disability, And Health: ICF*. Geneva: World Health Organization.

Chapter 2

Motor Skill Development in Infants with Down Syndrome Who Receive Treadmill Training With and Without Supramalleolar Orthoses

Introduction

Down syndrome (DS) is a genetic disorder that occurs in 13.65 out of every 10,000 live births (CDC, 2006).It is the most common cause of developmental delay. Though DS is most commonly known for its effects on cognitive ability, children with DS also have other impairments including difficulty with sensory perception (Chen & Fang, 2005), decreased ability to localize pain (Hennequin, Morin, & Feine, 2000) and delayed and atypical motor development. On average, children with DS sit independently at 11 months, pull to stand at 17 months, and walk independently at 24 months (Henderson, 1985). These delays, separately and in combination, lead to difficulties with function and social interaction.

International Classification of Functioning, Disability, and Health

To help differentiate between motor skill outcomes and functional outcomes, the The International Classification of Functioning, Disability, and Health (ICF) model (WHO, 2001) is a helpful tool. The ICF is a framework proposed by the World Health Organization to provide a common language about health-related and functional outcomes on multiple levels. It allows us to look at the functional and social interaction

outcomes related to specific interventions. The ICF model consists of three levels of functioning: body functions and structures, activity, and participation. Abnormalities in the body functions and structures level include physiological or anatomical impairments such as poor ankle alignment from ligamentous laxity and weakness. The next level of this model, activity, looks at the individual's ability to perform a task and includes a child's ability to achieve motor milestones. The final level, participation, focuses on the individual's ability to participate fully in society. This includes the ability to play, perform daily living skills, and to interact with friends and family members (WHO, 2001).

Body Functions and Structures

Children with DS display abnormalities on the body functions and structures level of the ICF. Therapists and researchers often assume that these abnormalities affect functional skills. For example, ligamentous laxity, hypotonia, and weakness are all cited as constraints to motor development in children with DS (Sacks & Buckley, 2003). In fact, treatment is often based on improving these impairments. However, functional skills, such as walking, are only minimally affected by a single intervention at the body function and structure level (Ostensjo, Carlbedrg, & Vollestad, 2004). As a consequence, therapists cannot treat impairments alone. Impairments should be treated within the context of functional activity.

Activity

The ability to perform gross motor skills falls in the activity level of the ICF. Infants with DS show delays in early motor skills (Palisano, 2001). These delays

continue to increase as the child grows older (Henderson, 1985). In addition to motor skill delay, movements in infants with DS are often atypical and may lead to additional movement abnormalities as the child develops. These early atypical movements lead to a limited movement repertoire with which to build future movements. In addition, physical activity levels decrease over time (Sayers Menear, 2007). This trend starts early. School age children with DS move less compared to their school age siblings (Whitt-Glover, O'Neill, & Stettler, 2006). Physical activity and motor skill level are related. Lloyd (2008) found a significant negative correlation between the age of walking onset and the level of physical activity in 3-5 year olds with TD and DS. The children who walked earlier were more physically active than the children who walked later (Lloyd, 2008). Both motor skill level and amount of physical activity have implications for overall health as well as inclusion in playground activities and organized sports.

Participation

Infants and children with DS have difficulties with participation level skills in addition to activity level skills. On the participation level of the ICF, children with DS have difficulty appropriately using play to explore their environment (Loveland, 1987) or developing independence in activities of daily living (Fidler, Hepburn, Mankin, & Rogers, 2005). Often, therapeutic interventions focus on the bodily function and structure and activity levels and neglect the participation level. However, participation level skills, by definition, have the largest impact on the ability to fully function in a societal context, which is the ultimate goal of intervention.

Orthotic Use

In the behavioral sciences, interventions at the body function and structure level are often measured at the activity level because this is where the outcomes are observable. Though the outcomes are smaller at the activity level than those expected at the bodily function and structure level, they remain important. Orthoses are a good example of a body function and structure level intervention that affects activity level skills in children with DS. Orthoses that are used at the foot and ankle are external devices that stabilize the subtaler joint, thus maintaining the calcaneus in an upright position. This improves the bony alignment of the foot and ankle, and influences postural and gait characteristics. Selby-Silverstein and colleagues (2001) looked at orthotic use in 3 to 6-year old children with DS. They found that foot orthoses affected body function and structure by decreasing ankle eversion while the children remained in static standing. They also affected activity level skills by decreasing foot progression angle and variability in this angle, changing the initial contact site from flat foot to heel strike, and increasing stance phase speed during gait. Martin (2004) studied orthotic use in 3-8 year old children with DS. She found that the children showed immediate and long term improvements in postural stability when they used a flexible supramalleolar orthosis (SMO). These studies show that orthoses are an effective intervention for children with DS in both the body function and structure level and activity level. However, little is known about the effects of orthoses on pre-walkers with DS.

Treadmill Training

Walking is an important activity level milestone. Like other motor skills, walking in children with DS is not only delayed, but also abnormal (Black, Chang, Kubo, Holt, &

Ulrich, in review; Cioni, Cocilovo, Rossi, Paci, & Valle, 2001; Looper, Wu, Angulo Barroso, Ulrich, & Ulrich, 2006; Parker, Bronks, & Snyder Jr, 1986; Ulrich, Haehl, Buzzi, Kubo, & Holt, 2004). Though infants with DS walk, on average, one year later than infants with TD (Palisano et al., 2001; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001), they are able to step when supported on a treadmill at a much earlier age. About 13 months before they walk, infants with DS respond to the treadmill with alternating steps (Ulrich, Ulrich, Collier, & Cole, 1995). This preference for alternating steps while being supported on a treadmill begins to emerge by 11 months, about the time when infants with DS begin to sit independently (Ulrich, Ulrich, & Collier, 1992).

Ulrich and colleagues (2001) used this stepping behavior to train infants with DS before they were able to walk independently. Children who received treadmill training as a supplement to physical therapy walked independently 101 days earlier than a control group of children with DS who were not receiving the treadmill intervention but continued to receive physical therapy. Though there were no group differences in age at study entry, the average chronological age at walking onset was 23.9 months in the control group and 19.9 months in the intervention group. A subsequent treadmill intervention study showed that a higher intensity individualized training protocol provides even greater outcomes. There was an improvement in gait parameters and a decrease in gait variability when compared to the original treadmill paradigm and to toddlers with DS who did not receive treadmill training (Looper et al., 2006; Wu, Looper, Ulrich, Ulrich, & Angulo-Barroso, 2007). The higher intensity individualized training also has a positive impact on toddlers' ability to cross barriers in their walking path (Wu, Ulrich, Looper, Tiernan, & Angulo-Barroso, 2008).

Purpose

This study is an extension of the previous treadmill training research involving infants with DS. The intent is to modify the lower intensity treadmill training of Ulrich and colleagues (2001) to provide a better outcome. The primary differences between this study and previous studies are that infants enter this study at a later developmental time point and use SMOs as part of the experimental protocol. This research focuses on the use of SMOs as a means to improve functional mobility in children with DS receiving treadmill training. Though the SMOs address joint stabililty, an impairment level problem, combining them with treadmill training, an activity level intervention, will allow me to look across multiple levels of disablement. By intervening with orthoses and treadmill training, we hope to see larger improvements in activity and participation than with treadmill training alone, as well as an improvement in the onset of independent walking.

Method

Participants

Twenty-two children with DS enrolled in this study. Over time, 5 children dropped out of the study (1 had emerging medical problems, 1 did not tolerate the treadmill, and 3 received orthoses prior to the end of the study.) The study was limited to infants with Trisomy 21. Onset of participation occurred when infants could pull to stand at the furniture but not yet cruise. Children were excluded from this study if they had a history of other developmental disabilities, uncorrected visual or hearing impairment,

previous orthotic use, orthotic use other than the ones provided for this study, or were unable to tolerate the orthoses. Infants were randomly assigned into a control (n=11) or experimental group (n=11). Children in the experimental group were measured for SMOs on their first visit and received them before their second visit. The children in the control group were measured for SMOs when they could take three independent steps and received them one to two weeks later. Children participated in this study from the time they could pull to stand until they had one month of independent walking experience. Once the infant could take three independent steps over ground, the treadmill training stopped. The Institutional Review Board at the University of Michigan approved this study.

Data collection

At the first visit to the family's home, parents reviewed and signed an informed consent document. Next, the researcher taught parents how to hold the infants on the treadmill in a safe manner and how to operate the treadmill (see figure 2.1). Once parents were comfortable with the procedures, the researcher held the infant on the treadmill to establish a baseline measure of treadmill stepping performance. This included three one-minute blocks of treadmill training that were videotaped.

At each subsequent monthly visit, three minutes of treadmill training were videotaped. The video camera was positioned perpendicular to the treadmill. It was adjusted to clearly view the child's legs and trunk. The child was supported on the running treadmill for one minute then rested briefly. This was repeated for three one-minute trials. During the testing, the child was supported by the researcher who held the child's trunk under his arms. During each monthly visit, the researcher took

anthropometric measures of each child. These measures included right shank length (from the lateral malleoli to the lateral joint line of the knee) and circumference at the midpoint of the shank, right thigh length (from the lateral knee joint line to the greater trochanter) and circumference at the midpoint of the thigh, body weight, and body length. For leg measurements, infants were seated on their parent's lap with knees flexed to 90 degrees. Infants laid in supine for the body length measurement. The researcher also updated progress in each child's motor skill developmental using the GMFM and the PEDI. This was done either by the researcher observing the specific skill or by the researcher asking the parent if the child was able to perform the skill on the GMFM. On the PEDI, the parent reported whether or not the child was capable of doing the test item.

Treadmill Training

Infants in both groups received treadmill training that began when the infants entered the study and continued until they took three independent steps overground. Each family was provided a small, motorized treadmill (Carlin Inc, Michigan) on the first visit. The parents were instructed to turn the treadmill on to a speed of 0.2 m/s and hold their child under the arms on the treadmill for 8 minutes a day for 5 days a week, allowing the child to support as much of his weight as possible. Parents were also instructed to have their infant were shoes (for the control group) or shoes and orthoses (for the experimental group) during the training. The 8-minutes of training did not have to be continuous. Parents recorded the training provided each day in a logbook provided by the researcher. In addition, the researcher recoded the odometer reading on the treadmill monthly to validate use.

Treadmill Stepping Data

Three one-minute treadmill responses were videotaped one time per month during the training period. Trained coders viewed all trials to code the number of steps produced and step type. Step types included alternating, single, double and parallel steps. An alternating right step occurred when the right foot stepped and was followed by a left step that crossed the midline of the right foot. If the left foot did not take a step or did not cross the midline of the right foot, it was coded as a right single step. If the child moved both feet forward at the same time, under his own power, it was coded as a parallel step. (See Ulrich & Ulrich, 1995 for further description of step coding.) There were a total of 5 coders. They had 96% agreement on the number of alternating steps on a random sample of trials that they all coded.

Developmental Test Data

Developmental progress was measured one time per month in each child's home while they were receiving treadmill training and in the lab when they had one month of independent walking experience. The GMFM (Russell, Rosenbaum, Avery, & Lane, 2002) was used to measure gross motor skill acquisition. The PEDI (Haley, Coster, Ludlow, Haltiwanger, & Andrellos, 1992) was used to measure the ability to use movement to participate in a societal context. Each test was updated monthly based on observation and parent report.

The Gross Motor Function Measure (GMFM) was used to test activity level skill development throughout the course of the study. The GMFM measures gross motor skill level in 5 subsections: (1) lying & rolling, (2) sitting, (3) crawling & kneeling, (4) standing, and (5) walking, running & jumping. Each item within the subsections is

scored on a scale from 0 (does not initiate) to 4 (completes). The test was developed for children with cerebral palsy but has since been validated for children with DS (Russell et al., 1998). It is widely used in the literature (Gemus et al., 2001; Martin, 2004; Palisano et al., 2001).

The Pediatric Evaluation of Disability Inventory (PEDI) was used to measures the children's ability to participate in tasks that are important for functioning in society. It has 2 parts, functional skills and caregiver assistance. Each part has three subsections, self-care, mobility, and social function. The PEDI is administered as a structured parent interview. On the functional skills section, the parent determines if the child is capable (1) or unable (0) to perform each test item. On the caregiver assistance portion of the test, the parent determines how much assistance the child needs to perform each test item on a scale of 0 (total assistance) to 5 (independent). The PEDI has been validated for children with developmental disabilities and is a good measure of participation level skills. It has been used in studies of children with DS (Dolva, Coster, & Lilja, 2004; Dolva, Lilja, & Hemmingsson, 2007; Volman, Visser, & Lensvelt-Mulders, 2007). In children with DS, the PEDI, as a measure of functional status, is more related to motor ability than mental ability (Volman et al., 2007). This makes the PEDI a good test of the influence of motor skills on functional performance.

Results

Study Sample Characteristics

The participants' physical characteristics at study onset are reported in table 2.1. The only a priori group difference was birth length, but by study onset this was no longer significantly different. There were no group differences at study onset in age,

anthropometric measures, or the amount of time the infants received physical therapy per week. There were also no differences between the groups in parental age or education levels, birth order, or number of siblings.

In terms of developmental test scores and the number of alternating steps, there were group differences at study entry (see table 2.2). The experimental group had a higher overall GMFM score that was made up of higher scores in GMFM subscales C (Crawling and Kneeling), D (Standing), and E (Walking, Running and Jumping). There was no difference in the total PEDI score, however the experimental group did score higher in the Social subsection of the PEDI at study entry. There was no difference between the groups in the total PEDI Assistance scale or in any of the subscales of the assistance section of the PEDI. The experimental group took more alternating steps on the treadmill at study entry. The initial differences between the groups were controlled for in the statistical analysis by including them as terms in the statistical models.

Though the stated treadmill training protocol was 8 minutes per day 5 days a week, the participants in both groups performed the training an average of 6±2 minutes per day 5 days a week. In addition, the experimental group wore the SMO 6.25±4 hours per day.

Walking Onset

The average age of walking onset was 846±249 days (28.2±8.3 months) for the control group and 848±176 days (28.3±5.9 months) for the experimental group. The average time in study was 268 ±88 days (8.9±2.9 months) for the control group and 206±109 days (6.9±3.6 months) for the experimental group. T-tests were performed to see if there were differences between the groups in age at walking onset and time in

study. There were no significant differences for age at walking onset (p=0.98) or time in study (p=0.23). There was a moderate effect (Cohen's D=0.63) in favor of the experimental group for time in the study (Cohen, 1988) indicating that a larger sample size may produce significant results.

Treadmill Stepping Patterns

The average number of steps at study entry was 20±9 for the control group and 35±15 for the experimental group. At walking onset, these numbers had increased to 38±13 and 50±14, respectively. There was a significant group difference at study entry which was controlled for in the statistical analysis. A mixed linear model was used to test the difference in developmental trends in number of alternating steps between the groups over the course of the training. The model included a term for the number of alternating steps at study entry, the linear effects of time, the quadratic effects of time, group effect, and both possible group by time interactions. The model also allowed time to vary randomly by subject because the infants were in the study for varying amounts of time. The predicted values of alternating steps over time, based on the model, are shown in figure 2.2. All children in the study increased the number of alternating steps they took on the treadmill over time (F(31,1)=15.28 p< 0.01). There was also a significant quadratic effect of time (F(11,1)=5.83 p=0.03). There were no significant group differences in the number of alternating steps (F(108,1)=0.69 p=0.41), nor were there significant group by time interactions (F(31,1)=2.11 p=0.16, linear and F(11,1)=2.26 p=0.16, quadratic.

Developmental Testing During Treadmill Training

GMFM

Mixed linear models were used to test the differences between groups and over time in the overall GMFM scores as well as subscale B, C, D, and E, scores. The models included a term for the linear effects of time, the quadratic effects of time, group effect, and both possible group by time (linear or quadratic) interactions. All models except the one for subscale B, also included a term for initial score to account for differences between the groups at study onset. The models also allowed time to vary randomly by subject. Predicted GMFM scores, based on the model, as well as the raw data are shown in figures 2.3 – 2.5.

All children in the study improved their overall GMFM scores over time ($F(23,1)=106.96$ $p< 0.01$). There was also a significant quadratic effect of time in the total GMFM score ($F(12,1)=38.83$ $p< 0.001$) indicating that the developmental trend to gross motor skill acquisition has a significant non-linear component to it. There was no significant group difference on the overall GMFM score ($F(122,1)=0.24$ $p=0.623$). There was a significant linear time by group interaction for the overall GMFM score ($F(22,1)=4.92$ $p=0.04$) but no quadratic time by group interaction ($F(12,1)=2.91$ $p=0.11$).

All children in the study improved their subscale B (Sitting) scores over time ($F(123,1)=52.51$ $p<0.01$). There was also a significant quadratic effect of time ($F(103,1)=22.13$ $p<0.001$). There was not a significant group difference on subscale B ($F(119,1)=1.90$ $p=0.17$). There were no significant linear or quadratic time by group interactions ($F(123,1)=0.34$ $p=0.56$ and $F(103,1)=0.20$ $p=0.66$, respectively).

All children in the study improved their subscale C (Crawling and Kneeling) scores over time ($F(10,1)=43.42$ $p<0.01$). There was also a significant quadratic effect of time ($F(7,1)=21.82$ $p<0.01$). There was a trend toward a group difference on subscale C ($F(115,1)=3.29$ $p=0.07$). There was also significant linear time by group interaction (($F(10,1)=6.43$ $p=0.03$) and quadratic time by group interaction ($F(7,1)=5.48$ $p=0.05$). These significant interactions indicate that the two groups followed a different developmental *p*rogression from study entry through independent walking.

All of the children improved their subscale D (Standing) scores over time ($F(82,1)=32.41$ $p<0.01$). There was also a significant quadratic effect of time for subscale D ($F(107,1)=5.16$ $p=0.03$). There was no significant group difference on the subscales D scores ($F(110,1)=0.61$ $p=0.44$). There were significant linear and quadratic time by group interactions ($F(79,1)=8.30$ $p=0.01$ and $F(104,1)=6.54$ $p=0.01$, respectively).

All of the children in the study improved there subscale E (Walking, Running and Jumping) scores over time ($F(24,1)=53.12$ $p<0.01$). There was also a significant quadratic effect of subscale E ($F(10,1)=13.32$ $p=0.01$). There was no significant group difference on subscale E ($F(121,1)=0.05$ $p=0.82$). There were no significant linear or quadratic time by group interactions ($F(24,1)=0.77$ $p=0.39$ and $F(10,1)=0.22$ $p=0.65$, respectively).

PEDI Functional Skills Section

Mixed linear models were used to test the difference in developmental trends between groups and over time in the PEDI functional skill section including total scores and the Self –Care, Mobility, and Social subsection scores. Each model included the

linear effects of time, the quadratic effects of time, group effect, and both possible group by time (linear or quadratic) interactions. The Social subsection model also included a term for the initial score of the Social subsection because there was a group difference in this parameter at study entry. The models also allowed time to vary randomly by subject because the infants were in the study for varying amounts of time. The values of the PEDI functional skills subsection scores over time are found in figures 2.6 – 2.8.

All children in the study improved their PEDI functional skills section total scores linearly over time ($F(8,1)=15.12$ $p= 0.01$). There was no significant quadratic effect of time ($F4,1)=0.63$ $p=0.47$). There was no significant group effect on overall PEDI functional skills score ($F(16,1)=0.21$ $p=0.65$). There were no significant linear or quadratic time by group interactions ($F(8,1)=1.10$ $p=0.33$ and $F(4,1)=0.16$ $p=0.71$, respectively).

There was a linear change over time on the Self Care subsection of the functional scale of the PEDI ($F(29,1)=3.81$ $p=0.06$). There was no quadratic change over time ($F(24,1)=0.96$ $p=0.34$). There were no significant group differences for the PEDI functional skills section Self Care subsection scores ($F(94,1)=0.81$ $p=0.37$). There were not significant linear or quadratic time by group interactions ($F(29,1)=0.02$ $p=0.90$ and $F(24,1)=0.05$ $p=0.83$, respectively).

All of the children showed a linear improvement over time on the Mobility subsection of the PEDI functional skill section ($F(13,1)=6.23$ $p=0.03$). There was not quadratic effect of time ($F(12,1)=0.190$ $p=0.67$). There was no significant group effect ($F(13,1)=0.12$ $p=0.74$). There were no time by group interactions ($F(13,1)=0.11$ $p=0.75$, linear and ($F(12,1)=0.02$ $p=0.89$, quadratic).

The children showed a linear improvement over time on the Social subsection of the PEDI functional skill section ($F(25,1)=6.73$ $p=0.02$. There was no quadratic time effect ($F(7,1)=0.13$ $p=0.73$) on the Social subsection scores. There were no significant group differences for the Social subsection scores ($F(89,1)=0.82$ $p=0.37$). In addition, there were no significant group by time interactions ($F(26,1)=1.34$ $p=0.26$, linear and $F(7,1)=0.30$ $p=0.60$, quadratically).

PEDI Assistance Section

Mixed linear models were used to test the difference in developmental trends in the total scores of the assistance section of the PEDI as well as its subsections: Self Care, Mobility, Social. Each model included the linear effects of time, the quadratic effects of time, group effect, and both possible group by time (linear or quadratic) interactions. The models also allowed time to vary randomly by subject. The values of the PEDI assistance scale subsection scores over time are in figures 2.9 – 2.11.

The children in the study improved their PEDI assistance section total scores scores linearly over time ($F(30,1)=17.42$ $p= 0.01$). There was no significant quadratic change over time ($F(18,1)=2.22$ $p=0.15$). The experimental group scored higher on the PEDI assistance section total scores than the control group ($F(112,1)=7.65$ $p=0.01$). There were no linear or quadratic time by group interactions ($F(30,1)=1.21$ $p=0.28$, linear and $F(18,1)=0.79$ $p=.39$, quadratic).

The children in the study improved their PEDI assistance section Self Care subsection scores linearly over time ($F(44,1)=4.56$ $p= 0.04$). There was no significant quadratic change over time ($F(19,1)=0.05$ $p=0.83$). The experimental group scored higher on the PEDI assistance section Self Care subsection than the control group

($F(110,1)=12.58$ $p<0.01$). There were significant linear or quadratic time by group interactions ($F(44,1)=8.82$ $p<0.01$, linear and $F(19,1)=10.41$ $p<0.01$, quadratic).

The children in the study improved their PEDI assistance section Mobility subsection scores linearly over time ($F(12,1)=24.85$ $p< 0.01$). There was a trend toward a quadratic change over time ($F(10,1)=4.09$ $p=0.07$). There was no significant group difference ($F(12,1)=0.91$ $p=0.36$). There were no significant linear or quadratic time by group interactions ($F(12,1)=0.01$ $p<0.01$, linear and $F(10,1)=0.12$ $p=0.74$, quadratic).

The children in the study improved their PEDI assistance section Social subsection scores linearly over time ($F(123,1)=14.60$ $p<0.01$). There was no significant quadratic change over time ($F(123,1)=2.64$ $p=0.11$). There was no significant group difference ($F(114,1)=1.65$ $p=0.20$). There were no significant linear or quadratic time by group interactions ($F(123,1)=1.33$ $p=0.25$, linear and $F(123,1)=1.54$ $p=0.22$, quadratic).

Developmental Testing in New Walkers

GMFM

At one month of walking experience, the mean scores for the overall GMFM and subscales C (Crawling and Kneeling), D (Standing), and E (Walking, Running and Jumping) were 195.67±8.12, 35.67±0.82, 28.67±4.5, and 20.33±3.72, respectively, for the control group and 183.78±7.22, 35.89±1.17, 21.00±5.12, and 15.89±2.80, respectively, for the experimental group (see Figure 2.12). T-test were performed to determine if the scores differed by group.

There was no overall group difference for the GMFM subscale C score ($p=0.69$). The control group scored significantly higher on the GMFM total score, subscale D, and

subscale E (p=0.01, p=0.01, and p=0.02, respectively). In addition, there was a large effect in favor of the control group in GMFM total score as well as subscale D and E (Cohen's D = 1.55, 1.59, and 1.36, respectively). In other words, the average infant in the control group had a GMFM total score that was 1.55 standard deviations higher than the average infant in the experimental group. There was no difference in subscale C (Cohen's D = 0.22).

PEDI Functional Skills Section

At one month of walking experience, the mean scores for the overall PEDI functional skills section and the Self Care, Mobility, and Social subsections were 77.67±10.50, 23.00±4.29, 31.33±5.79, and 23.33±1.63, respectively, for the control group and 79.67±11.08, 24.11±5.04, 30.44±5.34, and 25.22±3.69, respectively for the experimental group (see Figure 2.13). T-tests were performed to see if the scores differed by group.

There were no significant group differences for the PEDI functional scale total score or the Self Care, Mobility, or Social subsections (p=0.73, p=0.67, p=0.76, and p=0.29, respectively. There was a moderate effect in favor of the experimental group for the Social subsection (Cohen's D= 0.67).

PEDI Assistance Section

At the time when the infants has one month of walking experience, the mean scores for the overall PEDI assistance section and the Self Care, Mobility, and Social subsections was 38.67±5.43, 7.17±2.04, 23.83±2.99, and 7.67±1.37, respectively, for the control group and 35.22±5.89, 6.67±2.50, 20.78±3.31, and 7.89±1.62, respectively for the

experimental group (see Figure 2.13). T-tests were performed to see if the scores differed by group. There was a trend in favor of the control group on the Mobility subsection (p=0.09). There were no group differences on the total PEDI assistance scale, the Self Care subsection, or the Social subsection (p=0.29, p=0.69, and p=0.79, respectively). There was a moderate effect size in favor of the control group for the overall PEDI assistance section score and a large effect size in favor of the control group for the Mobility subsection score (Cohen's D= 0.59 and 0.97, respectively).

Discussion

The purpose of this study was to determine whether the addition of SMOs to a treadmill training protocol for children with DS would lead to improved developmental outcomes, especially on the activity and participation sections of the ICF. These outcomes included raw developmental test scores and time in study until each infant could take 3 steps independently. The hypothesis was that the addition of SMOs would lead to improved developmental test scores and decreased time in study. The results, however, were not so straightforward.

"Traditional" treadmill training, as described by Ulrich and colleagues in 2001 and 2008, is an extremely effective intervention for most infants with DS. It leads to a large decrease in time to independent walking onset and leads to improved gait at walking onset when compared to children who did not receive treadmill training (Ulrich et al., 2001; Wu, et al., 2007). In the current study, infants with DS who received traditional treadmill training were compared to a group of children with DS who received traditional treadmill training as well as early orthotic use. Though there was not a

significant difference between the groups, there was a moderate treatment effect in favor of the group that received SMOs. This suggests that the SMOs are positively affecting the rate of walking development. The fact that the treadmill training is consistently successful (Ulrich et al., 2008; Ulrich et al., 2001) may make small improvements hard to detect statistically without having a large sample size. This is due to the high level of variability in most developmental measures in the DS population.

Though the major difference between this study and previous treadmill studies on infants with DS is the addition of orthoses, another large difference was the time that treadmill training was initiated. Previous studies began the treadmill training protocol when children could sit independently (Ulrich et al., 2001) or take 6-10 supported steps on the treadmill (Ulrich et al., 2008). Because the current study focuses on SMO use, the intervention did not begin until the children were able to pull to stand and bear weight on their feet. This corresponds to Ulrich's study (1992) that found children with DS began to prefer alternating stepping patterns on the treadmill when they could pull to stand and make forward progress in prone. On average, the children in this study pulled to stand at 20.5 months or about 2 weeks after the children who received treadmill training in the original study began to walk. Though the cohorts from these two studies were different and may have varied in factors affecting gross motor development, this large difference in developmental level at the age of 20 months points to the importance of early implementation of the treadmill intervention. Perhaps a better experimental combination of treadmill training and orthoses would include treadmill training, beginning at 10 months of age, and SMOs when the children can pull to stand independently. This would

allow the infants to derive the maximal benefits from the treadmill training while still introducing the orthoses at a developmentally appropriate point.

While it appears that earlier practice is better than later practice, treadmill training is still an effective way to practice supported alternating stepping in older infants with DS. The average number of alternating steps per minute on the treadmill increased over the course of the intervention for all children in the study. The increase was nonlinear showing a rapid rise at the beginning of the intervention and a leveling out toward walking onset. Ulrich and colleagues (2008) found a similar trend. This leveling out seems to coincide with the child's ability to walk well behind a push toy.

According to the ICF, gross motor skills are activity level skills (WHO, 2001). In this study, activity level skills were measured using the time to walking onset as well as the GMFM scores. As mentioned above, the orthoses appear to have a moderate effect on the time to attainment of walking. The effect of orthoses on GMFM scores is slightly more complex. As expected, all children in the study showed an improvement in their gross motor skills during the course of the intervention. In addition, there was no group difference over the course of the intervention in the overall GMFM score. This was expected because all the children entered the study at the same gross motor level, pulls to stand, and ended the intervention at the same gross motor level, 3 independent steps. However, the control group showed larger improvements in the Crawling and Kneeling section of the GMFM than the experimental group. In addition, the predicted developmental trends for the Crawling and Kneeling section and Standing section differ by group, leading to significant group by time interactions. For both of these sections, the control group displays a rapid increase in scores followed by a leveling out while the

experimental group shows a much more linear improvement over time. The Crawling and Kneeling section contains items such a crawling up and down 4 steps, kneeling, and half kneeling. Orthoses that limit the mobility around the ankle may make these activities difficult to do leading to a slower rate of progression for the children with orthoses on this subscale. The Standing section contains items that test balance such as standing independently for 3 seconds or 20 seconds, and lifting one leg for 3 seconds. Though the orthoses provided external stability to the foot and ankle, which helps balance in older children with DS (Martin, 2004), these children have not yet learned how to balance. The use of orthoses while learning these skills may have limited the available solutions for solving the problem by limiting the amount of movement at the foot and ankle. In turn, the initial rate of increase on the Standing section scores was not as large in the experimental group as it was in the control group.

At 1 month of walking experience, differences between the groups in the GMFM scores persisted. The control group scored significantly higher on the overall GMFM as well as on the Crawling and Kneeling, Standing, and Walking Jumping and Running sections. This suggests that the children who learned to walk without the orthoses had an advantage in terms of 4-point mobility, balance, and upright mobility. During the development of a skill, infants experiment with and explore multiple solutions for solving movement tasks (Thelen & Corbetta, 1994). Through this process, they learn how to perform a skill and how to adapt that skill to new or differing circumstances. Perhaps the SMOs externally prescribe limits in ankle and foot alignment and range of motion during this important developmental period that detract from the variability of practice and thus the adaptability of the learned skills. The orthoses lead to an improvement in the targeted

activity level skills (because the intervention is specifically designed to practice walking) but to an overall detriment in the development of other gross motor skills.

In the ICF, participation is an activity within a societal context (WHO, 2001). In this study, the PEDI tested the effect of treadmill training and orthotic use on participation. On the functional skill section of the PEDI, all children improved over time but the use of orthoses did not seem to affect the rate of change in the scores. On the assistance section of the PEDI, all children increased their level of independence over time. The self-care subsection asks parents to rate how independent their child is at eating, grooming, bathing, dressing, and toileting. The use of orthoses affected the rate of change on this subsection. The children who wore orthoses scored higher overall than then children who did not wear orthoses. The children without orthoses made rapid gains in this subsection at the beginning of the intervention but then began to level off while the children with orthoses started with a somewhat flat trajectory and began to increase at the point when the other children leveled off.

However, any differences between the groups on the PEDI disappeared by the time the children had one month of independent walking experience. On the functional skills section of the PEDI, the use of orthoses had a moderate effect on the social subsection. On the assistance section, the children who did not receive orthoses during the intervention showed more independence on the mobility subsection. The lack of orthoses also had a moderate effect on the total score of the assistance section of the PEDI. As with the GMFM outcomes, the imposed limitations of the orthoses during a critical period in development could account for the decreased level of independence in the children who learned to walk wearing orthoses as opposed to those who did not

receive orthoses until after they were able to walk. In addition, the lack of variability and adaptability in the development and practice of the gross motor skills of children who wore orthoses early may have contributed to their increased need for assistance to complete motor tasks. The SMOs appear to have negative affects on the development of both activity and participation level skills.

Conclusions

The use of SMOs does appear to have a moderate benefit over and above that of treadmill training in terms of time to walking onset in children with DS. However, the SMOs also appear to have an overall detrimental effect on activity level and participation level skills in infants and new walkers who have learned to walk while wearing them. Based on this information, health care professionals may want to postpone the use of SMOs in children with DS until the children have learned to walk independently. In addition, this study suggests that in order to receive the maximum benefit from the treadmill intervention, therapists must begin its implementation before infants display walking readiness; waiting until they can pull to stand is too late.

There are a few limitations that should be kept in mind when considering the results and conclusions of this study. This study compared a group of children with DS who received treadmill training to another group of children with DS who received treadmill training and orthoses. We cannot predict the differences between a group that received orthoses and a group that did not receive treadmill intervention. Future studies should contain an orthoses only group. We visited parents in this study 1 time every month. In previous studies, the researchers visited the families' homes every 2 weeks. This increased contact with the families may have resulted in greater protocol

compliance. Future studies may benefit from increased family contact. This study did not consider individual differences in terms of orthotic type. Less supportive orthoses may be more beneficial to some children with DS. Future studies are needed to determine how to choose orthotic type and whether other forms of orthoses may be more beneficial. The sample size in this study was smaller than originally planned due to the loss of 5 infants mostly due to recommendations to infants in the control group to begin SMO use before the end of the study, requiring them to drop out of the study. The small sample size negatively affected statistical power. Further studies should include a larger sample size.

References

Black, D., Chang, C-L., Kubo, M., Holt, K., & Ulrich, B. D. (in review) Developmental Trajectory of Dynamic Resource Utilization During Walking: Toddlers with and Without Down Syndrome

CDC. (2006). Improved national prevalence estimates for 18 selected major birth defects - United States, 1999-2001. *Morbidity Mortality Weekly Report, 54*(51/52), 1301-1305.

Chen, Y. J., & Fang, P. C. (2005). Sensory evoked potentials in infants with Down syndrome. *Acta Paediatrica, 94*(11), 1615-1618.

Cioni, M., Cocilovo, A., Rossi, F., Paci, D., & Valle, M. S. (2001). Analysis of ankle kinetics during walking in individuals with Down syndrome. *Americal Journal on Mental Retardation, 106*(5), 470-478.

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (Second Edition ed.). Hillsdale: Lawrence Erlbaun Associates, pp. 20-26.

Dolva, A.-S., Coster, W., & Lilja, M. (2004). Functional performance in children with Down syndrome. *American Journal of Occupational Therapy, 58*(6), 621-629.

Dolva, A.-S., Lilja, M., & Hemmingsson, H. (2007). Functional performance characteristics associated with postponing elementary school entry among children with Down syndrome. *American Journal of Occupational Therapy, 61*(4), 414-420.

Fidler, D. J., Hepburn, S. L., Mankin, G., & Rogers, S. J. (2005). Praxis skills in young children with Down syndrome, other developmental disabilities, and typically developing children. *American Journal of Occupational Therapy, 59*(2), 129-138.

Gemus, M., Palisano, R., Russell, D., Rosenbaum, P., Walter, S. D., Galuppi, B., et al. (2001). Using the gross motor function measure to evaluate motor development in children with Down syndrome. *Physical & Occupational Therapy in Pediatrics, 21*(2-3), 69-79.

Haley, S. M. (1986). Postural reactions in infants with Down syndrome. *Physical Therapy, 66*(1), 17-22.

Haley, S. M., Coster, W. J., Ludlow, L. H., Haltiwanger, J. T., & Andrellos, P. J. (1992). *Pediatric Evaluation of Disability Inventory: Development, Standardization, and Administration Manual, Version 1.0*. Boston: Trustees of Boston University , Health and Disability Research Institute.

Henderson, S. E. (1985). Motor skill development. In D. Lane & B. Stratford (Eds.), *Current approaches to Down's syndrome* (pp. 187-218). London: Holt, Rinehart, & Winston.

Hennequin, M., Morin, C., & Feine, J. S. (2000). Pain expression and stimulus localisation in individuals with Down's syndrome. *Lancet, 356*(9245), 1882-1887.

Lloyd, M. C. (2008). *Physical Activity of Preschool Age Children With and Without Down Syndrome*. Unpublished Dissertation. Univeristy of Michigan, Ann Arbor.

Looper, J., Wu, J., Angulo Barroso, R., Ulrich, D., & Ulrich, B. D. (2006). Changes in step variability of new walkers with typical development and with Down syndrome. *Journal of Motor Behavior, 38*(5), 367-372.

Loveland, K. A. (1987). Behavior of young children with Down syndrome before the morror: Exploration. *Child Development, 58*(3), 768-778.

Martin, K. (2004). Effects of supramalleolar orthoses on postural stability in children with Down syndrome. *Developmental Medicine & Child Neurology, 46*(6), 406-411.

Ostensjo, S., Carlberg, E. B., & Vollestad, N. K. (2004). Motor impairments in young children with cerebral palsy: relationship to gross motor function and everyday activities. *Developmental Medicine & Child Neurology, 46*(9), 580.

Palisano, R. J., Walter, S. D., Russell, D. J., Rosenbaum, P. L., Gemus, M., Galuppi, B. E., et al. (2001). Gross motor function of children with Down syndrome: creation of motor growth curves. *Archives of Physical Medicine & Rehabilitation, 82*, 494-500.

Parker, A. W., & Bronks, R. (1980). Gait of Children with Down Syndrome. *Archives of Physical Medicine & Rehabilitation, 61*, 345-351.

Parker, A. W., Bronks, R., & Snyder Jr, C. W. (1986). Walking patterns in Down's syndrome. *Journal of Mental Deficiency Research, 30*, 317-330.

Russell, D., Palisano, R., Walter, S., Rosenbaum, P., Gemus, M., Gowland, C., et al. (1998). Evaluating motor function in children with Down syndrome: validity of the GMFM. *Developmental Medicine & Child Neurology, 40*(10), 693-701.

Russell, D. J., Rosenbaum, P. L., Avery, L. M., & Lane, M. (2002). *Gross Motor Function Measure (GMFM-66 and GMFM-88) User's Manual*. Cambridge: Mac Keith Press.

Sacks, B., & Buckley, S. J. (2003). What do we know about the movement abilities of children with Down syndrome? *Down Syndrome News and Update, 2*(4), 131.

Sayers Menear, K. (2007). Parents' perceptions of health and physical activity needs of children with Down syndrome. *Down Syndrome Research and Practice, 12*(1), 60-68.

Selby-Silverstein, L., Hillstrom, H. J., & Palisano, R. J. (2001). The effect of foot orthoses on standing foot posture and gait of young children with Down syndrome. *Neurorehabilitation, 16*(3), 183-193.

Thelen, E., & Corbetta, D. (1994). Exploration and selection in the early acquisition of skill. *International Review of Neurobiology, 37*, 75-102; discussion 121-103.

Ulrich, B. D., Haehl, V., Buzzi, U. H., Kubo, M., & Holt, K. G. (2004). Modeling dynamic resource utilization in populations with unique constraints: preadolescents with and without Down syndrome. *Human Movement Science, 23*(2), 133-156.

Ulrich, B. D., & Ulrich, D. A. (1995). Spontaneous leg movements of infants with Down syndrome and nondisabled infants. *Child Development, 66*(6), 1844-1855.

Ulrich, B. D., Ulrich, D. A., & Collier, D. H. (1992). Alternating stepping patterns: hidden abilities of 11-month-old infants with Down syndrome. *Developmental Medicine & Child Neurology, 34*(3), 233-239.

Ulrich, B. D., Ulrich, D. A., Collier, D. H., & Cole, E. L. (1995). Developmental shifts in the ability of infants with Down syndrome to produce treadmill steps. *Physical Therapy, 75*(1), 14-23.

Ulrich, D. A., Lloyd, M. C., Tiernan, C. W., Looper, J. E., & Angulo-Barroso, R. M. (2008). Effects of Intensity of Treadmill Training on Developmental Outcomes and Stepping in Infants With Down Syndrome: A Randomized Trial. *Physical Therapy, 88*(1).

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill training of infants with Down syndrome: evidence-based developmental outcomes. *Pediatrics, 108*(5), E84.

Volman, M. J., Visser, J. J., & Lensvelt-Mulders, G. J. (2007). Functional status in 5 to 7-year-old children with Down syndrome in relation to motor ability and performance mental ability. *Disability Rehabilitation, 29*(1), 25-31.

Whitt-Glover, M. C., O'Neill, K. L., & Stettler, N. (2006). Physical activity patterns in children with and without Down syndrome. *Pediatric Rehabilitation, 9*(2), 158-164.

WHO. (2001). *International classification of functioning, disability, and health: ICF*. Geneva: World Health Organization.

Wu, J., Looper, J., Ulrich, B. D., Ulrich, D. A., & Angulo-Barroso, R. M. (2007). Exploring effects of different treadmill interventions on walking onset and gait patterns in infants with Down syndrome. *Developmental Medicine & Child Neurology, 49*(11), 839-945.

Wu, J., Ulrich, D. A., Looper, J., Tiernan, C. W., & Angulo-Barroso, R. M. (2008). Strategy adoption and locomotor adjustment in obstacle clearance of newly walking toddlers with down syndrome after different treadmill interventions. *Experimental Brain Research,* 186(2), 261-272.

Tables

Table 2.1. Characteristics of the Study Sample at Entry

	Control Mean (SD)	Experimental Mean (SD)	*p*-value
Corrected Age at Entry (Days)	578 (188)	642 (121)	0.41
Weight (kg)	9.41 (1.39)	10.26 (0.61)	0.11
Height (cm)	75.81 (7.93)	78.67 (2.74)	0.30
Shank Length (cm)	13.93 (1.50)	14.77 (1.43)	0.26
Shank Circumference (cm)	18.27 (0.95)	17.69 (1.39)	0.35
Thigh Length (cm)	15.00 (1.50)	15.88 (1.58)	0.27
Thigh Circumference (cm)	26.04 (1.95)	25.28 (1.62)	0.39
Physical Therapy (min/wk)	60 (85)	63 (75)	0.94
Birth Weight (kg)	2.79 (0.47)	3.18 (0.41)	0.10
Birth Height (in)	47.09 (2.29)	50.37 (2.49)	0.02*
Number of Siblings	2 (2)	1 (1)	0.23
Birth Order	2 (2)	2 (1)	0.20
Maternal Age (years)	32 (5)	34 (6)	0.43
Paternal Age (years)	34 (5)	33 (7)	0.87
Maternal Education (years)	15 (3)	16 (1)	0.36
Paternal Education (years)	14 (3)	15 (2)	0.65
Income (x1000)	80-100 (40)	60-80 (20)	0.64

Mean (SD)
* $p<0.05$

Table 2.2. Developmental Level at Study Entry

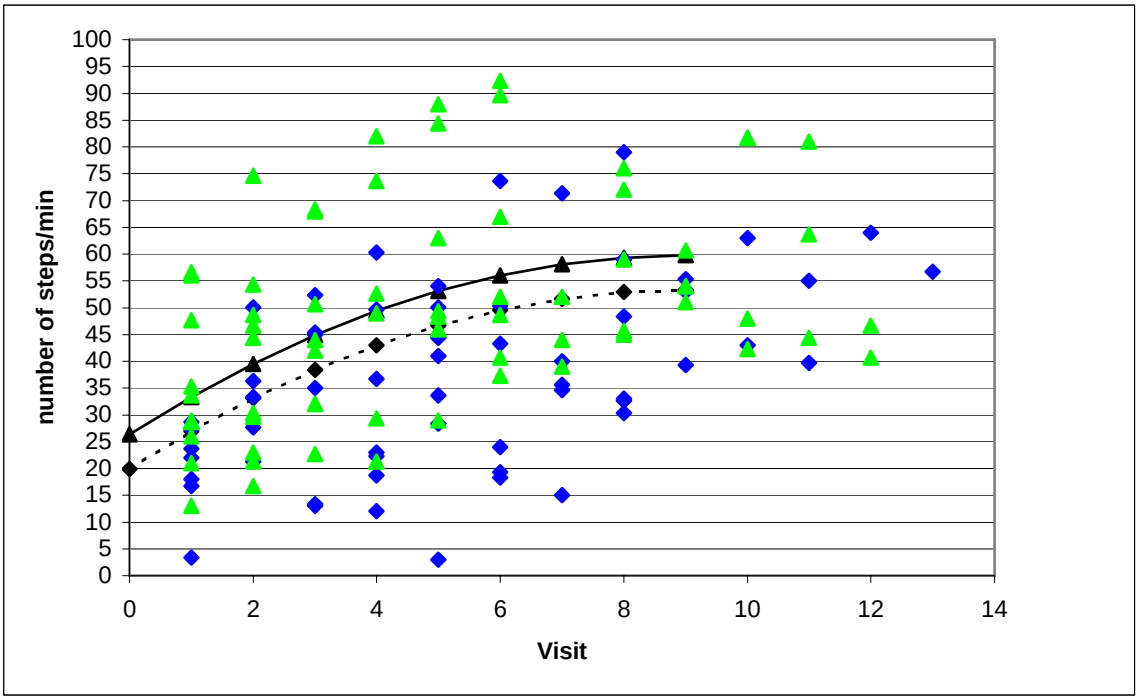
	Control Mean (SD)	Experimental Mean (SD)	*p*-value
GMFM Total	131 (7)	148 (9)	0.01*
GMFM Sitting (B)	52 (4)	55 (4)	0.20
GMFM Crawling and Kneeling (C)	22 (5)	27 (4)	0.05*
GMFM Standing (D)	4 (2)	10(3)	0.01*
GMFM Walking Running and Jumping (E)	1 (1)	6 (4)	0.01*
PEDI Functional Total	44 (11)	53 (13)	0.17
PEDI Functional Self-Care	13 (4)	17 (7)	0.19
PEDI Functional Mobility	17 (7)	18 (6)	0.67
PEDI Functional Social	15 (3)	18 (3)	0.04*
PEDI Assistance Total	16 (6)	19 (5)	0.28
PEDI Assistance Self-Care	3 (1)	4 (2)	0.18
PEDI Assistance Mobility	9 (5)	10 (5)	0.76
PEDI Assistance Social	4 (2)	5 (1)	0.17
Steps per Minute	20(9)	35 (15)	0.03*

Mean (SD)
* $p<0.05$

Figures

Figure 2.1. Infant and parent during treadmill training

Figure 2.2. Number of Alternating Steps Over Time



Predicted Steps/Min. Control Group

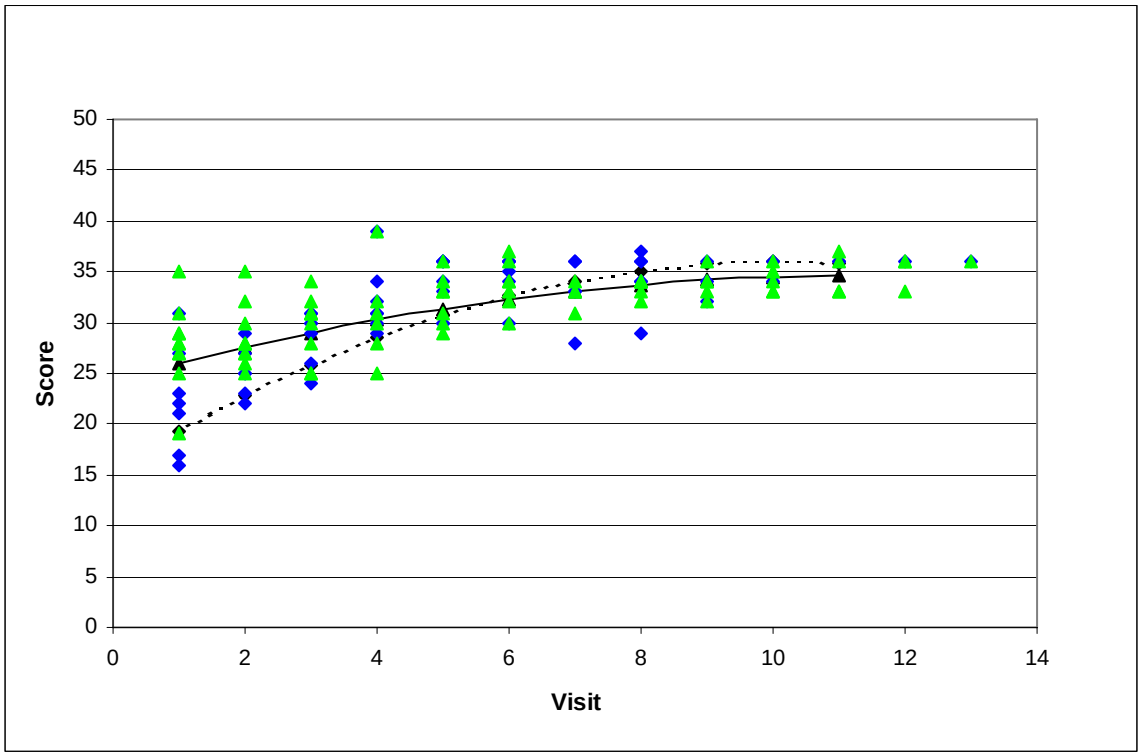
Predicted Steps/Min. Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Non-significant interactions were removed from the model.

Figure 2.3. GMFM Subtest C scores over time



Predicted Subtest C Scores – Control Group

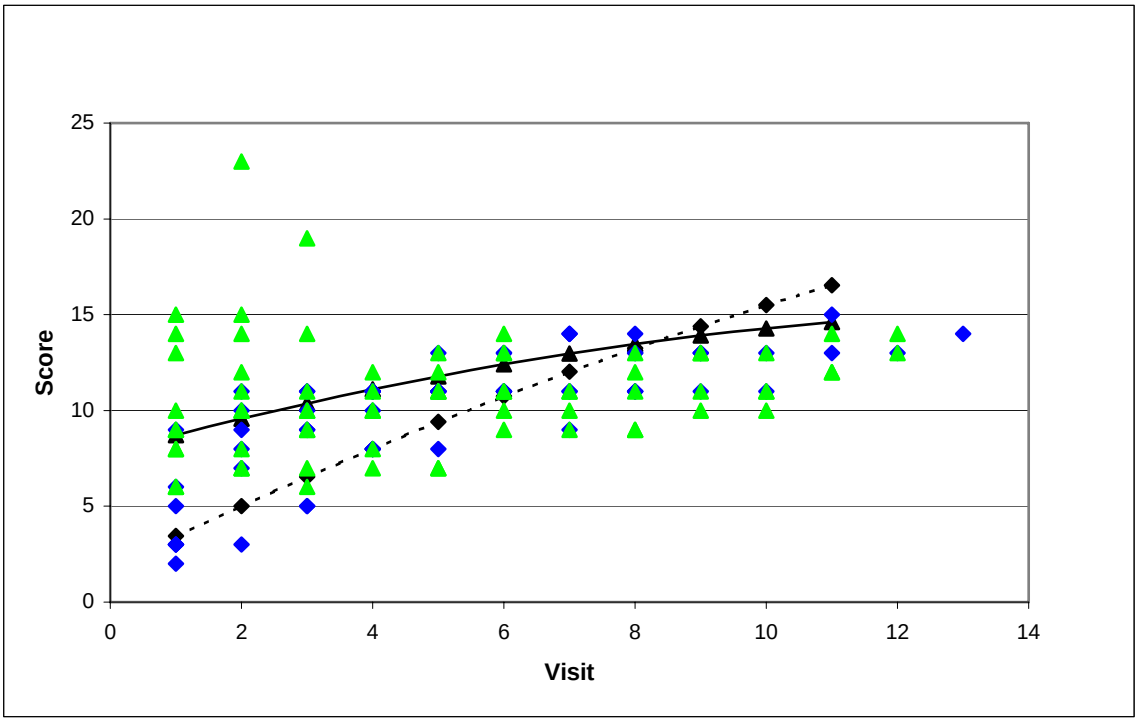
Predicted Subtest C Scores – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Non-significant interactions were removed from the model.

Figure 2.4. GMFM Subtest D scores over time



Predicted Subtest D Scores – Control Group

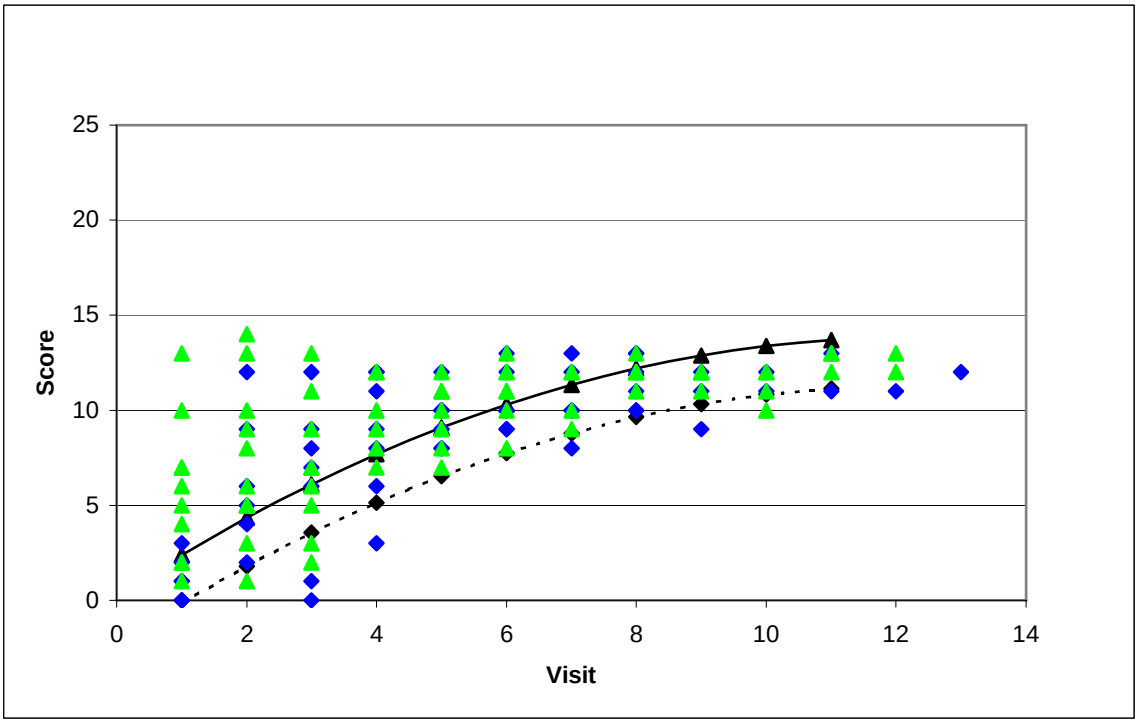
Predicted Subtest D Scores – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Non-significant interactions were removed from the model.

Figure 2.5. GMFM Subtest E scores over time



Predicted Subtest E Scores – Control Group

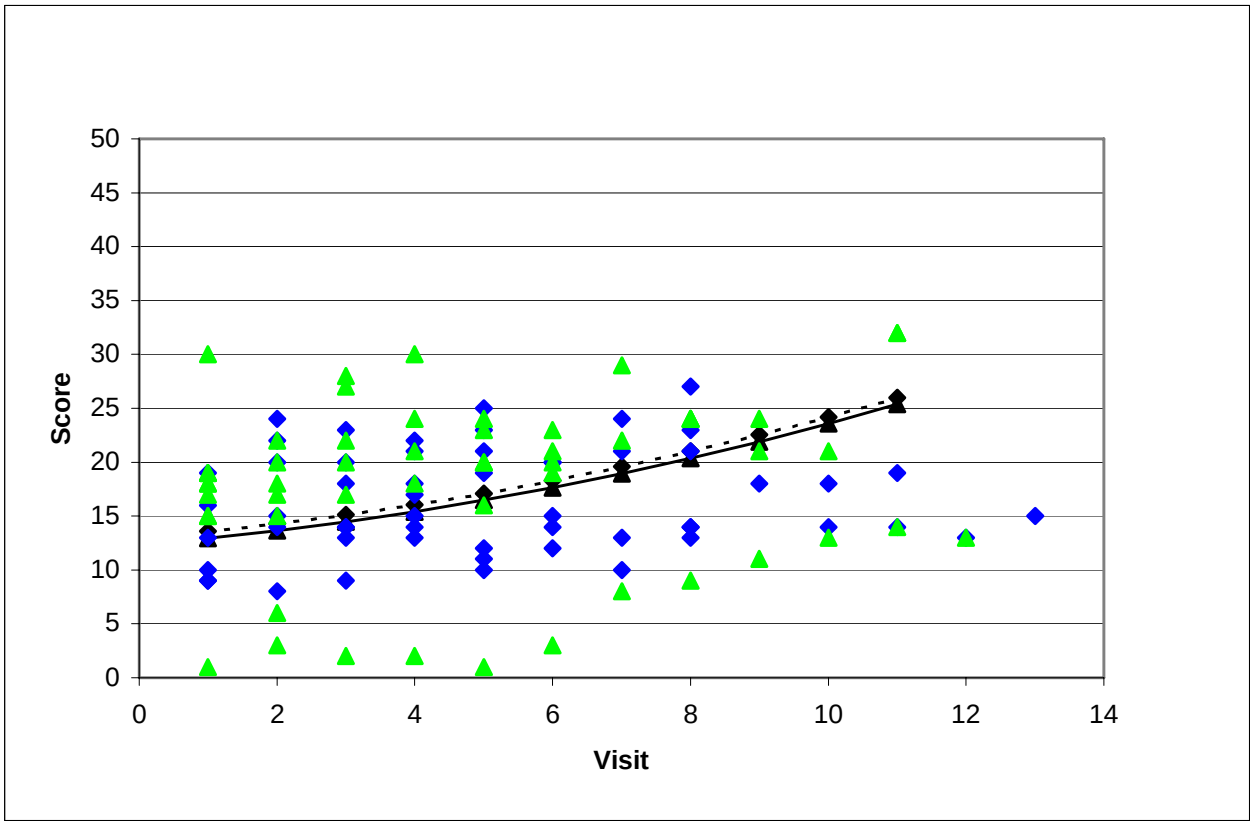
Predicted Subtest E Scores – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

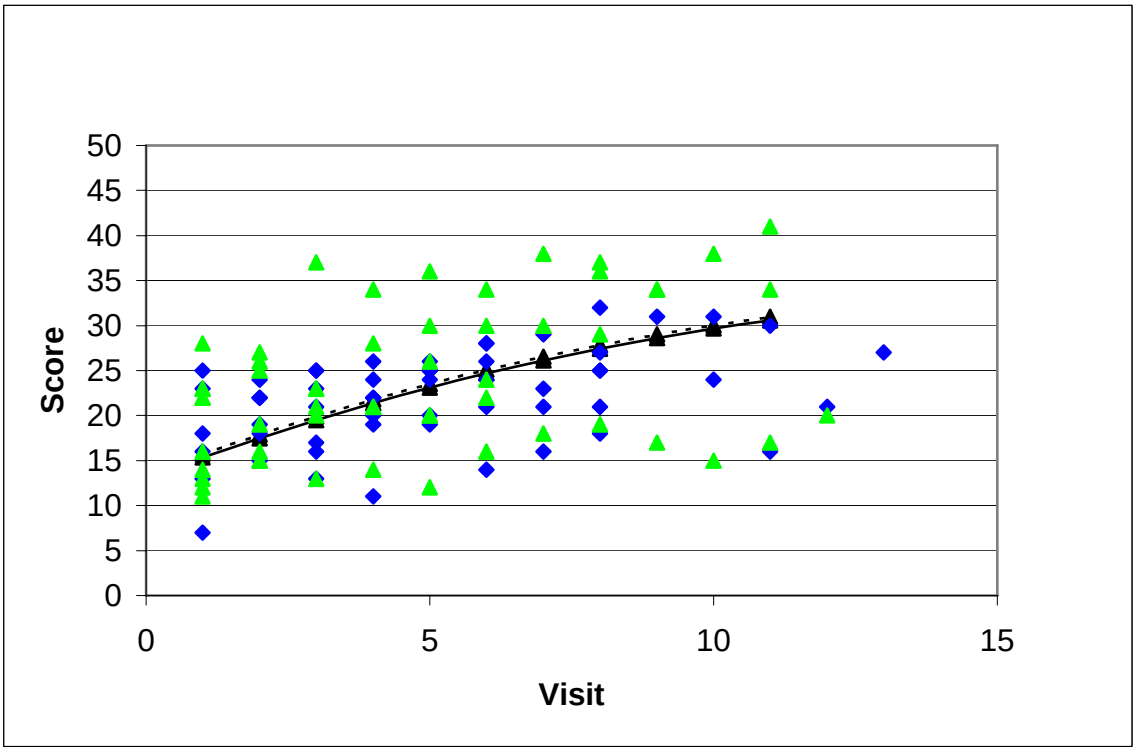
Non-significant interactions were removed from the model.

Figure 2.6. PEDI functional skills subsection SC scores



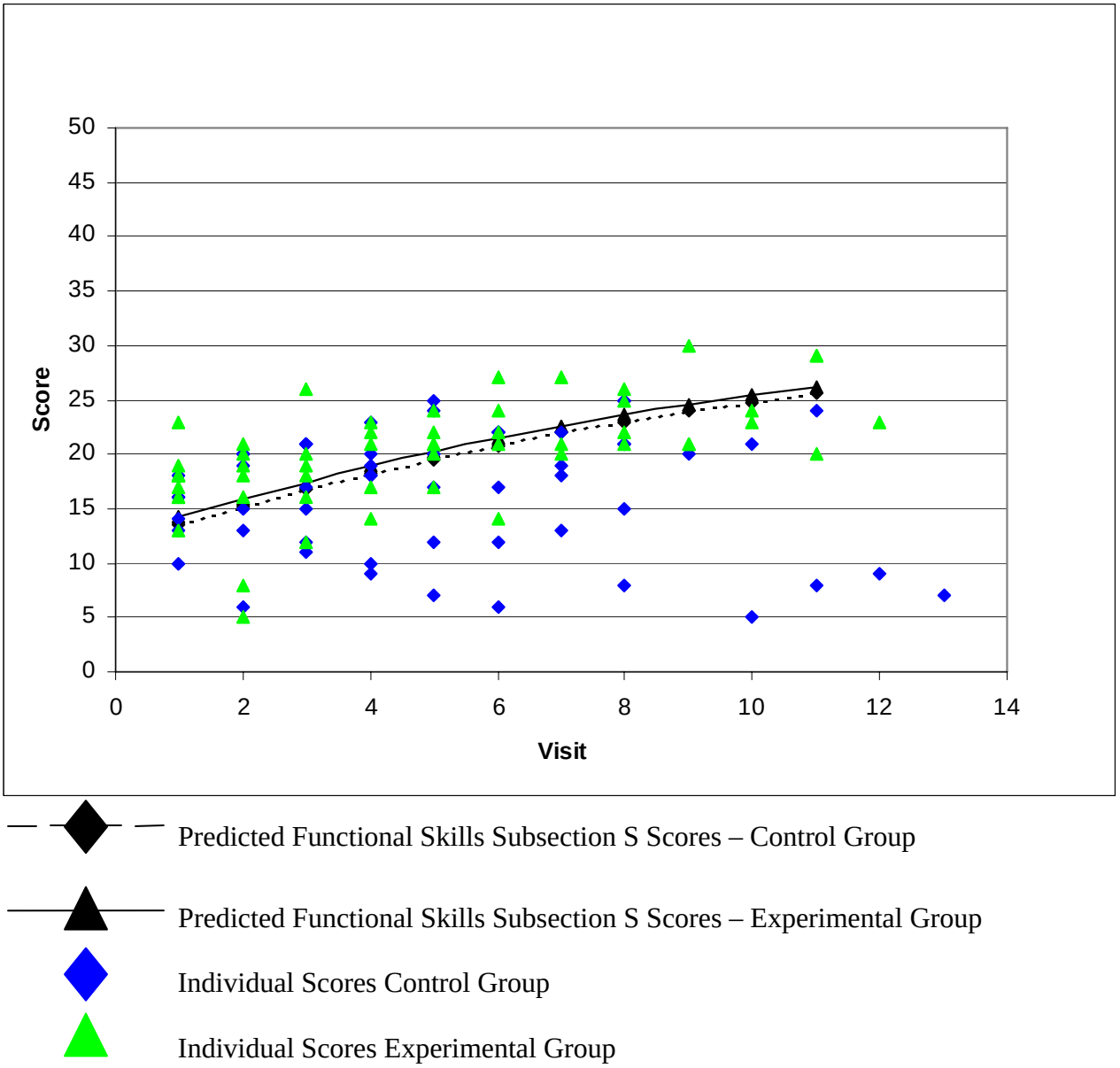
Non-significant interactions were removed from the model.

Figure 2.7. PEDI functional skills subsection M scores



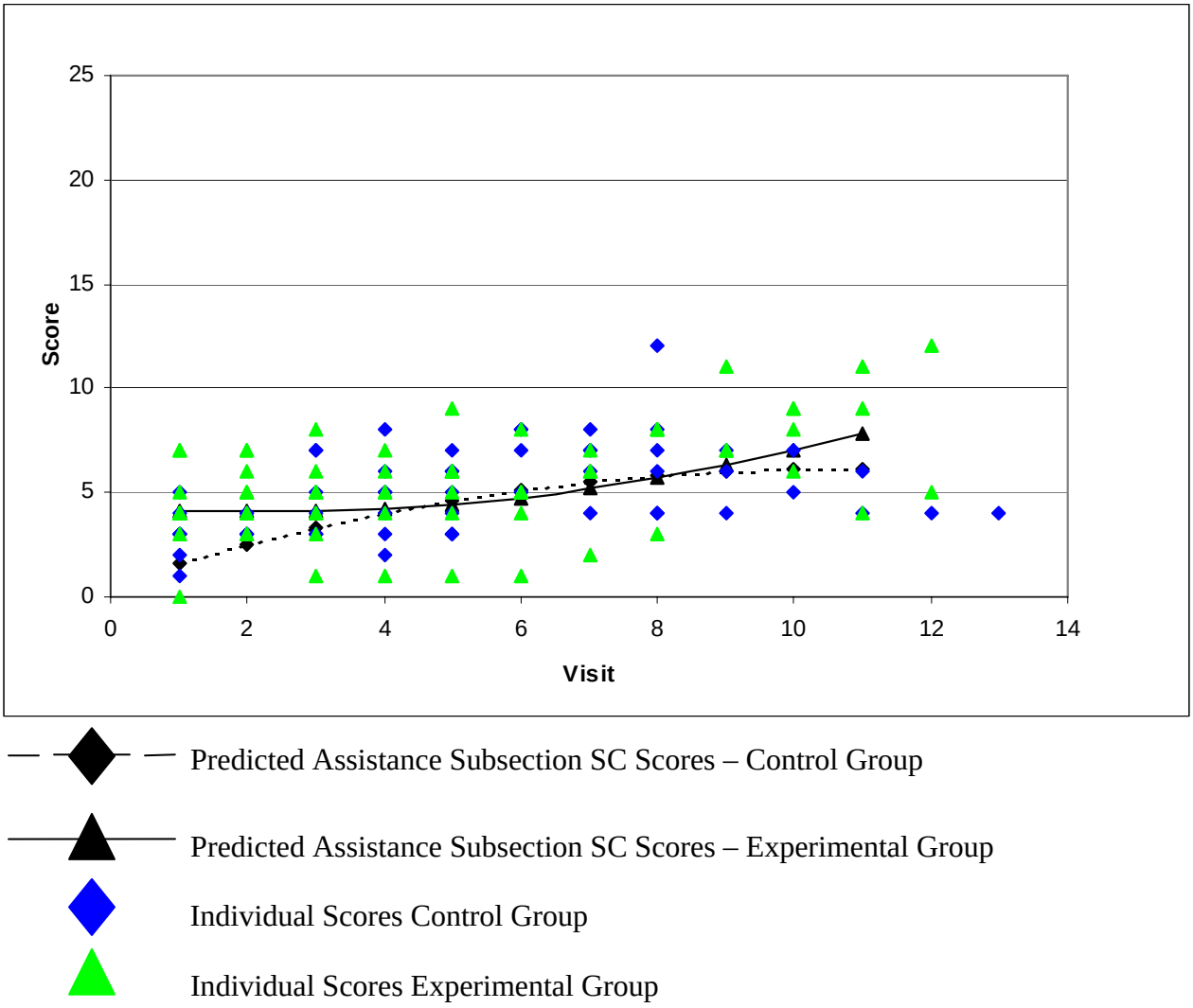
Non-significant interactions were removed from the model.

Figure 2.8. PEDI functional skills subsection S scores



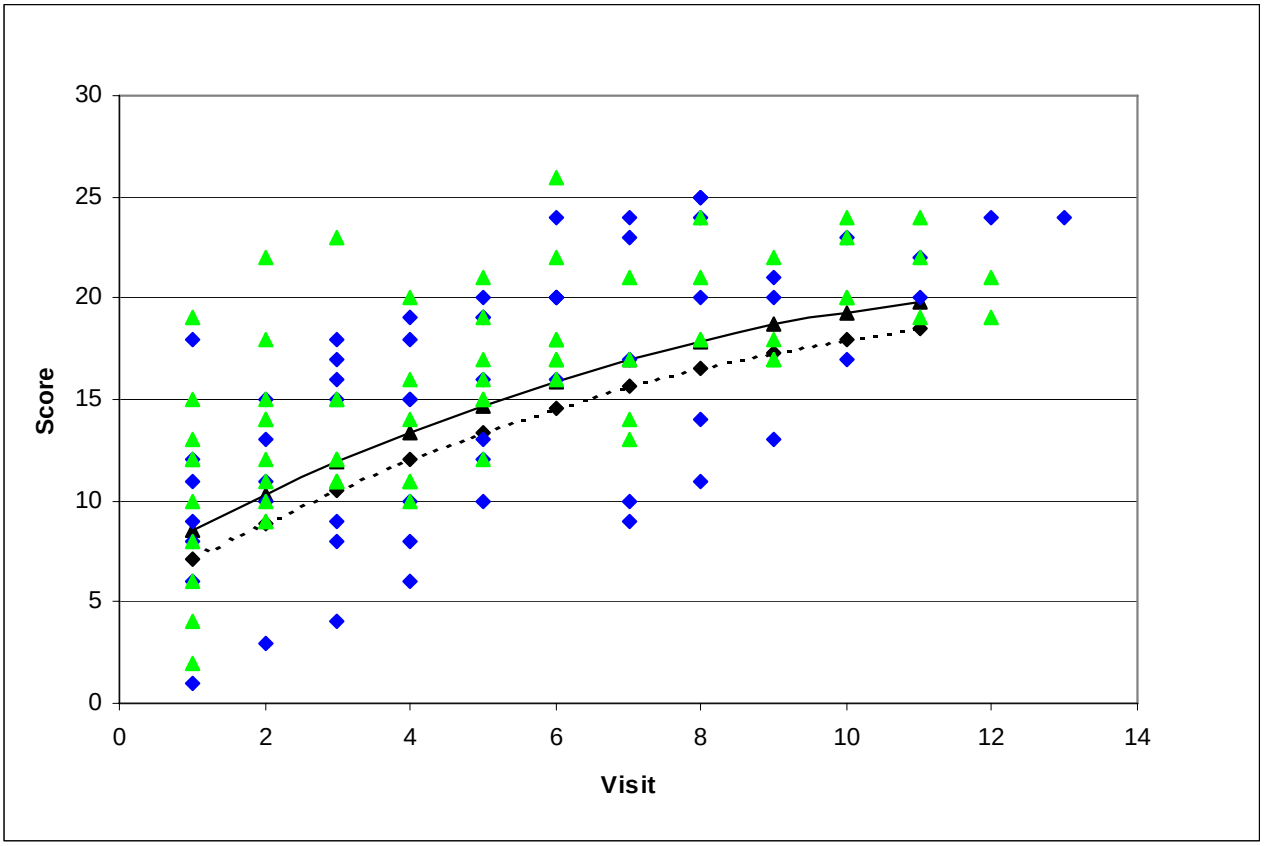
Non-significant interactions were removed from the model.

Figure 2.9. PEDI assistance subsection SC scores



Non-significant interactions were removed from the model.

Figure 2.10. PEDI assistance subsection M scores



Predicted Assistance Subsection M Scores – Control Group

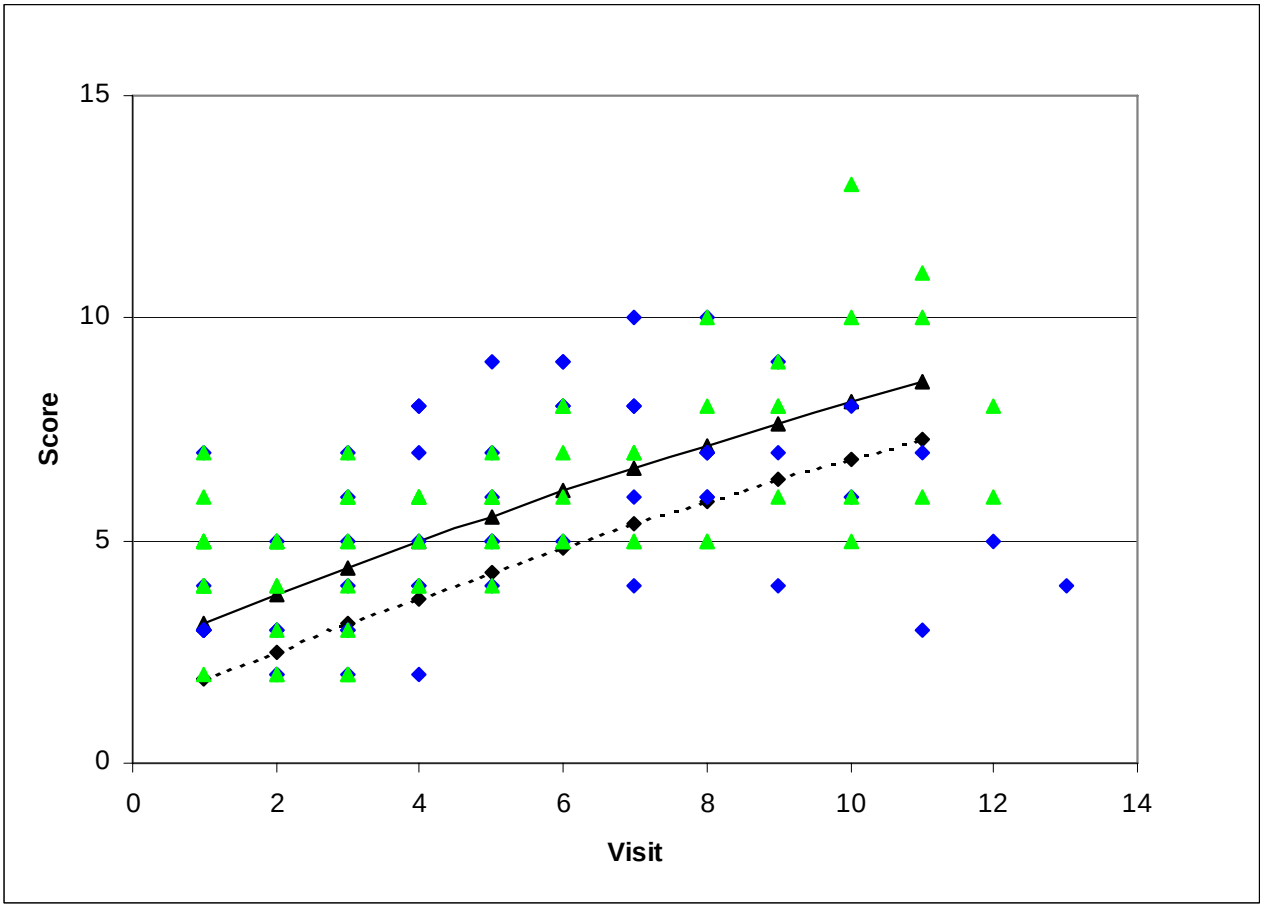
Predicted Assistance Subsection M Scores – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Non-significant interactions were removed from the model.

Figure 2.11. PEDI assistance subsection S scores



Predicted Assistance Subsection S Scores – Control Group

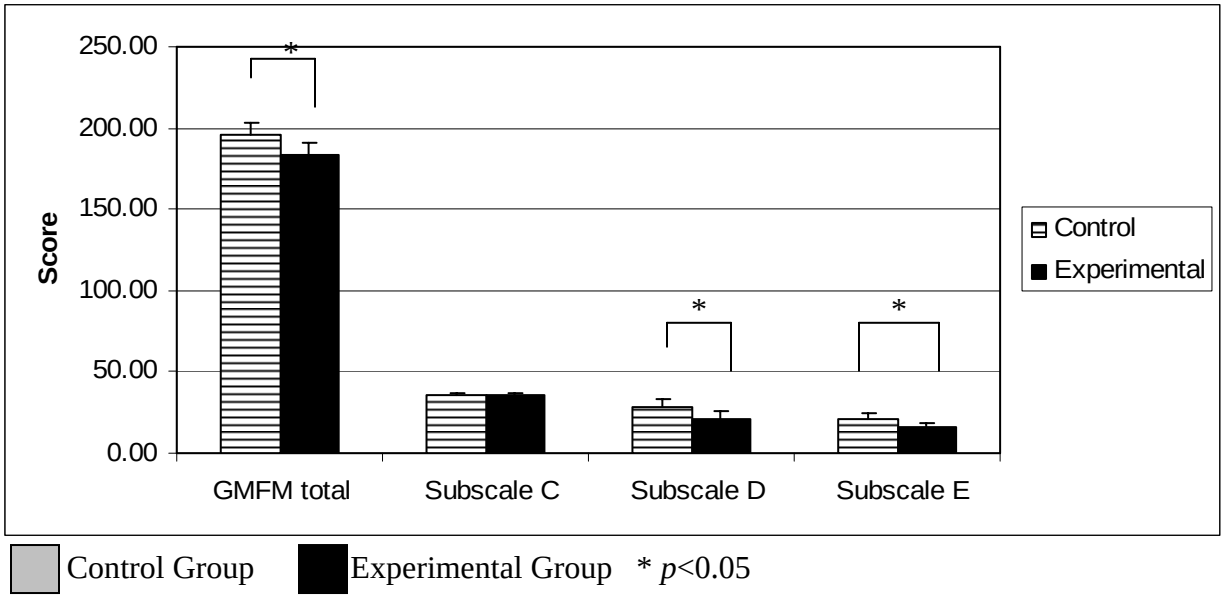
Predicted Assistance Subsection S Scores – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

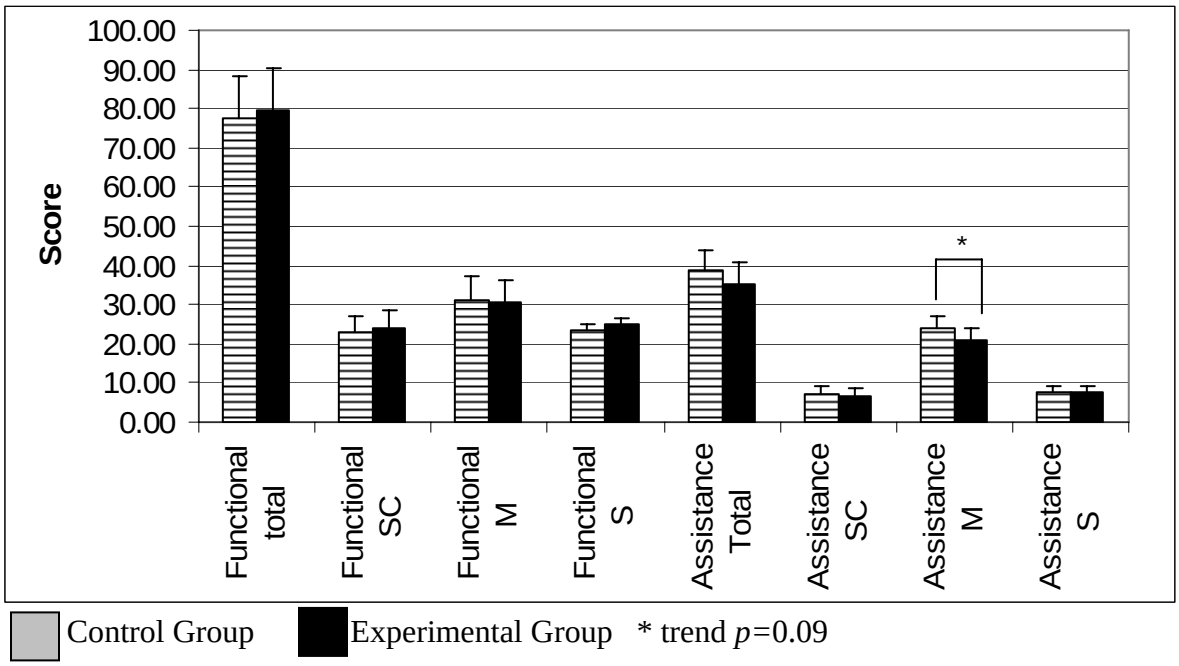
Non-significant interactions were removed from the model.

Figure 2.12. GMFM scores and subscores at 1 month of walking experience



Control Group Experimental Group * $p<0.05$

Figure 2.13. PEDI scores and subscores at 1 month of walking experience



Chapter 3

The effects of orthotic use on upright play participation in infants with Down syndrome

Introduction

Play is an essential part of childhood. Play is the activity by which children explore their environment and to learn how to deal effectively with their surroundings. It may be difficult to perform exploratory play in an upright posture when balance control is poor, as is the case in infants with Down syndrome (DS) (Kubo & Ulrich, 2006). However, supramalleolar orthose (SMOs) can improve balance in children with DS (Martin, 2004). Orthoses may improve the ability of children with DS to engage in upright exploratory play by providing foot and ankle stability. If their ability to control their base of support increases, they may be more able to use their hands to interact with objects during upright play, rather than for stability purposes.

Play is important for multiple aspects of development. The varied pattern of environmental stimulation that children obtain through play is important for optimal brain development (Rowland, 1998). The ability to move around and explore via crawling, walking, or powered mobility facitilates children's acquisition of cognitive skills, affective behaviors, and language skills (Biringen, Emde, Campos, & Appelbaum, 1995; Butler, 1986; Campos et al., 2000). Playing with peers, siblings, and parents is also an important way to explore and develop social skills (National Research Council and

Institute of Medicine, 2000). Play allows children to engage in and learn about the world around them.

Similar to children with typical development (TD), children with DS also use play to learn about their surroundings and how to move within their surroundings. However, Loveland (1987) states that children with DS may learn less effectively due to their less selective exploration style. Children with TD use exploration to figure out the relevant perceptual information and identify the affordances (or movement possibilities) within the environment (Berger & Adolph, 2007). Children with DS do not pick out the salient features of an object to explore but instead explore the whole object equally, leading to more repetitive play when compared to children with TD (Loveland, 1987). Playing in this manner may limit what a child is able to perceive about an object because it does not allow this child to attend to specific information about the object (Bushnell, 1993). In children with DS, this decreased ability to identify affordances may lead to delayed motor development because the movement possibilities associated with the environmental context are not fully explored.

Hand use is vital to play and exploratory behavior. When children are able to sit independently, they are able to use their hands to explore the area immediately around them. When children can support themselves in an upright position, they can see and explore a new visual perspective. Initially, though, the need to use their hands for weight bearing while upright may limit their manual exploration. In addition, infants with DS may not quickly learn to perceive the affordances available within reach. Children with DS may not understand that they can let go of a support surface and explore with their hands. There are also organismic constraints that limit infants' ability to let go of a

support surface to explore with their hands (Newell, 1986). These include impairments such as decreased muscle tone, ligamentous laxity, and slow reaction times (American Academy of Pediatrics, 2001; Shumway-Cook & Woollacott, 1985). These constraints lead to decreased postural control in children with DS, which affects their ability to play in an upright position and requires them to use their hands to support their body longer, when upright, instead of to explore.

Postural control

The development of postural control is important for the ability to explore the environment in an upright position. As children gain increased control of their posture, they are able to increase their exploratory ability. Bernstein (1967) hypothesized that during skill acquisition, there is first a freezing of the degrees of freedom, followed by a freeing of the degrees of freedom, and finally exploration and exploitation of the degrees of freedom. Typical development of postural control follows a similar process. Sveistrup and Woollacott (1996) looked at how postural ability changed over time in infants with TD. In their longitudinal study, they identified infants' postural reactions in static standing beginning when the infants could pull to stand and continuing through late independent walking. They found that in upright, as the infants behavior progressed, the postural activation pattern went from a high level of tonic activity to phasic muscle activation. In addition, Hadders-Algra and colleagues (1998) also found high levels of tonic activity with children who were learning to stand and walk. They proposed that the fixed extensor pattern seen in these children may be a temporary solution for balance problems. Sveistrup and Woollacott (1996) suggested this tonic activity is an attempt by children to stabilize multiple joints. As infants begin to learn to stand independently,

they use increased tonic activity to stabilize their joints and a fixed extensor pattern as an "all or nothing" response. These strategies are used until they gain enough experience to begin to free up the degrees of freedom. A similar pattern is seen in typically developing children when they learn to cruise, or walk sideways and use their upper extremities for support. Haehl and colleagues (2000) suggest that typically developing infants begin to cruise only moving one limb at a time. As the infants' trunk control improves, they begin use their trunk as the interface between the arms and legs which allows freer movement of the arms and legs (Haehl, Vardaxis, & Ulrich, 2000). As the children begin to free up the degrees of freedom associated with postural control, they are able to explore their environment more thoroughly and devote less energy to maintaining balance and more energy to the process of exploration.

In children with DS, abnormal postural control may contribute to their atypical exploratory play behavior. Poor postural development leads to a decreased set of movement possibilities (affordances) within a given environment (Berger & Adolph, 2007). While children with DS develop upright balance, they have delayed and atypical postural control when compared to children with TD. Kubo and Ulrich (2006) found that new walkers with DS take longer to learn to couple oscillations in the saggital and frontal planes during gait. In addition to this, Schuway-Cook and Woollacott (1985) hypothesize that children with DS are not only delayed, but also show a different developmental trend in their postural development when compared to children with TD. They found that children with DS between the ages of 1 and 3 have sway responses that are slow, poorly organized, and inconsistent compared to the consistent sway response seen in children with TD at the same age range. This atypical postural response may lead to different

solutions to maintaining upright or loss of balance and may influence a child's ability to explore multiple environmental contexts.

Orthoses

An intervention that may help children with DS maintain balance is lower extremity orthotic use. Supramalleolar orthoses (SMOs) are external devices that are placed in shoes to provide support to the foot and ankle. They help prevent calcaneal eversion by aligning the subtalar joint in a neutral position, thus improving the bony alignment of the foot and ankle, and influencing postural and gait characteristics (Orner, Turner, & Worrell, 1994)). Supramalleolar orthoses lead to improved postural stability in children with DS. Martin (2004) studied SMO use in children with DS, and found that the children, age 3 years 6 months through 8 years, showed immediate and long term improvements in postural stability, as measured by standardized test scores, when they used flexible SMOs. The orthoses provided foot and ankle stability that lead to improved balance. These results are limited to older children, but infants with DS may also benefit from SMO use. The improved balance and stability gained from SMO use may lead to an increase in hands-on exploratory play.

Purpose

The purpose of this study is to determine if SMO use in pre-walkers with DS during an upright play situation contributes to increased hands-on exploratory play.

Method

Participants

Twenty-two children with DS enrolled in this study. Overtime, 5 children dropped out of the study (1 had emerging medical problems, 1 did not tolerate the treadmill, and 3 received orthoses prior to the end of the study.) The study was limited to infants with trisomy 21 and onset of participation occurred when infant were able to pull to stand at the furniture but not yet cruise. Children were excluded from this study if they had a history of other developmental disabilities, uncorrected visual or hearing impairment, previous orthotic use, orthotic use other than the ones provided for this study, or were unable to tolerate the orthoses. The 22 children were randomly assigned into a control (n=11) or an experimental group (n=11). As a result of drop out, the final distribution of subjects was 7 (control group) and 10 (experimental group). The children in the experimental group were measured for SMOs on their first visit and received them before their second visit. The children in the control group were measured for SMOs when they could take three independent steps and received them in one to two weeks. (See figure 3.2 for a picture of the SMOs.) Each child participated in this study from the time he could pull to stand until he could take 3 steps independently. A parent of each child provided written informed consent prior to study entry. The Institutional Review Board at the University of Michigan approved this study.

Data Collection

Upright play activity was recorded with the following set-up (see figure 3.2): A Leap Frog Learning Table (Leap Frog ™) was placed next to the family's couch. A

mirror was placed behind the table and couch (opposite the camera) at a slight angle to allow researchers to view the side of the child that faced away from the video camera. The digital video camera was placed so that the infant's legs and trunk were within the frame. The video carmera recorded at 30 hz. The researcher placed each child in an upright position between the table and couch, facing the table. The 20 minutes of recorded play time began when the primary researcher took her hands off of the child. Researchers and family members sat on the other side of the table and encouraged the child to stand and play. If the child sat or fell down, the parent and researchers encouraged the child to pull to stand at either the table or the couch but did not assist the child in standing up. A child who moved out of view of the camera (by crawling) was placed in sitting between the table and the couch and then encouraged to pull to stand using the activities on the table or toys that the child likes. During the play sessions, children in the experimental group wore SMOs and children in the control group wore shoes.

Data Reduction

Each recorded 20-minute session was behavior coded in the lab. Behavior coders determined the amount of time spent in upright and, when in upright, how children supported their weight and played. The length of these bouts in upright was recorded in seconds via the time code embedded in the digital video tape. Coders counted only behaviors that were 30 consecutive frames or longer. When children were in an upright position, coders determined whether their trunk was leaning on the support surface or not, and whether they were using 0, 1, or 2 hands for weight bearing. Coders considered the children in upright when their hips were no longer moving in an upward motion. The

children were no longer considered upright when a portion of their body, other than feet, touched the ground, or when another person touched them. Following contact, the children would be considered upright again. The children's behavior was coded as cruising when they took 2 or more steps in one direction. A step required the children to pick up their foot and placed it down in a different spot. When the children moved from the floor to standing, the behavior was coded as pulling to stand. For each behavior, coders recorded time of onset and end, from which durations were calculated.

Seven coders participated in the upright behavior coding described above. They all coded a random selection of play sessions and showed high inter-rater reliability (ICC=1.00 for time in upright, ICC=0.998 for time in leaning, ICC=0.990 for time in 2-handed support, and ICC=0.969 for time in 1 handed support.)

Data Analysis

The data were used in three separate analyses. First, hand support as a percentage of total time in upright was analyzed. Second, hand support while leaning was analyzed. Third, hand support while not leaning was analyzed. The slopes of these changes over time were compared using mixed linear models. The models included a term for the linear effects of time, the quadratic effects of time, group effect, and both possible group by time (linear or quadratic) interactions. The models also allowed time to vary randomly by subject. Alpha was set at 0.05.

Results

Participant Characteristics at Study Onset

The participants' characteristics at study onset are reported in table 3.1. The control group had a longer birth length than the experimental group. However, length was not different at study onset. The experimental group wore the SMO 6.25 (±4) hours per day. Table 3.2 shows that there were no group differences for time in upright and leaning at study onset.

Time in upright

The predicted percent of time spent in upright over the course of the study is shown in figure 3.3. Across the course of the study, the mean amount of time spent in upright during a 20-minute (1200 second) play session was 12.33 (±5.15) minutes (740 (±309) seconds) for the control group and 12.28 (±5.28) minutes (737 (±317) seconds) for the experimental group. There was no significant difference between the groups in the amount of time that the children spent in upright ((*F*(52,1)=0.93 *p*=0.34). As the study progressed, the amount of time spent in upright during the 20-minute play period did not change as a linear function (*F*(42,1)=1.78 *p*=0.19) or a quadratic function (*F*(44,1)=0.70 *p*=0.41). There were no significant group by time interactions (*F*(42,1)=1.38 *p*=0.25 linear, (*F*(44,1)=1.18 *p*=0.28 quadratic).

Time in Leaning

Figure 3.3 shows that the mean amount of time spent leaning across the course of the study was 8.03 (± 4.48) minutes (485 (±269) seconds) in the control group and 7.57

(±5.32) minutes (454 (±319) seconds) in the experimental group. There was no difference between the groups in the amount of time the children spent leaning on the play surface ($F(44,1)=2.16$ $p=0.15$). As the study progressed, the amount of time in leaning did not increase as linear function ($F(52,1)=1.74$ $p=0.19$)or a quadratic ($F(50,1)=1.47$ $p=0.23$. There was a trend towards a group X time (linear and quadratic) interaction for time in leaning ($F(52,1)=2.98$ $p=0.09$ and $F(50,1)=3.20$ $p=0.08$, respectively).

The mean percentage of time spent leaning compared to the time spent in upright over the course of the study was 65% (±23) for the control group and 62% (±35) for the experimental group. There was no significant difference in the percentage of time spent leaning between the groups ($F(46,1)=0.09$ $p=0.77$). There was no significant change over time in the percent of time spent leaning ($F(52,1)=0.02$ $p=0.89$ linear, ($F(48,1)=0.18$ $p=0.67$ quadratic). There was also no significant group by time interaction ($F(52,1)=0.20$ $p=0.65$ linear, $F(48,1)=0.45$ $p=0.51$ quadratic).

Total hand use while upright

The average percentage of time spent throughout the study in leaning with 2 hands on the table while in upright was 32.50% (±27.69) for the experimental group and 26.90% (±18.68) for the control group over the course of the intervention (fig 3.4). There was no statistical difference between the groups ($F(54,1)=1.17$ $p= 0.28$). Over the course of the intervention, there was a significant linear decrease in 2-hand support ($F(46,1)=4.15$ $p=0.05$). There was no significant quadratic change over time ($F(48,1)=1.43$ $p=0.24$). There were no significant group by time interactions (linear ($F(46,1)=2.38$ $p=0.13$), quadratic ($F(48,1)=1.97$ $p=0.17$).

The average percentage of time spent in leaning with 1 hand on the play surface while in upright was 20.38% (±16.18) for the experimental group and 27.88% (±14.00) for the control group over the course of the intervention (figure 3.5). There was no statistical difference between the groups ($F(54,1)=0.31$ $p=0.58$). There were no linear or quadratic effects of time (linear $F(48,1)=0.30$ $p=0.59$, quadratic $F(50,1)=0.17$ $p=.68$). There was no significant linear group by time interaction ($F(48,1)=0.02$ $p=0.88$). There was no significant quadratic group by time interaction ($F(50,1)<0.01$ $p=0.99$).

The average percentage of time spent in leaning with 0 hands on the play surface while in upright was 6.80% (±11.90) for the experimental group and 10.71% (±10.39) for the control group over the course of the intervention (Figure 3.6). There was no statistical difference between the groups ($F(54,1)=0.12$ $p=0.73$). There were no linear or quadratic effects of time (linear $F(44,1)=0.97$ $p=0.33$, quadratic $F(47,1)<0.01$ $p=0.98$). There was no significant linear group by time interaction ($F(44,1)=1.82$ $p=0.18$). There was no significant quadratic group by time interaction ($F(47,1)=1.73$ $p=0.20$).

The average percentage of time spent not leaning with 2 hands on the play surface while in upright was 19.68% (±21.24) for the experimental group and 16.55% (±15.27) for the control group over the course of the intervention (figure 3.4) . There was no statistical difference between the groups ($F(53,1)=0.81$ $p=0.37$). There were no linear or quadratic effects of time (linear $F(44,1)=0.18$ $p=0.68$, quadratic $F(44,1)=0.05$ $p=0.83$). There was no significant linear group by time interaction ($F(44,1)=0.26$ $p=0.61$). There was no significant quadratic group by time interaction ($F(44,1)=0.04$ $p=0.85$).

The average percentage of time spent not leaning with 1 hands on the play surface while in upright was 19.13% (±19.99) for the experimental group and 13.84% (±14.43)

for the control group over the course of the intervention (figure 3.5). There was no statistical difference between the groups ($F(46,1)=0.50$ $p=0.48$). There was a trend towards a linear increase over time ($F(42,1)=3.54$ $p=0.07$). There was not significant quadratic effect of time ($F(42,1)=0.64$ $p=0.43$). There was no significant linear group by time interaction ($F(42,1)=0.47$ $p=0.50$). There was no significant quadratic group by time interaction ($F(42,1)=0.59$ $p=0.48$).

The average percentage of time spent not leaning with 0 hands on the play surface (or standing) while in upright was 1.48% (±5.20) for the experimental group and 0.40% (±1.22) for the control group over the course of the intervention (figure 3.6). There was no statistical difference between the groups ($F(39,1)=1.00$ $p=0.33$). There was no significant linear effect of time ($F(36,1)=0.75$ $p=0.39$). There was a significant quadratic increase over time ($F(36,1)=5.26$ $p=0.03$). There was no significant linear group by time interaction ($F(36,1)=0.03$ $p=0.86$). There was no significant quadratic group by time interaction ($F(36,1)=0.01$ $p=0.91$).

Hand use while upright and not leaning

The average percentage of time spent throughout the study with 2 hands on the table while upright but not leaning was 43.70% (±29.01) for the experimental group and 54.36% (±28.14) for the control group over the course of the intervention (figure 3.7). The control group spent significantly more time with 2 hands on the support surface than the experimental group ($F(55,1)=8.76$ $p= 0.01$). There was no significant linear effect of time ($F(44,1)=1.55$ $p=0.22$). There was no significant quadratic effect of time ($F(45,1)=0.68$ $p=0.42$). There was no significant linear group by time interaction

($F(44,1)=2.44$ $p=0.13$). There was no significant quadratic group by time interaction ($F(45,1)=0.91$ $p=0.35$).

The average percentage of time spent throughout the study with 1 hand on the table while not leaning 38.28% (±26.47) for the experimental group and 40.97% (±26.33) for the control group over the course of the intervention (figure 3.8). There was no significant difference between the groups ($F(52,1)=1.01$ $p= 0.32$). There was a significant linear increase over time ($F(43,1)=5.00$ $p=0.03$). There was no significant quadratic effect of time ($F(44,1)=1.12$ $p=0.30$). There was no significant linear group by time interaction ($F(43,1)=1.44$ $p=0.24$). There was no significant quadratic group by time interaction ($F(44,1)=1.25$ $p=0.27$).

The average percentage of time spent throughout the study with 0 hands on the table while not leaning (or standing) was 2.43% (±8.57) for the experimental group and 1.82% (±5.18) for the control group over the course of the intervention (figure 3.9). There was no significant difference between the groups ($F(47,1)=1.25$ $p= 0.27$). There was no significant linear effect of time ($F(35,1)=0.67$ $p=0.42$). There was a significant quadratic effect of time ($F(36,1)=4.66$ $p=0.04$). There was no significant linear group by time interaction ($F(35,1)=0.13$ $p=0.72$). There was no significant quadratic group by time interaction ($F(36,1)<0.01$ $p=0.95$).

Hand use while upright and leaning.

The average percentage of time spent throughout the study with 2 hands on the table while leaning was 47.59% (±27.65) for the experimental group and 41.87% (±23.76) for the control group over the course of the intervention (figure 3.10). There

was a trend towards a group difference indicating that the experimental group spent more time with 2 hands on the support surface while leaning ($F(54,1)=2.77$ $p= 0.10$). Over the course of the intervention, the percentage of time spent with 2 hands on the play surface while leaning showed a significant linear decrease for both groups ($F(47,1)=7.22$ $p=0.01$). There was a trend towards a quadratic change over time ($F(49,1)=3.31$ $p=0.08$). There was a significant group by time (linear) interaction ($F(47,1)=3.98$ $p=0.05$) and a trend toward a significant group by time (quadratic) interaction ($F(49,1)=3.57$ $p=0.07$) indicating that the experimental group showed a steeper decrease in the amount of time spent with 2 hands on the table while leaning

The average percentage of time spent with 1 hand on the play surface while leaning was 34.60% (±21.92) for the experimental group and 42.57% (±15.93) for the control group over the course of the intervention (figure 3.11). This was a trend in favor of the control group ($F(48,1)=3.86$ $p=0.06$). Over the course of the intervention, the percentage of time spent with 1 hand on the play surface significantly increases linearly ($F(40,1)=9.53$ $p<0.01$) and quadratically ($F(42,1)=6.73$ $p=0.01$). In addition, there were trends towards group by time interactions both linearly and quadratically ($F(40,1)=3.02$ $p=0.09$, $F(42,1)=3.09$ $p=0.09$, respectively).

The average percentage of time spent with 0 hands on the play surface while leaning was 11.47% (±19.64) for the experimental group and 15.45% (±15.78) for the control group over the course of the intervention (figure 3.12). There was no statistical difference between the groups ($F(54,1)$ $p=0.61$). There were no linear effects on time ($F(44,1)=0.65$ $p=0.42$). There were also no quadratic effects on time ($F(47,1)=0.10$

p=0.75). There was no significant linear group by time interaction (F(44,1)=2.84 p=0.10). There was a trend towards a quadratic group by time interaction (F(47,1)=3.11 p=0.08).

Discussion

The purpose of this study was to determine if SMO use in pre-walkers with DS improved upright balance during a play situation leading to increased exploratory play while in upright. While it was hypothesized that orthotic use would improve upright exploratory play, we found no positive effect of orthotic use on this behavior. In fact, pre-walkers with DS who did not use SMOs spent a larger percentage of time in leaning with one hand and no hands on the play surface than the pre-walkers who wore SMOs. Though exploratory play is an important part of development and the infants in this study all participated in hands-on exploratory play to some extent, the need to remain stable in upright appeared to trump the the ability to free up their hands for play and exploration.

Total time in upright and leaning

All of the infants in this study spent a similar amount of time in upright and leaning throughout the course of the study. Table 3.3 shows the average age of the infants and the average time in upright and leaning at each visit for the total study sample. It shows that the average amount of time in upright did not change much over time but the there was large individual variablity. It also shows that the amount of time in leaning was highly variable and peaked at visit 7. Though there were no differences over time or between groups in total time in upright and leaning, there were trends toward both linear and quadratic group by time interactions in total time in leaning. These indicate that the developmental path followed by each group over time is different (see figure 3.3). The experimental group follows an “inverted-U” trajectory, leaning the most

at visit number 5, while the control group shows a more linear trend in the amount of time leaning. This suggests that orthoses are influencing the developmental trends. The infants in the experimental group lean less than the control group at the beginning of the study. The amount of time in leaning increases as the time in upright increases. After visit 5, the absolute amount of time in upright continues to increase for both groups of children but the amount of time in leaning begins to decrease for the infants in the experimental group. At this point, the infant in the experimental group begin to support their weight by leaning on their hands instead of their trunks.

Total hand use while upright

The orthoses do not seem to effect hand use while in upright. There were no group differences in 2-,1-,and 0-hand support, whether leaning or not leaning as a percentage to time in upright. There were also no group by time interactions. The passage of developmental time appear to have the most effect on hand use while in upright. The time in 2-handed support while leaning as a percentage of time in upright significantly decreased during the study. The time in 1-handed support while not leaning as a percentage of time in upright also seemed to increase as time passed. In addition, time in standing increased quadratically at the end of the study just prior to walking onset. Both groups spent the majority of their time in upright in a leaning posture with 2 hands on the support surface. As time progressed they were able to free up 1 or 2 hands both in leaning and non-leaning to explore and play.

Hand use during non-leaning and leaning portions of upright

Hand use was also analyzed separately during non-leaning and leaning portions of the time in upright. During the non-leaning portion of time in upright, the control group spent a significantly larger percentage of time with 2 hands on the support surface. During the leaning portion of time in upright, the statistical trend suggests that the control group spent more time in 1-hand support. In addition, there was a significant group by time interaction indicating that the groups were developing in a different manner. The control group does appear to explore more with their hands but also appears to lean more in order to explore. When infants in both groups removed their trunks from the support surface, they spent more time with both hands in a weight bearing, stabilizing position.

Though the control group spent a smaller percentage of time with 2 hands on the support surface during the non-leaning portion of the time in upright, there was no overall difference between the groups over the course of the study in 2-handed support while leaning. However, all infants decreased the time they spent in with 2 hands on the support surface in leaning and non-leaning. The 2-handed leaning position in very stable; it is the dominant posture at the beginning of the study, when infants are just learning how to stand. At this point they used their trunk and both hands to stabilize their body. As children learn to control their bodies better, they use their trunk to support their body in a more adaptive manner, first by leaning against the support surface and then by supporting their trunk in upright. This leads to the observed decrease in 2-hand support as the study progressed and allows infants to begin playing and exploring with their hands.

While there was no difference between the groups in the percentage of time spent in 1-hand support during non-leaning, the control group performed better than the experimental group in the percentage of time spent in 1-hand support while leaning over the course of the study. Over time, all the infants in the study increased the amount of time in 1-hand support in both leaning and non-leaning. In leaning, the results showed that although there was not a linear effect over time there was a quadratic effect. This reflects the "inverted-U" shaped pattern in the statistical predictions. One-hand support peaks at visit 5 and 7 then begins to decline as 0-hand support begins to increase. The trend toward significant group by time interactions suggests that these 2 groups are developing in a slightly different manner. The control group begins with a larger percentage of time in 1-handed support while leaning and peaks on visit 5 where the experimental group builds up to a peak at visit 7.

There was no difference between the groups for 0-handed support during the time in leaning and non-leaning. However, there was a trend toward a quadratic group by time interaction in 0-hand support during the time in leaning. There was also a significant quadratic effect of time on 0-hand support during the time in non-leaning indicating an abrupt appearance of unsupported stance just before walking onset. The use of 2 hands for play (0 hands for support) while leaning signals a non-linear switch from trunk leaning as necessary to maintaining upright to trunk leaning as an adaptive skill that allows 2-handed exploration. Though there were no group differences over the course of the study, the predicted means show that this shift occurs earlier in the control group. The control group begins to play and explore using 2 hands while leaning at the 5-month visit while the experimental group achieves this at the 7-month visit. This increased

amount of time to explore may give the control group the opportunity to develop a deeper understanding of and experience with their surrounding.

When looking at how hand support changes over time, it is important to note that both groups spent the largest amount of time in 2-hand support while leaning in this play context. However, the control group appears to have an advantage when not in 2-hand support during leaning. At the time when 1-hand support during leaning appears higher in the experimental group, the control group is more likely to be spending a longer period of time leaning with no hands on the support surface. Interestingly, the predicted percentages indicate that the experimental group steadily decreases the time they spend leaning while the control group remains somewhat constant. However, while both groups show an increase decrease in the amount of 1- and 0-handed support, the control group appears to spend more time in 1- and 0-handed support though they tend to be leaning more. Perhaps the orthoses provided enough stability for the infants to begin exploring their trunk stability while exploring and playing with toys in their environment less. As the infants' ability to control their legs and trunk increased, their ability to explore with one hand while not leaning also increased. Overall the infants who did not wear orthoses spent more time using their hands to explore because they relied more on their trunk for support.

Engaging in and learning about the world is implicit in the act of playing. In addition, hand use is a key component to exploratory behavior. The children in this study displayed a progression from limited ability to free their hands from a support role to decreased hand support while in upright. Children with TD would show a similar progression from 2-handed support to 1- or 0-handed support. However, they may be

able to explore more than children with DS in an upright position because they require less hand support in an upright play situation. In addition, treadmill training in infants also leads to improved postural control so treadmill training may also influence the exploratory ability in children with TD.

Though the use of SMOs in infants with DS seems to improve stability enough to decrease the amount of time spent leaning in this pre-walking population, it inadvertently may further limit the ability of infants with DS to use their hands to explore while in upright. Because the infants are actively exploring their trunk movements, they are relying on upper extremity weight bearing for stability. This increased stability comes at the cost of hands-on exploration.

Conclusion

Orthoses are often recommended for pre-walkers with DS despite little to no empirical evidence to support the practice. The results of this study question this practice. While the use of SMOs may improve upright stability, it does not appear to improve upright exploration with the hands in pre-walkers with DS. In fact, the early use of SMOs may inhibit this behavior and could have a negative impact on future development. Based on this information, health care professionals may want to refrain from using SMOs in pre-walkers with DS.

There are a few limitations that should be kept in mind when considering the results and conclusions of this study. This study did not have an orthoses only group. Both groups received treadmill training. It is possible that the treadmill training sufficiently improves postural control in infants with DS to allow for less support in upright. Though orthoses in addition to treadmill training appear to lead to a different

developmental pattern than treadmill training alone, orthoses alone may lead to yet a different developmental pattern. In future studies, an orthoses only group should be included. This study did not consider individual differences in terms of orthotic type. Less supportive orthoses may be more beneficial to some children with DS and may have less developmental implications. Future studies are needed to determine how to choose orthotic type and whether other forms of orthoses may be more beneficial. In addition, the sample size in this study is small due to a high number of dropouts in the control group whose physical therapists recommended orthotic use before the child could walk. This resulted in lower statistical power. Future studies should include a larger sample size. Future studies should also monitor the infants' level of physical activity over developmental time to determine if physical activity is influenced by orthotic use, pre and post walking onset.

References

American Academy of Pediatrics Counsel On Genetics (2001). American Academy of Pediatrics: Health supervision for children with Down syndrome. *Pediatrics,* 107(2), 442-449.

Berger, S. E., & Adolph, K. E. (2007). Learning and development in infant locomotion. *Progress in Brain Research,* 164, 237-255.

Bernstein, N. (1967). *The co-ordination and regulation of movements*. Oxford: Pergamon Press.

Biringen, Z., Emde, R. N., Campos, J. J., & Appelbaum, M. I. (1995). Affective reorganization in the infant, the mother, and the dyad: the role of upright locomotion and its timing. *Child Development,* 66(2), 499-514.

Butler, C. (1986). Effects of powered mobility on self-initiated behaviors of very young children with locomotor disability. *Developmental Medicine & Child Neurology,* 28(3), 325-332.

Campos, J. J., Anderson, D. I., Barbo-Roth, M. A., M., H. E., Hertenstein, M. J., & Witherington, D. C. (2000). Travel broadens the mind. *Infancy,* 1, 149-219.

Hadders-Algra, M., Brogren, E., & Forssberg, H. (1998). Developmentof postural control - differences between ventral and dorsal muscles. *Neuroscience and Biobehavioral Reviews,* 22(4), 501-506.

Haehl, V., Vardaxis, V., & Ulrich, B. D. (2000). Learning to cruise: Bernstein's theory applied to skill acquisition during infancy. *Human Movement Science,* 19(5), 685-715.

Kubo, M., & Ulrich, B. D. (2006). Early Stage of Walking: Development of Control in Mediolateral and Anteroposterior Directions. *Journal of Motor Behavior, 38*(3), 229-237.

Loveland, K. A. (1987). Behavior of young children with Down syndrome before the morror: Exploration. *Child Development,* 58(3), 768-778.

Martin, K. (2004). Effects of supramalleolar orthoses on postural stability in children with Down syndrome. *Developmental Medicine & Child Neurology,* 46(6), 406-411.

National Research Council and Institute of Medicine. (2000). Making friends and getting along with peers. In J. P. Shonkoff & D. A. Phillips (Eds.), *From Neurons to*

Neighborhoods; The Science of Early Childhood Development (pp. 165-169). Washington, DC: National Academy Press.

Newell, K. M. (1986). Constraints on the development of coordination. In M. G. Wade & H. T. A. Whiting (Eds.), *Motor Development in Children: Aspects of Coordination & Control* (pp. 341-360). Dordrecht: Martinus Nijhoff.

Orner, C. E., Turner, D., & Worrell, T. (1994). Effect of foot orthoses on the balance skills of a child with a learning disablility. *Pediatric Physical Therapy*, 6, 10-14.

Rowland, T. W. (1998). The biological basis of physical activity. *Medical Science in Sports and Exercise*, 30, 392-399.

Shumway-Cook, A., & Woollacott, M. H. (1985). The growth of stability: postural control from a development perspective. *Journal of Motor Behavior*, 17(2), 131-147.

Sveistrup, H., & Woollacott, M. H. (1996). Longitudinal development of the automatic postural response in infants. *Journal of Motor Behavior*, 28(1), 58-70.

Tables

Table 3.1. Characteristics of the Study Sample at Entry

	Control	Experimental	p-value
Corrected Age at Entry (Days)	578 (188)	642 (121)	0.41
Weight (lbs)	20.75 (3.07)	22.62 (1.35)	0.11
Height (CM)	75.81 (7.93)	78.67 (2.74)	0.30
Shank Length (CM)	13.93 (1.50)	14.77 (1.43)	0.26
Shank Circumference (CM)	18.27 (0.95)	17.69 (1.39)	0.35
Thigh Length (CM)	15.00 (1.50)	15.88 (1.58)	0.27
Thigh Circumference (CM)	26.04 (1.95)	25.28 (1.62)	0.39
Physical Therapy (min)	60 (85)	63 (75)	0.94
Birth Weight (Lbs)	6.14 (1.04)	7.01 (0.91)	0.10
Birth Height (in)	18.54 (0.9)	19.83 (0.98)	0.02*
Number of Siblings	2 (2)	1 (1)	0.23
Birth Order	2 (2)	2 (1)	0.20
Maternal Age (years)	32 (5)	34 (6)	0.43
Paternal Age (years)	34 (5)	33 (7)	0.87
Maternal Education (years)	15 (3)	16 (1)	0.36
Paternal Education (years)	14 (3)	15 (2)	0.65
Income (x1000)	80-100 (40)	60-80 (20)	0.64

Mean (SD)
$p<0.05$

Table 3.2. Upright skill level at Study Entry

	Control	Experimental	p-value
Time in Upright (sec)	657.43 (524.89)	573.75 (339.88)	0.69
Time in Leaning (sec)	481.02 (442.09)	327.83 (263.78)	0.38
% Time in Leaning	67.13 (23.64)	64.99 (34.35)	0.90
Leaning with 2 hands down (sec)	310.20 (318.73)	206.69 (184.59)	0.43
Leaning with 1 hand down (sec)	140.99 (134.22)	98.17 (105.86)	0.49
Leaning with 0 hands down (sec)	29.84 (32.44)	12.61 (15.76)	0.18
% Leaning with 2 hands down (sec)	66.99 (19.74)	70.12 (19.88)	0.76
% Leaning with 1 hand down (sec)	27.42 (14.09)	25.38 (19.60)	0.82
% Leaning with 0 hands down (sec)	5.59 (6.84)	4.19 (4.94)	0.64

Mean (SD)
* $p<0.05$

Table 3.3. Age and Time in Upright at Each Visit for the Total Sample

VISIT	AVERAGE AGE (months)	TIME IN UPRIGHT (seconds)	TIME IN LEANING (seconds)
1	20.5 (5.0)	612.38 (414.40)	393.42 (346.25)
3	22.2 (5.1)	763.97 (258.34)	511.14 (234.36)
5	23.8 (5.4)	727.91 (317.97)	469.33 (323.15)
7	24.5 (5.2)	770.59 (315.00)	546.47 (345.04)
9	28.7 (6.3)	754.10 (221.98)	458.46 (289.96)
11	33.0 (6.5)	882.41 (143.95)	425.58 (211.49)
Total Sample	24.1 (6.2)	720.34 (326.66)	458.94 (296.44)

Mean (SD)

Figures

Figure 3.1. Supramalleolar orthoses

Figure 3.2. Experimental Set-up

Figure 3.3. Predicted total time in upright and leaning

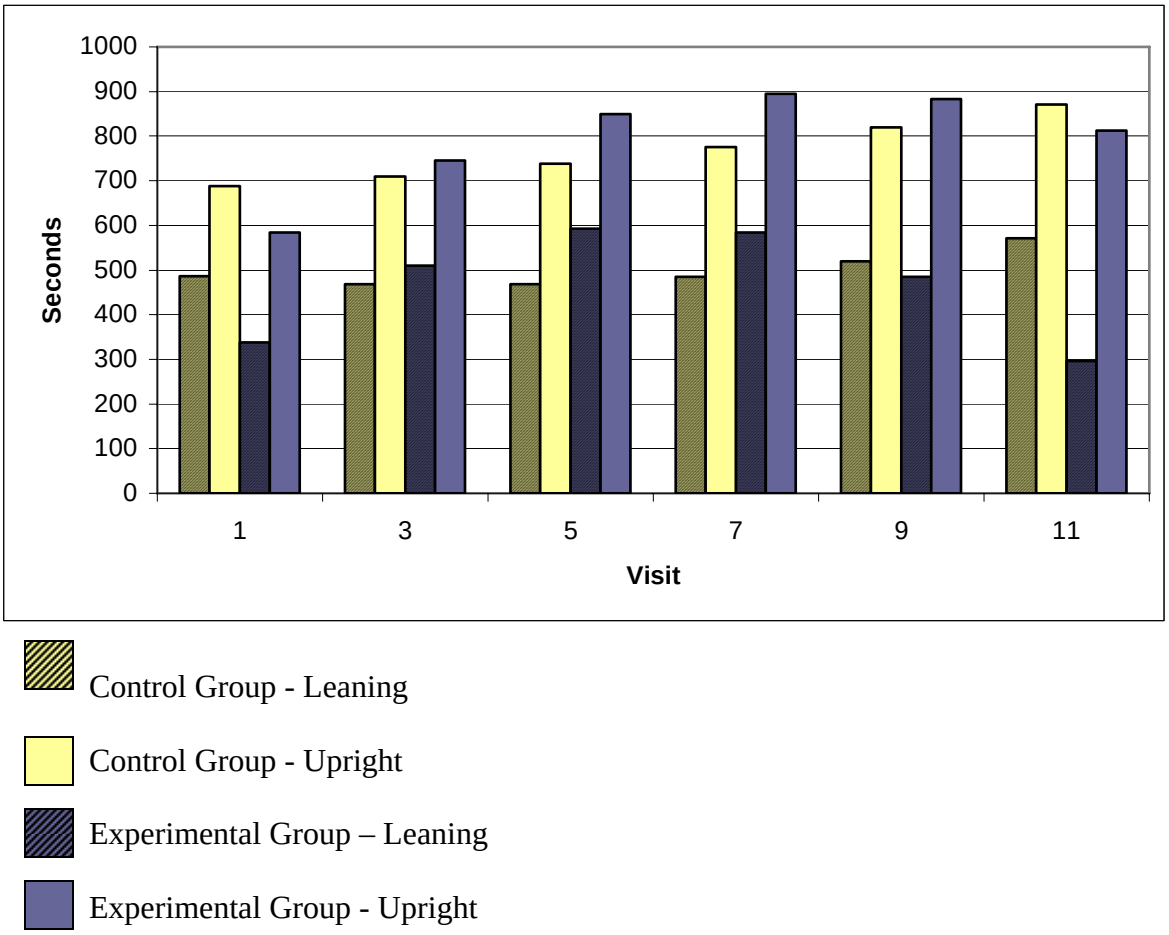


Figure 3.4. Percent of time in 2-hand support while in upright

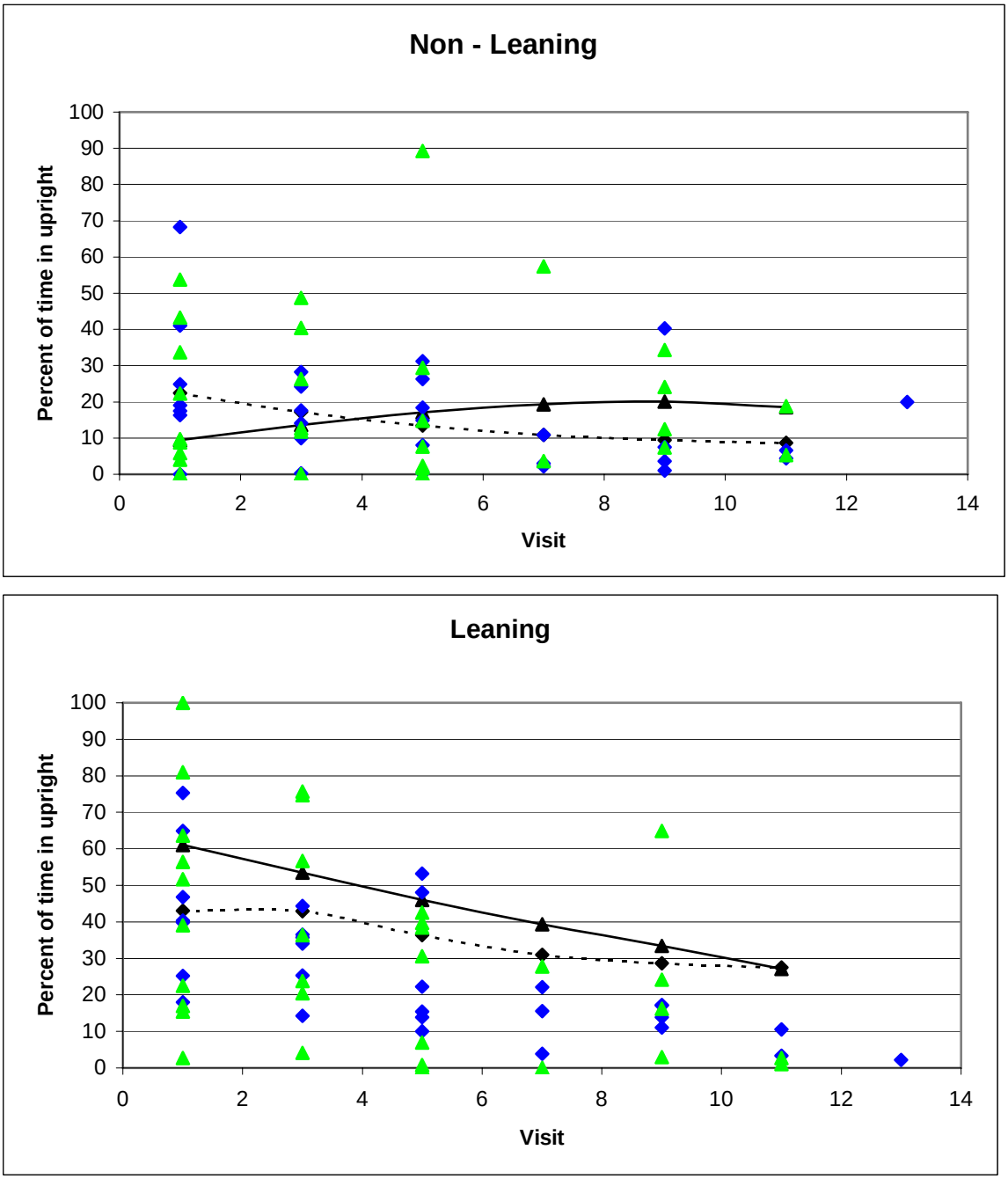


Figure 3.5. Percent of time in 1-hand support while in upright

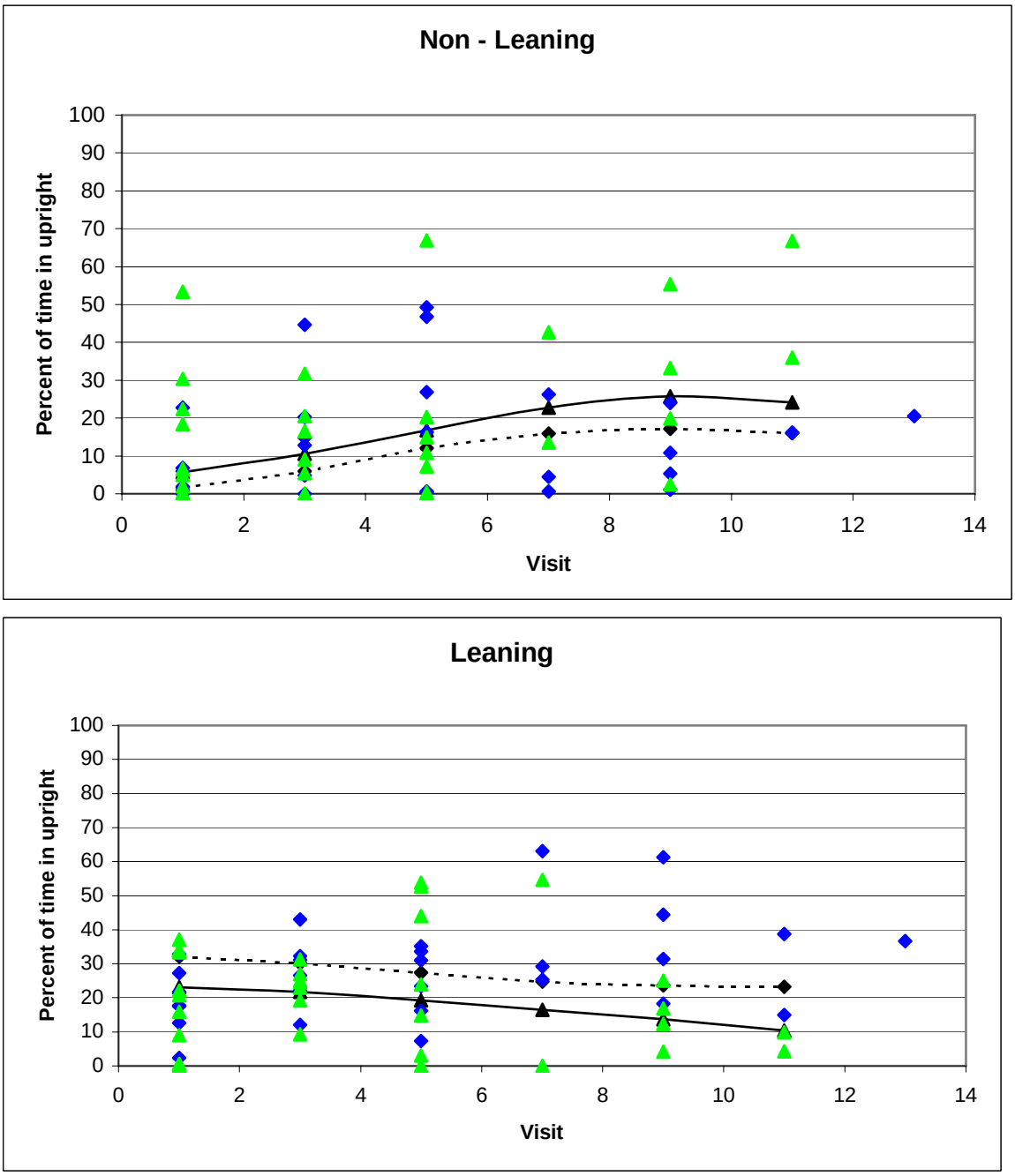
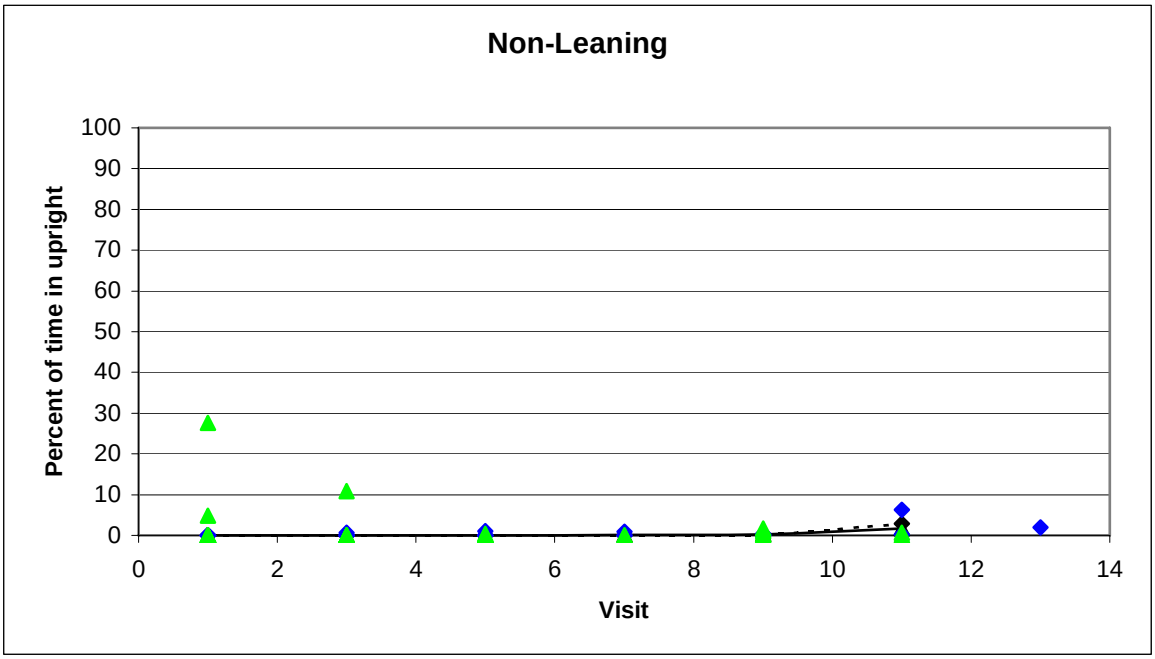
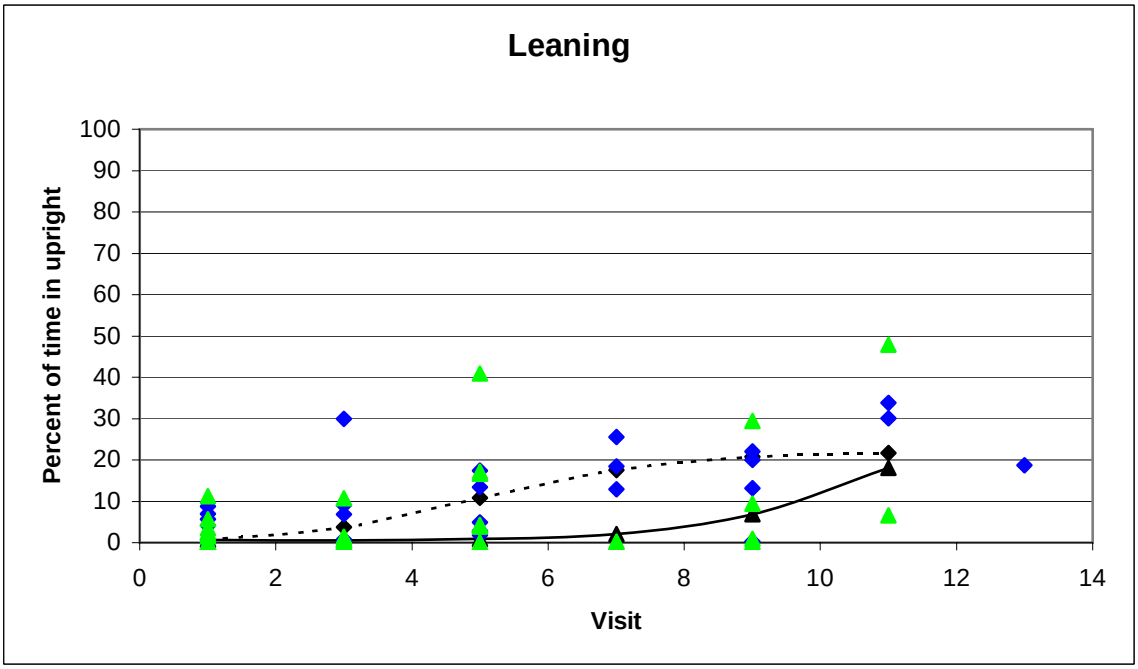


Figure 3.6. Percent of time in 0-hand support while in upright





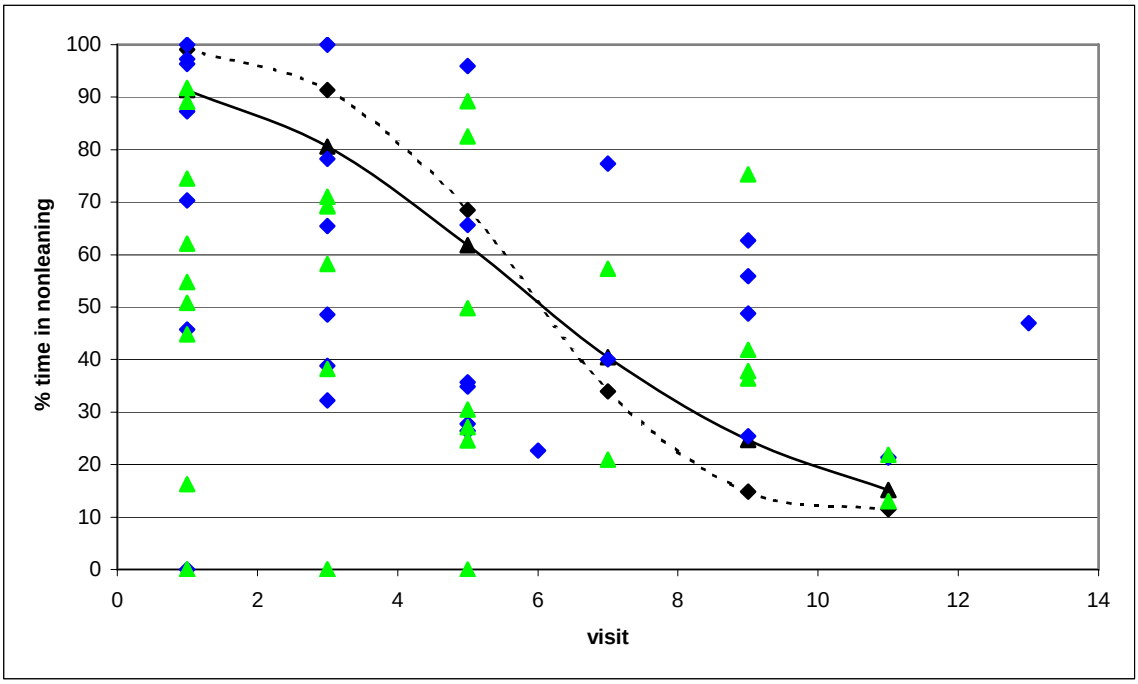
Predicted % time in upright – Control Group

Predicted % time in upright – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.7. Percent of time in 2-hand support while in non-leaning



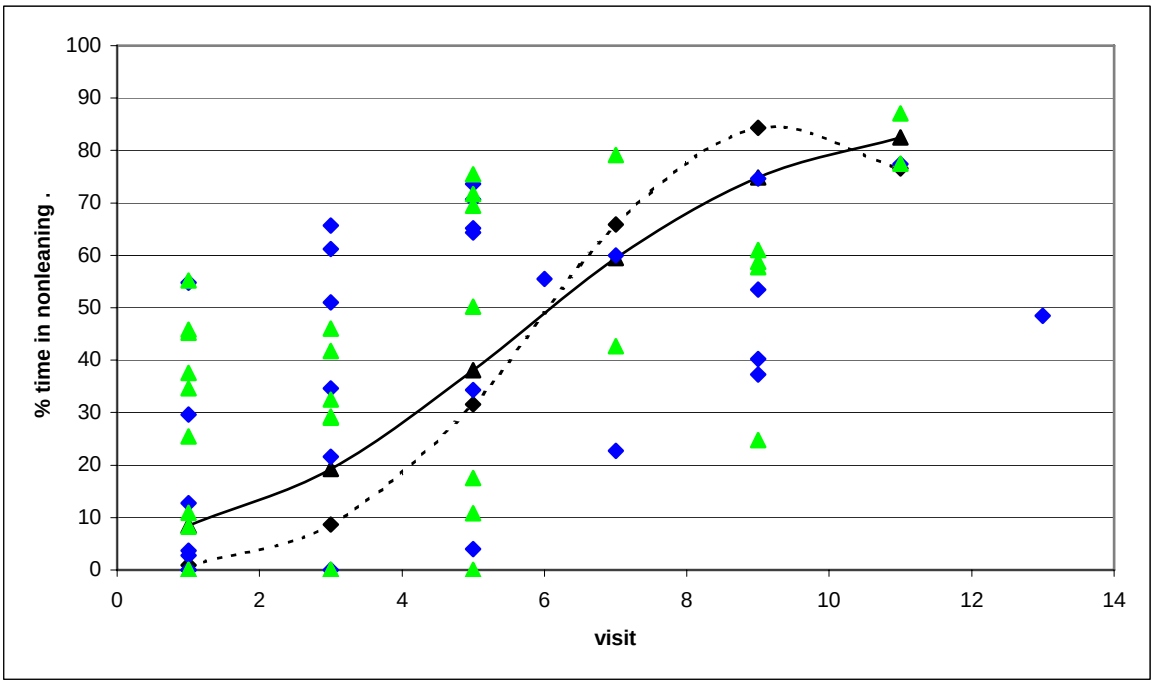
Predicted % time in non-leaning – Control Group

Predicted % time in non-leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.8. Percent of time in 1-hand support while in non-leaning



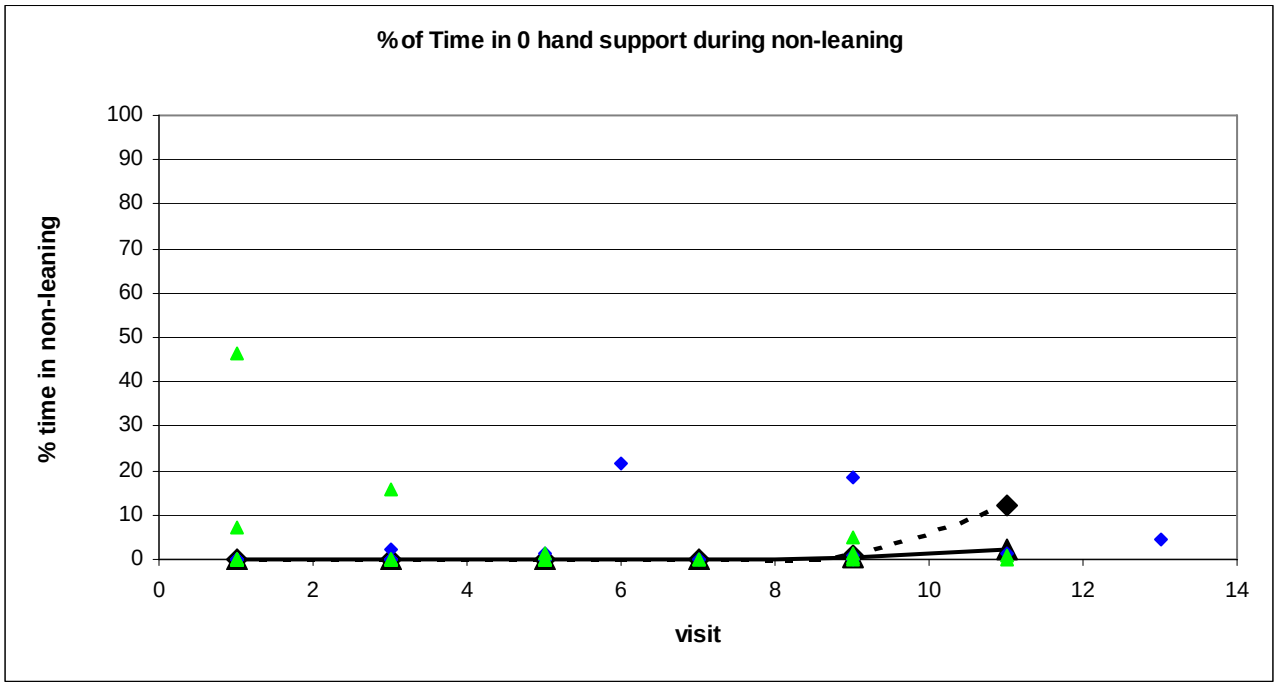
Predicted % time in non-leaning – Control Group

Predicted % time in non-leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.9. Percent of time in 0-hand support while in non-leaning



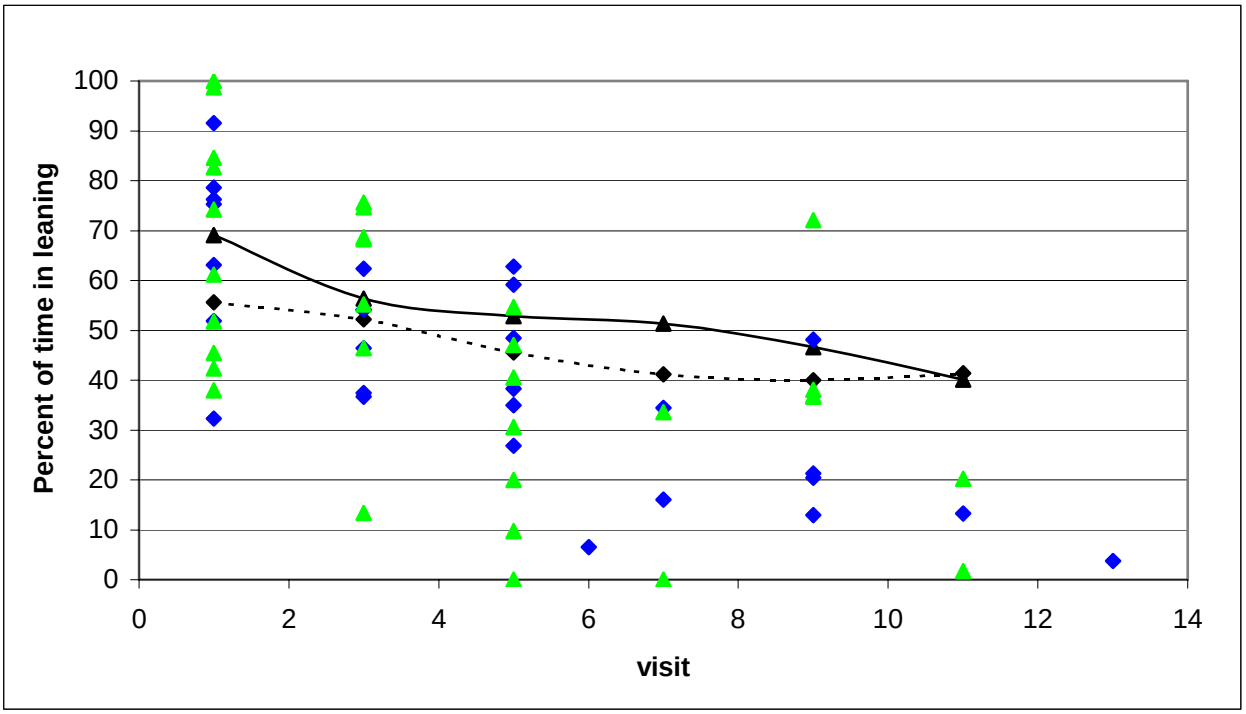
Predicted % time in non-leaning – Control Group

Predicted % time in non-leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.10. Percent of time in 2-hand support while in leaning



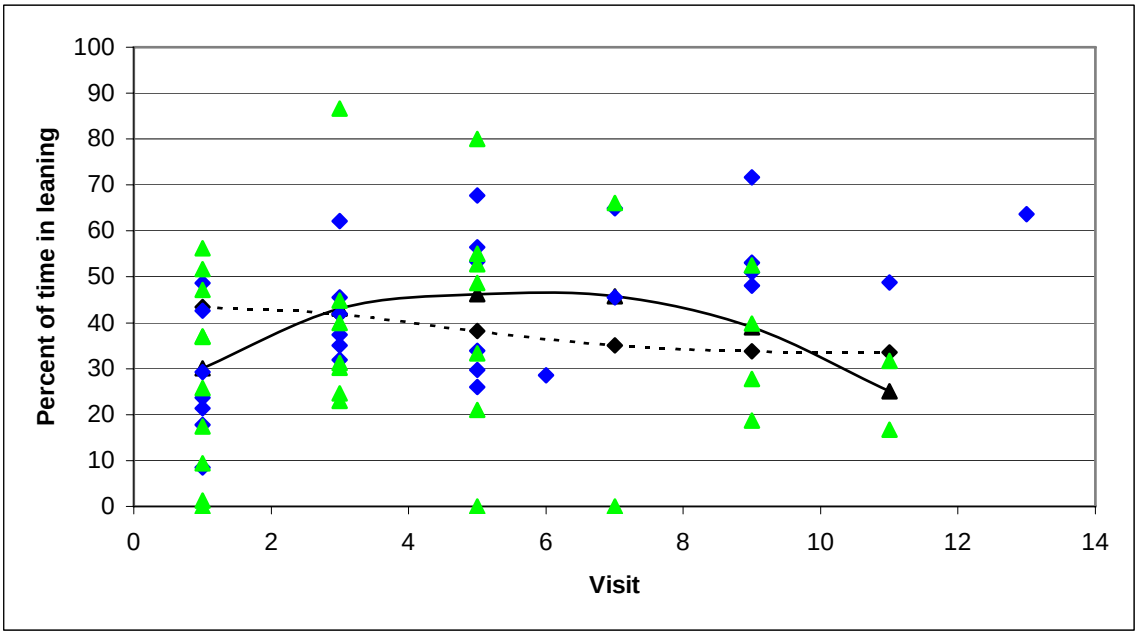
Predicted % time in leaning – Control Group

Predicted % time in leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.11. Percent of time in 1-hand support while in leaning



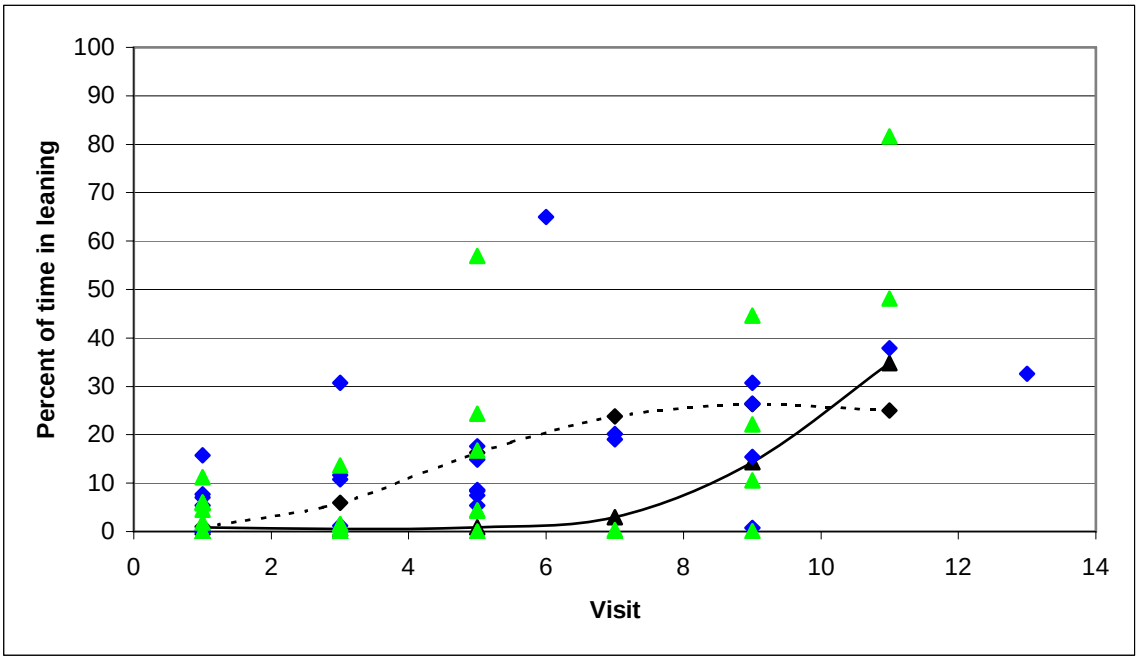
Predicted % time in leaning – Control Group

Predicted % time in leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Figure 3.12. Percent of time in 0-hand support while in leaning



Predicted % time in leaning – Control Group

Predicted % time in leaning – Experimental Group

Individual Scores Control Group

Individual Scores Experimental Group

Chapter 4

The effect of early orthotic use on the gait parameters of new walkers with Down syndrome

Introduction

Locomotion is an activity that is important for all children. When a child has a disability, such as Down syndrome (DS), walking becomes a primary concern of parents. The instinctive question, "When will my child walk?" is an important one because locomotion influences many aspects of development including social skill and cognitive development (Campos et al., 2000). In addition, age of walking onset may also influence the physical activity level of preschool aged children (Lloyd, 2008). The ability to walk well and explore while walking has a positive impact on development.

Development of gait

There are many factors that may contribute to the aquisition of walking. These factors, include the presence of the relevantneural substrates, enough strength to support the body on one foot, sufficient postural control, and level of motivation. There is evidence that children with DS display differing neural substrates (Brandt & Rosen, 1995) as well as decreased postural control (Haley, 1986; Kubo & Ulrich, 2006, Shumway-Cook & Woollacott, 1985). Children with DS also contend with ligamentous laxity and hypotonia (American Academy of Pediatrics, 2001). These factors contribute

to a delay in walking in children with DS that averages 12 months (Palisano et al., 2001; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001).

In addition to delayed onset, the gait of children with DS is atypical. Children with typical development (TD) learn to control their gait in 2 phases (Bril & Breniere, 1992). The first is a rapid phase in the first 4 months of walking. The second is a slower refining period that lasts up until 7 years of age. During the first phase, children with TD display a rapid decrease in step width, an increase in step length, and an increase in foot placement variability in the frontal and saggital plane (Bril & Breniere, 1993; Looper, Wu, Angulo Barroso, & Ulrich, 2006). As they learn to control their posture while moving forward, children with TD begin to improve joint coordination patterns beginning at the knees and hips (Chester, Tingley, & Biden, 2006). The muscular coordination pattern around the ankle joint takes longer to emerge (Ganley & Powers, 2005). These joint kinematics appear to develop relatively fast. However, the kinetics take longer. The hip and knee forces reach adult gait levels at roughly 7 years of age while the ankle forces reach adult levels at about age 9 (Chester et al., 2006).

Children with DS also show improvements in gait parameters; however, change more slowly and continue to show atypical gait patterns. (Cioni, Cocilovo, Rossi, Paci, & Valle, 2001; Smith, Kubo, Black, Holt, & Ulrich, 2007; Wu, Looper, Ulrich, Ulrich, & Angulo-Barroso, 2007). This may be related to decreased postural control in people with DS. In fact, Kubo and Ulrich (2006) found that the difference in gait between toddlers with DS and toddlers with TD were due to the difficulties that toddlers with DS have in gaining control of the movement of their center of mass in the frontal (mediolateral) plane. Decreased postural control appears to remain problematic during gait in a

preadolesents with DS as well (Kubo & Ulrich, 2006). In addition to poor postural control, adults with DS continue produce decreased ankle force compared to adults with TD (Cioni et al., 2001). These differences may be due to poor foot and ankle alignment caused by ligamentous laxity and hypotonia.

Foot and ankle alignment in gait

The ankle and foot play an important role in force production and absorption during gait. In adults who are typical, the foot and ankle alignment change during gait so that the foot and ankle complex can act as a shock absorber during initial contact and as a rigid lever arm for force transmission at terminal stance (Fukunaga et al., 2001). At initial contact, the ankle moves into pronation. This unlocks the navicular bone, the keystone of the medial longitudinal arch of the foot, allowing the arch of the foot to collapse and absorb energy. As the stance phase of gait continues with flexion during the loading response, the foot begins to move into supination. It reaches a neutral position at midstance and moves into slight supination by terminal stance. In supination, the navicular bone is locked in place to maintain a rigid arch and allow for effective force transfer through the foot into the ground for forward prolusion.

People with DS fall further into pronation than is typical, due to their low muscle tone and joint laxity (Genaze, 2000). They are less able to move out of pronation into supination. As a result, the foot is always in a shock absorbing, pronated position. The walker with DS is less able to transmit force through the foot to the ground and therefore requires greater force to gain the same forward motion as a walker with TD would (Cioni et al., 2001). In fact, Ulrich and colleagues (2004) found that preadolescents with DS required more forcing per step than preadolescents with TD.

Intervention and gait parameters

There is not much evidence on the effectiveness of specific interventions on quality of gait in children with DS. However, early treadmill training does impact gait in children with DS. Treadmill training alters the onset of walking in infants who have DS, lowering it by 101 days (Ulrich et al., 2001). It also results in an improvement in gait patterns at walking onset, especially in step length (Wu et al., 2007). Increasing the intensity of treadmill training leads to greater improvements in gait parameters and in the ability to clear an obstacle in the walking path that last for 6 months following the termination of training (Angulo-Barroso, Wu, & Ulrich, 2008; Wu, Ulrich, Looper, Tiernan, & Angulo-Barroso, 2008).

Another intervention that appears to improve gait in children with DS is orthoses at the foot and ankle complex. Orthoses that have been used in studies of children with DS include foot orthoses (FOs) (Selby-Silverstein, Hil lstrom, & Palisano, 2001) and supramalleolar orthoses (SMOs) (Martin, 2004).In children with DS, both FOs and SMOs have an effect on movement. Supramalleolar orthoses lead to improved balance in 3-8 year olds (Martin, 2004). Foot orthoses have an effect on gait in children with DS who are 3-6 years old by decreasing foot progression angle, changing initial contact from the forefoot to the heel, and increasing stance phase velocity (Selby-Silverstein et al., 2001). Though orthoses appear to have positive effects, they have not been tested in new walkers with DS. Despite this lack of evidence, orthoses continue to be a popular intervention for new walkers and pre-walkers with DS.

Orthoses work by holding the calcaneous in an upright position and limiting the amount of pronation the foot can achieve. The foot orthoses are worn on the bottom of

the foot and wrap around the side of the foot and the longitudinal arch and the lower portion of the calcaneous and are less restrictive than SMOs. SMOs wrap around the foot and ankle from the metatarsal heads to the malleoli. By controlling the calcaneal movement, to varying degrees, the orthoses attempt to limit the allowable motion into pronation and supination to an appropriate range around neutral.

In children with DS, both foot orthoses (Fos) and supramalleolar orthoses (SMOs) have an effect on movement. Supramalleolar orthoses lead to improved balance in 3-8 year olds (Martin, 2004). Foot orthoses have an effect on gait in children with DS who are 3-6 years old by decreasing foot progression angle, changing initial contact from the forefoot to the heel, and increasing stance phase velocity (Selby-Silverstein et al., 2001). Though orthoses appear to have positive effects, they have not been tested in new walkers with DS. Despite this lack of evidence, orthoses continue to be a popular intervention for new walkers and pre-walkers with DS

The rationale for prescribing orthoses for infants and toddlers with DS is that increased foot and ankle stability may lead to earlier walking onset. Although this is an important goal, orthotic use in this young population may also have adverse consequences. At this point in development, neuromuscular connections are forming in response to activity (Sporns & Edelman, 1993). This allows for the development of variable and adaptive movements. By limiting movement at the foot and ankle, orthoses may impede that process leading to decreased neuromuscular control. This would make movement while not wearing the orthoses difficult. In addition, the orthoses themselves may physically limit the children's ability to perform typical motor skills that require the ankle to attain a specific position (e.g. kneeling, creeping) due to their construction.

Health care professionals should carefully weigh the pros and cons of data on orthotic use in infants and toddlers before recommending their use.

Purpose

The purpose of this study is to determine whether the use of SMOs improves gait in new walkers with DS who have received treadmill training without orthoses and to determine if using orthoses prior to the onset of walking, during treadmill training, has positive effects on gait in children with DS.

Method

Infants were randomly assigned into a control (n=11) or experimental group (n=11). Children in the experimental group were measured for SMOs on their first visit and received them before their second visit. The children in the control group were measured for SMOs when they could take three independent steps and received them one to two weeks later. Children participated in this study from the time they could pull to stand until they had one month of independent walking experience. Once the infant could take three independent steps over ground, the treadmill training stopped. The Institutional Review Board at the University of Michigan approved this study.

Participants

Twenty-two children with DS enrolled in this study, however, 5 children dropped out of the study (1 had emerging medical problems, 1 did not tolerate the treadmill, and 3 received orthoses prior to the end of the study making them ineligible). Nine children (5

control and 4 experimental) walked well enough at one month of walking experience to participate in the gait analysis portion of this study. The study was limited to infants with Trisomy 21. Onset of participation occurred when infants could pull to stand at the furniture but not yet cruise. Children were excluded from this study if they had a history of other developmental disabilities, uncorrected visual or hearing impairment, previous orthotic use, orthotic use other than the ones provided for this study, or were unable to tolerate the orthoses. The toddlers had previously been randomly assigned into 2 groups: one in which parents were asked to provide treadmill training 8 minutes per day for 5 days a week for their infant (control) and one in which parents were asked to keep their infants in SMOs 8 hours a day in addition to the treadmill training (experimental). The infants in the experimental wore their shoes and orthoses during the treadmill training and the infants in the control group wore shoes during the treadmill training. The characteristics of each group at entry into the study are included in table 4.1. The children participated in the treadmill training from the time they pulled to stand independently until they took 3 independent steps. Children in the experimental group continued to wear SMOs after treadmill training was discontinued. Children in the control group were measured and fit with SMOs within a week after walking onset. They wore the SMOs for 8 hours a day. After one month of independent walking, each child came to the motor development laboratory for gait analysis. A parent of each child gave informed consent to participate in this study. The Health Sciences Institutional Review Board at the University of Michigan approved this study

Data Collection

All toddlers came to the Motor Development Laboratory at the University of Michigan one month after the onset of walking. .

At the beginning of the data collection period, the children had time to play and become comfortable in the setting. During this time, the experimental protocol was explained to the parents. Once the children were comfortable, the parents removed the children's clothes leaving the diaper on. An eyebrow pencil was used to mark the greater trochanter, the lateral joint line of the knee, and the posterior ankle midway between the malleoli. Retroreflective markers were placed on the skin surface at these locations. A marker was also placed on the toe section of the testing shoes over the second metatarsal head. The testing shoes were modified Nike Picos. A section was cut out of the heel counter to allow for marker placement. Velcro held this gap together. Shoe size was determined by the children's foot length measurement. Once the children had the shoes on, they were placed in standing on the walkway approximately 6-8 steps away from their parents. The parents encouraged the toddlers to walk toward them at the end of the mat. During data collection, each child walked a minimum of 4 steps across a walkway. This was repeated 4 times (4 trials) in each of 2 conditions (shoes only and shoes with SMOs). Order of conditions was random.

After the gait trials, anthropometric measures were taken. These included total height and weight, as well as leg length (ground to greater trochantor in standing), thigh length (lateral knee joint line to greater trochantor), thigh circumference (at the midpoint of the thigh, shank length (lateral malleolus to the lateral knee joint line), shank circumference (at the midpoint of the shank), ankle width (medial to lateral malleoli),

foot length (heel to toe), and foot width (at the widest point). Foot length and width were used to determine appropriate shoe size. Leg length was used to normalize step length. The other measurements were used to determine if there were physical differences between the groups.

Data Reduction and Analysis

The dependant variables in this analysis included: velocity, step width (horizontal distance between two consecutive foot falls), step length (distance between two consecutive foot falls in the direction of motion), dynamic base of support (angle formed by one stride and one step), and foot progression angle (the angle between the foot and the line of walking progression) (see figure 4.1). A 6-camera PEAK Motus motion capture system (Vicon Peak, Lake Forest, USA) was used to collect kinematic data at 60 Hz within a 2.6m (x) X 1.4m (y) X 1.2m (z) calibrated space. The dependent variables were calculated using a customized MATLAB program (The MathWorks, Natick, USA) using the PEAK data. The data for each variable were analyzed using a 2(group) X 2(condition) analysis of variance. Effect sizes were also calculated to help interpret group differences. An effect size of 0.2 through 0.49 was a small effect, 0.5-0.79 was a moderate effect, and 0.8 and above was a large effect (Cohen, 1988).

Results

Participant Characteristics

The participant characteristics are shown in table 4.1. At entry into the study, when the infants were able to pull stand, the children in the experimental group had a

mean age of 21.7 (±3.0) months and the control group had a mean of 16.7 (±4.1) months. At one month of walking experience, the toddlers in the experimental group were on average 31.0 (±6.3) months while the toddlers in the control group had a mean age of 25.8 (±4.2). The participants in the experimental group had been in the study for an average of 9.3 (±4.3) months and the participants in the control group had been in the study for an average of 9.1 (±2.0) months. There were no differences between the groups on the anthropometric measures.

Velocity

The mean velocity for each group and each condition are shown in figure 4.2. The average velocity for the control group was 0.29 (±0.13) meters per second. The average velocity for the experimental group was 0.19 (±0.07) meters per second (0.21 (±0.06) while wearing orthoses and 0.18 (±0.08) without orthoses). The ANOVA revealed a trend towards a group difference in velocity ($F(1,14)=3.47$ $p=0.08$), in favor of the control group. There was no significant difference in velocity between conditions ($F(1,14)=0.11$ $p=0.75$) and no significant group by condition interaction ($F(1,14)=0.03$ $p=0.87$). The group effect size was large, in favor of the control group (Cohen's D=1.00). This suggests that the average child in the control group walks at a velocity 1 standard deviation faster that the average child in the experimental group. The condition effect size was below meaningful(Cohen's D= 0.09)

Step Width

The step width normalized by hip width (obtained through PEAK) was 1.11 (±0.16) for the control group and 1.10 (±0.19) for the experimental group (see figure 4.3).

The ANOVA revealed no significant group difference for normalized step width ($F(1,14)=0.02$ $p=0.89$), no significant difference between the conditions ($F(1,14)=0.01$ $p=0.93$), and no significant group by condition interaction ($F(1,14)=0.05$ $p=0.83$). The calculated effect size statistic indicated there was no group effect (Cohen's D= 0.07) or condition effect (Cohen's D= 0.04).

Step Length

The average step length normalized by leg length was 0.49 (±0.14) for the control group and 0.40 (±0.12) for the experimental (see figure 4.4). The ANOVA revealed no significant group difference in normalized step length ($F(1,14)=1.97$ $p=0.18$), no significant difference between the conditions ($F(1,14)=0.83$ $p=0.38$), and no significant group by condition interaction ($F(1,14)=0.10$ $p=0.76$). The effect size statistic suggests there was a moderate effect for group in favor of the control group (Cohen's D= 0.70). There was also a small effect for condition in favor of the orthoses condition (Cohen's D=0.43).

Dynamic Base of Support

The average dynamic base of support for the control group was 126 (±13) degrees. The average dynamic base of support for the experimental group was 118 (±15) degrees (see figure 4.5). The ANOVA revealed no significant group difference ($F(1,14)=0.53$ $p=0.48$), no significant difference between the conditions ($F(1,14)=2.25$ $p=0.17$), and no significant group by condition interaction ($F(1,14)=0.02$ $p=0.90$). There was a moderate effect for group in favor of the experimental group (Cohen's D= 0.57).

There was also a large effect for condition in favor of the shoes condition (Cohen's D=0.93).

Foot Progression Angle

The average foot progress angle for the control group was 21 (±5) degrees. The average foot progression angle for the experimental group was 11 (±7) degrees (see figure 4.6). The ANOVA revealed a significant group difference in favor of the experimental group ($F(1,14)=10.33$ $p=0.01$), no significant difference between the conditions ($F(1,14)=0.19$ $p=0.63$), and no significant group by condition interaction ($F(1,14)=0.61$ $p=0.45$). There was a large effect for group in favor of the experimental group (Cohen's D= 1.67). There was little condition effect (Cohen's D= 0.13).

Discussion

This study provides a glimpse of both the short-term and long-term outcomes of SMO use in new walkers with DS. The toddlers were tested as they walked with and without orthoses to provide insight into the short-term effects of orthoses. The long-term effects of SMO use in infancy and during treadmill training are observed in the group differences. The experimental group received treadmill training and wore SMOs throughout the study while the control group received orthoses after they could take 3 independent steps. By looking at gait parameters both with and without SMOs in all children, we can determine whether the SMOs lead to a short-term improvement in gait patterns. In addition, we can determine if long-term SMO use in infancy improves gait parameters at 1 month post walking onset.

The results of this study suggest that short-term SMO use leads to impaired dynamic stability. Though the dynamic base of support measure was not statistically different by condition, the large effect size in favor of the shoes only condition indicate that dynamic stability was negatively influenced by orthotic use in this sample. Stabilizing the foot and ankle does not seem to immediately lead to a more stable gait in new walkers.

The long-term use of SMOs in infancy could have positive and negative effects. While SMOs provide foot and ankle stability, they limit the activity around the ankle. This limited activity could lead to delayed or abnormal neuromuscular development in infants (Sporns & Edelman, 1993) and could cause movement problems such as difficulty maintaining dynamic balance. This does not seem to be the case in older children with DS. Children between the ages of 3 and 8 with DS who wore SMOs for a 6-week period displayed improved balance skills (Martin, 2004). Again, the long-term effects of SMO use in infancy have not been previously studied.

The results of this study suggest that long-term orthotic use does affect the gait parameters of new walkers with DS. Infants who did not wear SMOs during treadmill training walked faster than those who wore SMOs. However, children in the SMO group walked close to the speed of the treadmill training (treadmill speed =0.2 meters per second) while the control group walked faster than that speed. Faster velocity is usually described as better because velocity increases with practice. It may be that toddlers who walked faster had less control of their walking while the toddlers in the experimental group walked at a more controlled speed, that is, close to their training speed. Bril and Breniere (Bril & Breniere, 1993) state that, for new walkers, walking is a process of

falling followed by regaining balance during double limb support. Perhaps the toddlers in the control group did not have the control to regain their balance in double support and were instead in a perpetual state of falling. On the other hand, the faster velocity and the moderate effect size for step length may indicate a positive effect for the control group. Perhaps the prolonged orthotic use in the experimental group imposed some movement restrictions in the saggital plane, which limited step length and velocity at one month of walking experience.

Increased control and stability for the experimental group is suggested by foot progression angle. The amount that the foot turns outward during stance is an indication of stability (Orner, Turner, & Worrell, 1994). As the angle of outward rotation increases, the child's base of support increases, indicating that gait is less stable and a larger base of support is needed to maintain control. The children in the experimental group had a significantly smaller foot progression angle than the children in the control group, suggesting that they were more stable while walking.

Further support for the increased stability of the experimental group is their dynamic base of support. Although there was no significant group difference in this measure, there was a moderate effect size, which, in light of the low statistical power, also suggests that the experimental group displayed more dynamic stability than the control group. This effect size could also be a random effect and the results concerning dynamic base of support should be interpreted cautiously. However, while there was no statistical difference between the groups in step width, the foot progression angle was significantly larger in the control group; the children in the control group have a larger base of support. This suggests that the children in the control group are not as

dynamically stable as the children in the experimental group and require a larger base of support to maintain an upright position. This lends weight to the interpretation of the moderate group effect size in dynamic base of support as a true effect rather than a random effect. It appears that the children in the experimental group, who wore the orthoses for a long period of time (on average 9 months), had a more stable gait pattern when they started walking independently.

While long-term orthotic use does lead to improved gait at 1 month of walking experience, the improvements were small. In addition, gait in children with TD is unstable at 1 month of walking experience (Bril & Breniere, 1993). The cost of orthoses in dollars and time is not negligible. The question of whether the small improvements seen justify this cost at a time when gait is inherently unstable is a valid one. Longer term follow up will help determine whether these improvements are clinically relevant or if similar or better results will arise from delaying orthotic use until after the gait patterns in new walkers has begun to stabilize.

Conclusion

Supramalleolar orthoses appear to impair gait stability in new walkers after short-term use. The results seem to improve with long-term use during infancy. Though these results are promising, they should be interpreted with caution due to the potential impact orthoses may have on the development of control for other behaviors. Future studies exploring the effect of long-term orthotic use on other aspects of development are necessary to give us a more complete picture of the impact of orthoses on neuromotor development. In addition, this study had a small sample size. The small sample size and the large variability in many of the variables lead to low statistical power. Future studies

should have a larger sample size. Future studies should also include a SMO only group in addition to the treadmill training only and treadmill training and SMO groups. This will help differentiate how much impact the treadmill training has on the development of postural control in infants with DS compared to SMOs.

References

American Academy of Pediatrics Council on Genetics, (2001). American Academy of Pediatrics: Health supervision for children with Down syndrome. *Pediatrics, 107*(2), 442-449.

Angulo-Barroso, R. M., Wu, J., & Ulrich, D. A. (2008). Long-term effect of different treadmill interventions on gait development in new walkers with Down syndrome. *Gait & Posture, 27*(2), 231-238.

Brandt BR, Rosen I (1995) Impaired peripheral somatosensory function in children with Down syndrome. *Neuropediatrics,* 26, 310–312

Bril, B., & Breniere, Y. (1992). Postural requirements and progresion velocity in young walkers. *Journal of Motor Behavior, 24*(1), 105-116.

Bril, B., & Breniere, Y. (1993). Posture and independent locomotion in early childhood: Learning to walk or learning dynamic postural control? In G. J. P. Savelsbergh (Ed.), *The development of coordination in infancy* (Vol. 97, pp. 337-358). Amsterdam: Elsevier Science.

Campos, J. J., Anderson, D. I., Barbo-Roth, M. A., M., H. E., Hertenstein, M. J., & Witherington, D. C. (2000). Travel broadens the mind. *Infancy, 1*, 149-219.

Chester, V. L., Tingley, M., & Biden, E. N. (2006). A comparison of kinetic gait parameters for 3-13 year olds. *Clinical Biomechanics, 21*, 726-732.

Cioni, M., Cocilovo, A., Rossi, F., Paci, D., & Valle, M. S. (2001). Analysis of ankle kinetics during walking in individuals with Down syndrome. *Americal Journal of Mental Retardation, 106*(5), 470-478.

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences* (Second Edition ed.). Hillsdale: Lawrence Erlbaun Associates.

Fukunaga, T., Kubo, K., Kawakami, Y., Fukashiro, S., Kanehisa, H., & Maganaris, C. N. (2001, February 7, 2001). In vivo behaviour of human muscle tendon during walking. *Proceedings: Biological Science,* 268(1464),229-233.

Ganley, K. J., & Powers, C. M. (2005). Gait kinematics and kinetics of 7-year-old children: a comparison to adults using age-specific anthropometric data. *Gait & Posture, 21*(2), 141-145.

Genaze, R. R. (2000). Pronation. The orthotist's view. *Clinics in Podiatric Medicine & Surgery, 17*(3), 481-503.

Haley, S. M. (1986). Postural reactions in infants with Down syndrome. *Physical Therapy, 66*(1), 17-22.

Kubo, M. & Ulrich, B. D. (2006). Coordination of pelvis-HAT (head, arms, and trunk) in anterior-posterior and medio-lateral diractions during treadmill gait in preadolescents with/without Down syndrome. *Gait & Posture, 23*, 512-518.

Kubo, M., & Ulrich, B. D. (2006). Early Stage of Walking: Development of Control in Mediolateral and Anteroposterior Directions. *Journal of Motor Behavior, 38*(3), 229-237.

Lloyd, M. C. (2008). *Physical Activity of Preschool Age Children With and Without Down Syndrome*. Unpublished Dissertation. Univeristy of Michigan, Ann Arbor.

Looper, J., Wu, J., Angulo Barroso, R., & Ulrich, D. U., B. (2006). Changes in step variability of new walkers with typical development and with Down syndrome. *Journal of Motor Behavior, 38*(5), 367-372.

Martin, K. (2004). Effects of supramalleolar orthoses on postural stability in children with Down syndrome. *Developmental Medicine & Child Neurology, 46*(6), 406-411.

Orner, C. E., Turner, D., & Worrell, T. (1994). Effect of foot orthoses on the balance skills of a child with a learning disablility. *Pediatric Physical Therapy, 6*, 10-14.

Palisano, R. J., Walter, S. D., Russell, D. J., Rosebaum, P. L., Gemus, M., Galuppi, B. E., et al. (2001). Gross Motor Function of Children with Down Syndrome: Creation of Motor Growth Curves. *Archives of Physical Medicine & Rehabilitation*, 82, 494-500.

Selby-Silverstein, L., Hillstrom, H. J., & Palisano, R. J. (2001). The effect of foot orthoses on standing foot posture and gait of young children with Down syndrome. *Neurorehabilitation, 16*(3), 183-193.

Shumway-Cook, A., & Woollacott, M. H. (1985). Dynamics of postural control in the child with Down syndrome. *Physical Therapy, 65*(9), 1315-1322.

Smith, B. A., Kubo, M., Black, D. P., Holt, K. G., & Ulrich, B. D. (2007). Effect of practice on a novel task--walking on a treadmill: preadolescents with and without Down syndrome. *Physical Therapy, 87*(6), 766-777.

Sporns, O., & Edelman, G. M. (1993). Solving Bernstein's problem: a proposal for the development of coordinated movement by selection. *Child Development, 64*(4), 960-981.

Ulrich, B. D., Haehl, V., Buzzi, U. H., Kubo, M., & Holt, K. G. (2004). Modeling dynamic resource utilization in populations with unique constraints:

preadolescents with and without Down syndrome. *Human Movement Science, 23*(2), 133-156.

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill training of infants with Down syndrome: evidence-based developmental outcomes. *Pediatrics, 108*(5), E84.

Wu, J., Looper, J., Ulrich, B. D., Ulrich, D. A., & Angulo-Barroso, R. M. (2007). Exploring effects of different treadmill interventions on walking onset and gait patterns in infants with Down syndrome. *Developmental Medicine & Child Neurology, 49*(11), 839-945.

Wu, J., Ulrich, D. A., Looper, J., Tiernan, C. W., & Angulo-Barroso, R. M. (2008). Strategy adoption and locomotor adjustment in obstacle clearance of newly walking toddlers with down syndrome after different treadmill interventions. *Experimental Brain Research,* 186(2), 261-272.

Tables

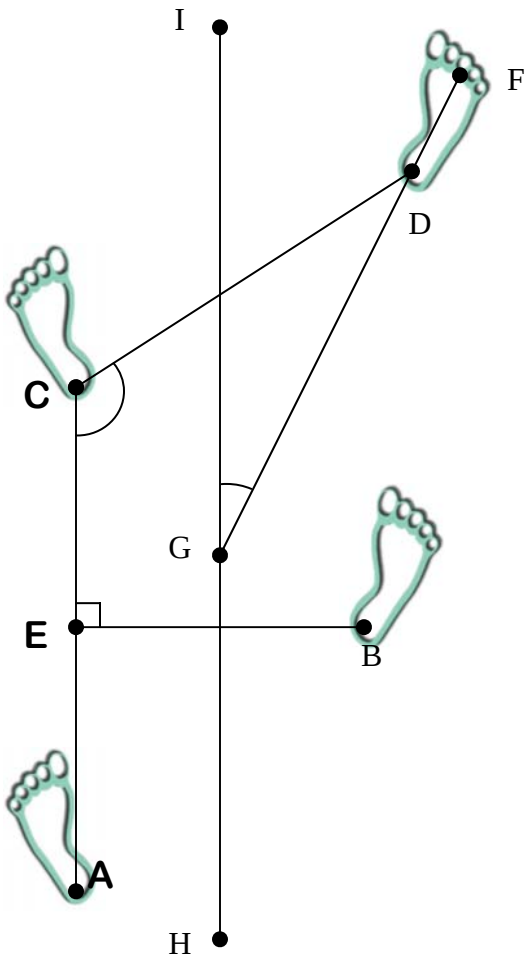
Table 4.1. Participant Characteristics at 1 Month of Walking Experience

	Total Sample	Experimental	Control	p-value
Total Height (cm)	79.6 (5.9)	82.4 (3.0)	77.4 (6.9)	0.23
Weight (lbs)	23.7 (2.6)	25.2 (1.1)	22.5 (3)	0.13
Leg Length (cm)	34.1 (3.8)	35.3 (1.6)	33.1 (5.0)	0.44
Thigh Length (cm)	16.6 (2.1)	17.9 (1.3)	15.6 (2.2)	0.11
Thigh Circumference (cm)	27.0 (1.5)	27.1 (2.2)	27.0 (0.8)	0.88
Shank Length (cm)	15.5 (1.3)	16.0 (1.1)	15.1 (1.5)	0.39
Shank Circumference (cm)	19.0 (1.2)	19.1 (1.8)	18.9 (0.6)	0.82
Ankle Width	3.9 (0.2)	3.8 (0.2)	4.0 (0.2)	0.21
Foot Length	12.0 (0.8)	12.3 (0.5)	11.8 (1.0)	0.39
Foot Width	5.1 (0.4)	5.1 (0.5)	5.1 (0.4)	0.76
Age at Study Entry (m)	18.9 (4.3)	21.7 (3.0)	16.7 (4.1)	0.08
Age at New Walker Visit (m)	28.1 (5.6)	31.0 (6.3)	25.8 (4.2)	0.18
Time in Study (m)	9.2 (3.0)	9.3 (4.3)	9.1 (2.0)	0.94

Mean (SD)

Figures

Figure 4.1. Gait Parameters



Step Length – line AE

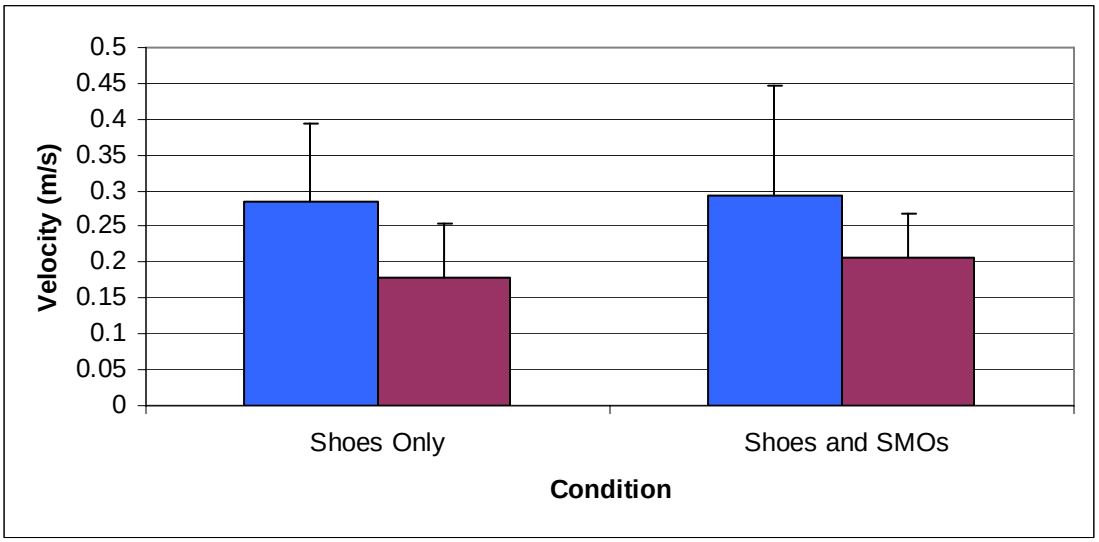
StepWidth – line BE

Dynamic Base of Support – angle ACD

Angle of Foot Progression – angle FGI

Line of Walking Progression – line HI

Figure 4.2. Velocity by Condition



Control Group

Experimental Group

Figure 4.3. Normalized Step Width by Condition

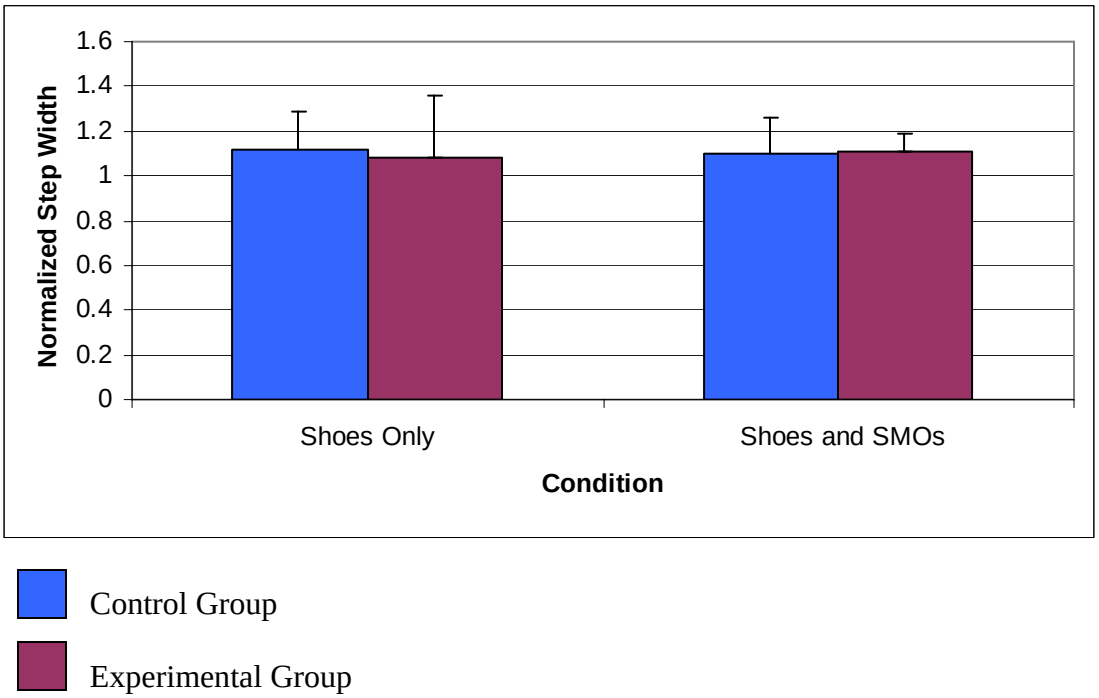
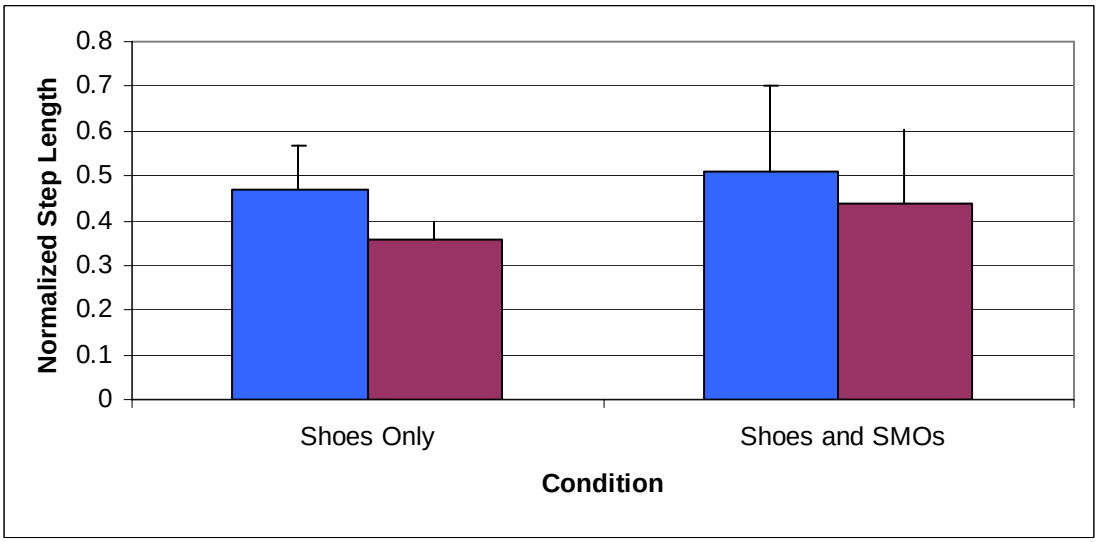


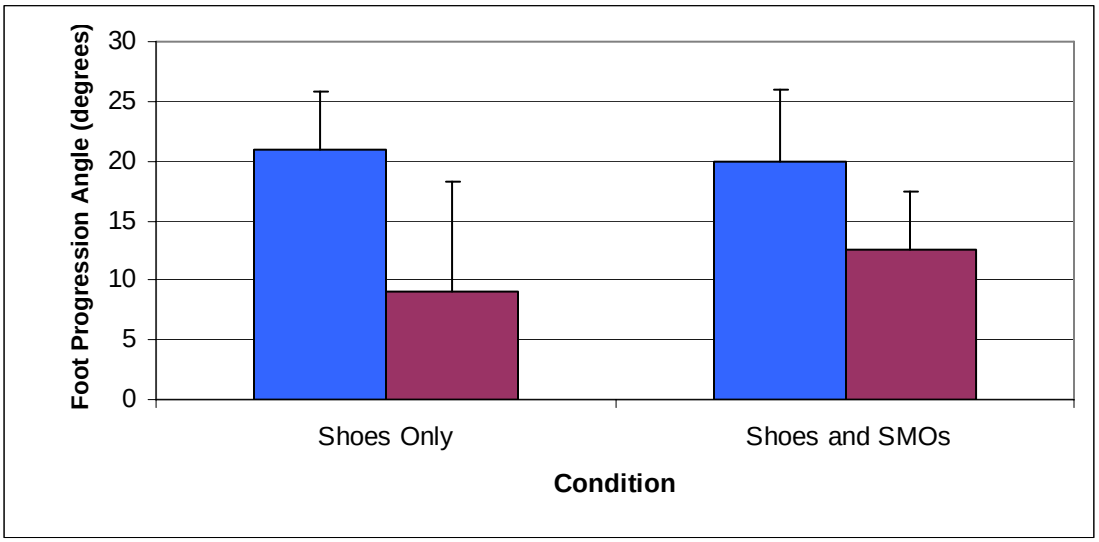
Figure 4.4. Normalized Step Length by Condition



Control Group

Experimental Group

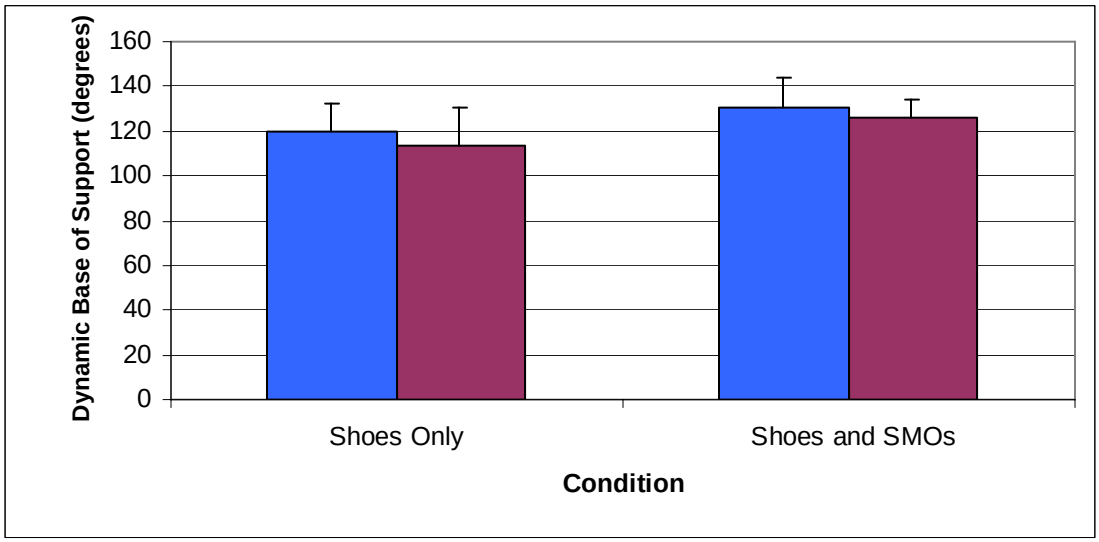
Figure 4.5. Foot Progression Angle by Condition



Control Group

Experimental Group

Figure 4.6. Dynamic Base of Support Condition



Control Group

Experimental Group

Chapter 5

Conclusion

Introduction

The purpose of this intervention study was to determine whether activity and participation level skill development improved by combining treadmill training with orthotic use in infants with Down syndrome (DS). In addition, I sought to gain a further understanding of the specific results of supramalleolar orthoses (SMO) use in infants with DS including the impact on age at walking onset, play performance, and walking ability. This information is an important addition to the growing body of knowledge of specific early interventions for children with DS. It will offer health care providers and parents information about appropriate treatment options for infants with DS.

I predicted specific study outcomes based on: (1) previous studies involving orthotic use, (2) previous research using treadmill training with infants who have DS, (3) theoretical tenants of dynamic systems theory, and (4) components of the ICF model. I expected the infants who wore orthoses while learning to walk would walk earlier than the infants who did not wear orthoses due to the increased stability around their foot and ankle. I also believed that the increased stability provided by the orthoses would allow the infants who wore orthoses to spend more time using their hands for play during a 20-minute play task than those without orthoses. Finally, I expected that new walkers would display better gait while wearing orthoses compared to their gait while not wearing

orthoses. Overall, I thought that SMO use would benefit children with DS who were learning to walk.

Summary of Results

Study 1

The first aspect of this study examined the trajectory of motor skill development in infants who received treadmill training with or without orthoses. Because postural control is a limiting factor in the development of children with DS (Haley, 1986) and SMO use leads to improved balance in older children with DS (Martin, 2004), I thought that SMO use in infants with DS would improve their developmental trajectory. I predicted that ankle stability was the control parameter for the development of upright motor skills in this population.

In general, the motor skill development of infants with DS did not benefit from SMO use. Though there was a moderate treatment effect in favor of the experimental group in time from study entry to independent walking, the raw developmental test scores showed better overall performance in the infants who did not wear orthoses. It appears that the increased stability around the foot and ankle may have positively influenced the ability to take independent steps, however, it had a negative impact on other motor skills and the adaptability of walking skills.

In this first study, the skill targeted by the treadmill intervention was walking. In previous studies, the consistent early practice of alternating stepping provided by the treadmill led to earlier onset of walking (Ulrich, Lloyd, Tiernan, Looper, & Angulo-Barroso, 2008; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001). In contrast to the current study, the children in these earlier studies began treadmill training at approximately 10

months corrected age, when they could sit independently or take at least 6 steps on the treadmill. Due to the emphasis on orthotic use, the children in the current study began the treadmill intervention when they could pull to stand independently. It appears that waiting for "walking readiness," as indicated by pulling to stand, is too late to begin the treadmill training intervention. The children in previous studies who received earlier treadmill intervention walked at approximately 20 months. In the current study, the average age at study entry (pull to stand) was 20.5 months. This suggests that in order to receive the maximum benefit from the treadmill intervention, therapists must begin its implementation before infants display walking readiness; waiting until they can pull to stand is to late.

Study 2

The second aspect of this study focused on the effect of SMOs on upright play ability in infants with DS. Again, I predicted that ankle stability was a control parameter for hand use during upright play in this population. Young children with DS who are 1-3 year olds with DS display uncoordinated upright postural control (Shumway-Cook & Woollacott, 1985). In addition, orthoses lead to improvement in balance related subscales of developmental tests in 3-8 year olds with DS (Martin, 2004). With this in mind, I predicted that SMO use in infants with DS would improve their balance enough for them to free up their hands and engage in exploratory play.

As with the motor development skills, the infants with DS who wore SMOs did not perform as well as the infants who did not wear SMOs in exploratory hand use when playing in upright. However, the infants who wore the orthoses spent more time in a non-leaning posture than the infants who did not wear orthoses. The infants who wore

SMOs displayed better trunk control but focused on this aspect of their movement while in upright instead of exploratory play. The infants with DS who did not use orthoses were able to use their hands more adaptively for play and exploration though they were leaning to perform this task. The increased stability around the foot and ankle provided by the SMOs negatively influenced the infants' ability to use their hands to play but allowed for improved postural control.

This second study centered on infants' ability to use their hand for play and exploration while in an upright position. Previous research has shown that children with DS participate in exploratory play but are not as good as children with TD at picking out the salient features of an object to explore (Loveland, 1987). I thought that providing the infants with a boost of stability might help facilitate their exploratory behavior. Instead, it had the opposite affect. We know that the interplay between upper and lower body is important in new walkers with DS (Kubo & Ulrich, 2006) and that the trunk serves as an interface between the upper and lower extremities in children who are cruising (Haehl, Vardaxis, & Ulrich, 2000). Perhaps the orthoses boosted the infants' balance enough to free up the trunk but the infants had not yet developed adequate trunk control to use the trunk as a link between the arms and legs. This resulted in a non-leaning posture with 2 hands on the support surface and decrease exploratory hand use. The orthoses may lead the infants to rely on their upper extremities for body weight support instead of using them for exploratory play.

Study 3

The third aspect of this study examined the gait, with and without orthotic use, of new walkers with DS who received treadmill training. The experimental group learned to

walk while wearing the orthoses and the control group received the orthoses after walking onset. Previous research suggests that treadmill use alone positively impacts gait in children with DS (Angulo-Barroso, Wu, & Ulrich, 2008; Wu, Looper, Ulrich, Ulrich, & Angulo-Barroso, 2007; Wu, Ulrich, Looper, Tiernan, & Angulo-Barroso, 2008). In addition, foot orthoses have a positive impact on gait parameters in 3-6 year old children with DS (Selby-Silverstein, Hillstrom, & Palisano, 2001). Based on this information, I hypothesized that SMO use would improve gait performance in new walkers with DS compared to gait with just shoes on (immediate effects) and that those children in the experimental group,who learned to walk while wearing orthoses, would display more typical gait parameters when compared to those children who did not wear orthoses while learning to walk (long-term effects).

The results of this study provide insight into both short-term and long-term SMO use. The results suggest that there is little effect of immediate SMO use on the measured gait parameters. There were no significant differences or effect sizes for velocity, step width, or foot progression angle. There was only a small effect size in favor of the orthoses condition for step length. For dynamic base of support, there was actually a large effect in favor of the shoes only condition. When comparing the use of SMOs during gait to walking with shoes only in new walkers with DS, orthoses seem to have a negative effect on dynamic stability.

Interestingly, long term SMO use leads to improved dynamic stability. New walkers in the experimental group appeared to be more stable than new walkers in the control group. There was a moderate effect size in favor of the experimental group in dynamic base of support. Though the toddlers in the experimental group walked slower

than the toddlers in the control group, they walked closer to the treadmill training speed (0.2 m/s). In addition, the toddlers in the experimental group had a much smaller foot progression angle and showed signs of improved dynamic base of support. This suggests that the toddlers in the experimental group had more control over their gait.

The use of orthoses from the time infants can pull to stand appears to improve gait parameters at walking onset. Though gait was slower in toddlers from the experimental group, the decreased foot progression angle and dynamic base of support suggest that this was due to increased stability and control. Perhaps the toddlers in the control group were relying on momentum to maintain upright and walk to the end of the mat. This would lead to a higher velocity but less control. In addition, the increased foot progression angle in the toddlers who were in the control group suggests that they require a wider base of support to maintain upright. In other words, they are less stable than the toddlers who trained with SMOs. This suggestion is further supported by the moderate effect size in favor of the experimental group on dynamic base of support. The long-term use of SMO in infants leads to a more dynamically stable gait pattern.

Overall Conclusions

While I hypothesized that SMO use would benefit children with DS who were learning to walk, the evidence collected in this study suggests that, overall, the results are mixed. The use of SMOs limited the ability of infants with DS to effectively learn how to adapt their movements. The infants who wore orthoses while learning to walk displayed poorer motor skill development and showed less independence in their mobility skills than infants who did not use orthoses. In addition, the infants who wore orthoses also had a more difficult time using their hands for play in an upright position than the

infants who did not wear orthoses. However, the use of orthoses did appear to improve upright balance during play and led to better dynamic stability during gait. Though these results matched the previous, though limited, research on orthotic use in children with DS that portrayed orthotic use in a positive light (Martin, 2004; Selby-Silverstein et al., 2001), the potentially negative effect of orthoses on non-gait aspects of development was surprising.

A primary difference between this study and previous studies of orthotic use in children with DS is the developmental level of the children who participated. The previous studies tested children whose walking skills were already established. This study, however, looked at the impact of orthotic use on infants with DS who were acquiring upright motor skills. In Bernstein's (1967) theory of skill acquisition, movers have to learn how to control movement by establishing flexible movement synergies. This is done by first freezing degrees of freedom and then, as movement control is learned, freeing them. Perhaps the orthoses interfere with this process of skill acquisition. The SMOs provide an external device that freezes the degrees of freedom but does not allow the infant to gain control of the movement at the foot and ankle or free the degrees of freedom at the foot and ankle. This limits the control over and adaptability of the movement that develops.

Though orthotic use may be helpful in older children with DS, I suggest that SMOs should not be used in infants with DS during skill acquisition. To facilitate the onset of walking skills and improve gait parameters in infants with DS, early treadmill training is effective. While the use of SMOs improved gait parameters, the use of SMOs at this stage is not beneficial and should be postponed until walking is established

because SMO use may have a negative effect on overall developmental skills. As mentioned earlier, this information is an important addition to the growing body of knowledge on early interventions for children with DS and provides healthcare providers with increased knowledge on how to effectively use orthoses as an intervention technique in children with DS.

Limitations

Though I am confident in the results of this study, there are limitations to consider when applying the conclusions to the population of infants with DS at large. This study compared two groups of children who both received treadmill training. The addition of a group that received SMOs only would allow us to distinguish between the effects of the treadmill and the effects of the SMO more clearly. Another limitation of this study is the type of orthoses used. Supramalleolar orthoses are more restrictive than other types of foot orthoses. Given the large variability in the population with DS, they may have been appropriate for some children but were clearly too restrictive for most of the children in this study. A less restrictive orthotic may provide better results, however, the external limiting of degrees of freedom during skill acquisition, at any level, would theoretically be detrimental to development. It is also important to note that this study differed from previous treadmill training studies in that the infants began treadmill training at a later developmental point. Future studies should begin treadmill training earlier and add orthoses as appropriate. One final limitation of this study is the sample size. Though 22 participants were recruited, 5 dropped out during the treadmill training portion of the study and only 9 were included in the gait portion of the study. A larger sample would be beneficial.

Future Research Directions

Orthotic use in children with DS is a controversial subject in the health care community. There is little published research and many conflicting theories about why orthoses are good or bad. In response, many practitioners rely on experience and anecdotal evidence to make decisions on the use of orthoses. More research will help inform clinical practice in this area. Of primary concern is when is the most appropriate time, if any, to begin orthotic intervention. Also of concern is what are the most appropriate type of orthoses for a child with DS and what factors can a health care professional use to make that decision. Another important set of questions is what, other than gait kinematics, do orthoses effect. For instance, what is the effect of orthotic use on gait kinetics, energy expenditure, and physical activity level in children with DS? Though these questions are just a start, answering them will greatly enhance evidence that health care providers can use to make decisions about orthotic use in children with DS.

References

Angulo-Barroso, R. M., Wu, J., & Ulrich, D. A. (2008). Long-term effect of different treadmill interventions on gait development in new walkers with Down syndrome. *Gait & Posture, 27*(2), 231-238.

Bernstein, N. (1967). *The co-ordination and regulationof movements*. Oxford: Pergamon Press.

Haehl, V., Vardaxis, V., & Ulrich, B. D. (2000). Learning to cruise: Bernstein's theory applied to skill acquisition during infancy. *Human Movement Science, 19*(5), 685-715.

Haley, S. M. (1986). Postural reactions in infants with Down syndrome. *Physical Therapy, 66*(1), 17-22.

Kubo, M., & Ulrich, B. D. (2006). Early stage of walking: development of control in mediolateral and anteroposterior directions. *Journal of Motor Behavior, 38*(3), 229-237.

Loveland, K. A. (1987). Behavior of young children with Down syndrome before the mirror: Exploration. *Child Development, 58*(3), 768-778.

Martin, K. (2004). Effects of supramalleolar orthoses on postural stability in children with Down syndrome. *Developmental Medicine & Child Neurology, 46*(6), 406-411.

Selby-Silverstein, L., Hillstrom, H. J., & Palisano, R. J. (2001). The effect of foot orthoses on standing foot posture and gait of young children with Down syndrome. *Neurorehabilitation, 16*(3), 183-193.

Shumway-Cook, A., & Woollacott, M. H. (1985). The growth of stability: postural control from a development perspective. *Journal of Motor Behavior, 17*(2), 131-147.

Ulrich, D. A., Lloyd, M. C., Tiernan, C. W., Looper, J. E., & Angulo-Barroso, R. M. (2008). Effects of Intensity of Treadmill Training on Developmental Outcomes and Stepping in Infants With Down Syndrome: A Randomized Trial. *Physical Therapy, 88*(1).

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill training of infants with Down syndrome: evidence-based developmental outcomes. *Pediatrics, 108*(5), E84.

Wu, J., Looper, J., Ulrich, B. D., Ulrich, D. A., & Angulo-Barroso, R. M. (2007). Exploring effects of different treadmill interventions on walking onset and gait

patterns in infants with Down syndrome. *Developmental Medicine and Child Neurology*, *49*(11), 839-945.

Wu, J., Ulrich, D. A., Looper, J., Tiernan, C. W., & Angulo-Barroso, R. M. (2008). Strategy adoption and locomotor adjustment in obstacle clearance of newly walking toddlers with down syndrome after different treadmill interventions. *Experimental Brain Research*, 186(2), 261-272.

Appendices

Appendix 1. Orthotic use in children with DS: A theoretical perspective

Introduction

Orthotic use in infants with Down syndrome (DS) who cannot yet walk is a controversial subject. With little published research about the effects of orthoses on the motor development in infants with DS, clinicians often rely on anecdotal evidence and opinion to decide whether orthoses are appropriate. Ideally, the clinician's theoretical background influences decisions such as when to begin orthotic use, how to measure the impact of orthotic use, and whether the use of orthoses is harmful at any time in development. A theoretical background in Dynamic Systems Theory (DST) can provide clinicians and scientists with insight into these questions and may help define the most beneficial period to use orthotic therapy on children with DS

Dynamic Systems Theory (DST), as it relates to development, focuses on the contributions of many subsystems to advances in motor skills. These subsystems include systems of the body, the environment, as well as the constraints of the task (Newell, 1986). As these subsystems reach a threshold where they can successfully interact and facilitate movement, a behavior emerges.

A principle component of DST is self-organization. Self-organization of movement occurs spontaneously. Self-organized movement patterns are not preprogrammed in the brain but emerge based on function and the interaction of

subsystems (Heriza, 1991; Thelen & Corbetta, 1994; Ulrich, 1997). Self-organization occurs in response to the task at hand, the configuration of the body, and the context that the task is performed in (Thelen, 1992). Clinicians can influence the behavior by manipulating aspects of the task, the body, or the environment in which it is occurring.

By manipulation aspects of the task, body, or environment, clinicians are influencing control parameters. A control parameter causes a system to shift into a new behavior. In the case of human motor development, control parameters cause children to change their pattern of motor behavior. The developmental state of a subsystem, the environmental context, or even a specific aspect of the task can all act as control parameters. In order for therapists to effectively facilitate development and change behavior, it is critical to identify the control parameters of the behaviors in focus.

In addition to control parameters and self-organization, the concept of "degrees of freedom" is often employed in DST. In pioneering work, Bernstein (1967) recognized that the body is complex and that there are multiple ways to accomplish a single task. This redundancy in the system led him to conclude that there are too many variables to separately control when learning a task. Bernstein termed this the "degrees of freedom problem" (Bernstein, 1967). To gain control of the many available degrees of freedom, they are compressed into movement patterns and the immense number of possible movement combinations becomes limited (Heriza, 1991; Scholz, 1990). Infants figure out how to compress and limit the available degrees of freedom through active exploration of movement (Thelen & Corbetta, 1994). Early active movement is vital to an infant's ability to develop appropriate movement patterns.

Orthotic Use In Light Of Dynamic Systems Theory

When applying a DST perspective to treatment, clinicians attempt to manipulate one or more control parameters that will lead to the emergence of the desired behavior. Often this control parameter is the development of postural control. Children with DS have delayed development of postural control (Haley, 1986). Children (especially infants) with DS display low muscle tone and ligamentous laxity leading to joint laxity (American Academy of Pediatrics, 2001). Many people believe that this joint laxity limits motor development in children with DS. Clinicians often use orthoses to limit the joint laxity at the foot and ankle. In other words, the orthoses constrain the degrees of freedom at the foot and ankle. In infants, this manipulation of the musculoskeletal subsystem may produce more foot and ankle stability and could allow a shift in behavior from cruising to walking.

However, externally limiting the freedom of movement at the foot and ankle with orthoses could reduce the active exploration of the movement around this joint complex. Early voluntary exploration is important to the development of the neuromuscular system. In fact, perception and action leads neurons to form coherent groups (Sporns & Edelman, 1993). Based on principles of activity-dependent neuroplasticity, if activity is limited, the formation of neural connections as well as the firing patterns of established connections will be disrupted (Merzenich et al., 1996). As Thelen & Corbetta stated, there are neural consequences to behavioral exploration (Thelen & Corbetta, 1994). To facilitate skill aquisition, there is a period of intense exploration when an infant begins to learn a new movement (Thelen & Corbetta, 1994). This leads to a “soft-wired” neural system that can adapt to changes in context. A lack of early practice and exploration

could lead to decreased movement adaptability. If motion is externally limited during the formation of neuromuscular networks, infants may not develop the variation in movement necessary to adapt to changing contexts.

Orthotic use could be beneficial to children by limiting the degrees of freedom around the foot and ankle, leading to a shift in behavior. Conversely, they could limit the variation in movement practice and lead to a loss of adaptability. Perhaps both of these points are correct. If this is the case, we should explore the impact of orthoses over developmental time. The experience of moving gives us insight into the world around us, and how our bodies move through this world. In fact, many developmental researchers suggest that experimentally grouping infants' level of experience as opposed to age may provide us with better insight into the process of development (Adolph, Vereijken, & Shrout, 2003; Spencer & Thelen, 2000; Yaguramaki & Kimura, 2002). The effect of experience on muscular activation is task specific (Okamoto, Okamoto, & Andrew, 2003). In addition, the locomotor system reacts to a dynamic force environment (Thelen, 1992). If external devices such as orthoses limit foot and ankle movement while learning a new task, the typical dynamic force environment is altered and appropriate muscular activation patterns may not develop. In this case, use of orthoses during skill acquisition would result in poor movement adaptability because context and postures vary when tasks are being repeated. Coordinative patterns must be flexible because each movement variation may require a slightly different muscle activation pattern (Thelen & Corbetta, 1994; Ulrich et al., 1997). If the coordinative patterns are not yet formed, orthoses could limit the development of flexibility in these patterns.

During skill acquisition, the orthoses may limit infants' ability to fully explore movement patterns. Exploration of movement leads to the development of a varied movement repertoire through the formation and shaping of neuronal connections (Sporns & Edelman, 1993). Early exploration acts as a perceptual modality that allows infants to build a sense of where their limbs are in space how to use their muscles to changes this (Thelen & Corbetta, 1994; Thelen & Smith, 1994). By limiting the infant's movement ability at the foot and ankle, orthoses decrease the amount of perceptual information that goes into the formation of neural connections and into the infants' sense of body awareness.

The outcomes of orthotic use are potentially very different in children with DS who have already acquired a varied movement repertoire. Parents often report that older children with DS have difficulty keeping up with their peers or participating in recreational activities along side children with TD (Sayers Menear, 2007). In this case, the musculoskeletal system of the children with DS may not be able to adequately support the activities they wish to pursue. For instance, many children with DS maintain their feet in a pronated position throughout their gait cycle. This may require increased energy expenditure during walking and running because the foot never reaches a supinated position where it is used as a rigid lever arm for push off (Cioni, Cocilovo, Rossi, Paci, & Valle, 2001). Increased energy expenditure could make it difficult for children with DS to keep up with their peers. In addition, decreased muscle reaction times make balance difficult for children with DS (Shumway-Cook & Woollacott, 1985a). This could negatively impact the ability of children with DS to participate in sports with TD children that require balance and agility such as soccer or basketball. In

these cases, an intervention that improves the biomechanical alignment could improve the children's ability to participate in games and activities with their peers.

Orthoses help to facilitate biomechanical alignment and functioning. In children whose motor skills are stable but whose feet and ankles are poor at biomechanically supporting their behavior, the use of orthoses may switch behavior into a more participatory mode; Biomechanical alignment is the control parameter and the orthoses support the appropriate alignment allowing for the emergence of the new behavior. In children over 3 with DS, orthoses have been shown to improve gait parameters and motor skills level, as demonstrated by standardized test scores (Martin, 2004; Selby-Silverstein, Hillstrom, & Palisano, 2001). These positive outcomes associated with orthotic use are similar to the gait and balance outcomes found in this dissertation study, however, this study has also revealed negative effects of the orthoses on adaptive motor skills and manual exploration. This may be due to the children's skill level at the time of the intervention. Though the orthoses may have a negative effect during the time of skill acquisition, they could enhance movement once the skills have been learned.

Orthoses may limit the exploration of movement, or they could enhance the use of movement for the exploration of the environment. They can potentially be prohibitive or adaptive. The question is when are orthoses prohibitive or adaptive? The answer might lie within a continuum of importance between movement exploration versus environmental/societal exploration with upright mobility skills. While there are components of both in every movement, movement exploration appears to be more important in infancy and environmental/social exploration may gain importance into childhood. The trick for therapists and other health professionals is recognizing when

skill acquisition has given way to an increased focus on environmental exploration and social interactions using movement as a means to achieve these goals. At this point, therapists must determine if orthoses could help improve the child's ability to participate in appropriate social situations.

Contributing systems

We may be able to solve this problem for each child individually by examining the development of the subsystems that contribute to walking. In the process of development, we often think of the subsystems' states as control parameters because they act like rate-limiters to the emergence of a new behavior. These subsystems develop at different rates in a non-linear way (Thelen, 1989). The interaction of the subsystems, at anytime, both constrain and allow for the emergence of behaviors. By following the development and interaction of subsystems, relevant control parameters for upright stepping and walking behavior can be identified and provide information on the appropriateness of orthotic use.

The primary question is which subsystems play a role in upright mobility. Beyond that we need to distinguish between subsystems that play a larger role in the acquisition of the skill and subsystems that are prominent once the skill is learned. Thelen (1986) suggested that, on an organismic level, pattern generation, articulator differentiation, postural control, visual flow sensitivity, tonus control, extensor strength, body constraints, and motivation are important subsystems for locomotor development. However, the use of orthoses may not affect body constraints or motivation in children with DS. Thelen suggests that balance and strength are the most important of these to the emergence of walking in infants with typical development while others are already in

place and others such as visual flow sensitivity develop further with the onset of walking (Thelen, 1986).

Pattern generation is one of the subsystems that are already in place at a young age. Very young infants display coordinated stepping patterns when held in upright. However, these stepping patterns are not similar to the stepping patterns found in walking (Thelen & Fisher, 1982). How do these early patterns influence later walking? Thelen (1986) suggests that the synergies of these early patterns develop and change, though not in isolation, leading to a mature gait pattern. In children with DS, this development takes longer than in children with TD (Palisano et al., 2001; Ulrich, Ulrich, Angulo-Kinzler, & Yun, 2001). However, the ability to produce a gait-like alternating stepping pattern is present in infants with DS at 11 months, long before walking onset (Ulrich, Ulrich, Collier, & Cole, 1995). That is not to say that the alternating stepping pattern is obligatory or unchangeable. In fact, if orthoses are used while infants are learning how to exploit the developing gait pattern, they may influence the development of the appropriate muscle synergies around the foot and ankle.

It has long been postulated that the rhythmical patterns seen in walking are produced by a central pattern generators (CPGs). Forssberg (1985) proposed that infants rely on an innate CPG for primitive stepping. Additionally, he proposed that as the nervous system matures, higher brain level systems begin to influence the network. However, updated views of CPGs are less hierarchical. Cheron and colleagues showed that CPGs are tuned by activity and environment, leading to rapid changes in the first 6 months of walking experience (Cheron, Bengoetxea, Bouillot, Lacquaniti, & Dan, 2001; Cheron, Bouillot et al., 2001). Because CPGs are tuned by sensory feedback from

activity, restricting movement may have a negative effect on the neural networks and neuromuscular control. Orthoses limit the available movement of the foot and ankle in an attempt to provide stability. Orthoses may not be an appropriate intervention during this period of CPG fine-tuning.

Other subsystems that Thelen (1986) proposed as contributing to locomotion are articular differentiation and tonus control. During the process of articular differentiation, early synchronous joint movements become differentiated as muscle-firing patterns move from co-activation to reciprocal activation. This frees the joints to achieve a mature gait pattern.

An immature control pattern is often characterized by tonic background activity and antagonist co-activation. This tonic activity decreases over time (Hadders-Algra, 1993). Experience plays a large role in the development of muscle activation patterns. In a case study on the development of walking, Okamoto and colleagues (2003) found that as the child progressed from infant stepping to supported walking, antagonist co-activation decreased and reciprocal activation became more and more likely. At the onset of independent walking, antagonist co-contraction became the primary muscle activation pattern seen. When the task changed from a familiar task to a novel one, muscle activation patterns changed as well. This is also true in people with DS. People with DS also display co-contraction during novel tasks to stabilize their bodies (Aruin, Almeida, & Latash, 1996). However, with practice, there is a decrease in co-contraction patterns in people with DS (Almeida, Corcos, & Latash, 1994). It appears that the effect of experience on muscular activation is task specific. When observing the developmental

process of bipedal walking, it becomes clearer that task specific experience may be the parameter that controls muscle activation patterns.

If infants with DS learn to walk with orthoses on, articular differentiation and muscular activation patterns may be effected in multiple ways. If the ankle is always supported by the orthoses, the progression from co-activation to reciprocal activation described by Okamoto and colleagues (2003) for children with TD and Almeida and colleagues (1994) for people with DS may be inhibited. The lack of active practice would make the emergence of a reciprocal activation patterns less likely. In the extreme case, there could be little to no activation pattern if the infant is relying on relies totally on the orthoses for support instead of the musculoskeletal system. In addition, removing the orthoses from a child who is walking independently would constitute a novel task and could lead to a pattern of increased co-activation. From an intervention standpoint, orthotic use would be most appropriate once a reciprocal muscle activation pattern is present in independent walking.

As Thelen (1986) states, postural control is an important subsystem to the locomotor system. It has elements in place from before independent walking occurs and continues to develop with walking experience. Children with DS have delayed and atypical postural control when compared to children with TD. Haley (1986) found that infants with DS develop postural reactions and motor skills at a slower rate than children with TD. In addition to this, Shumway-Cook and Woollacott (1985b) found that children with DS develop postural control in a delayed and atypical manner when compared to children with TD. Children with DS between the ages of 1 and 3 have sway responses that are slow, poorly organized, and inconsistent compared to the consistent sway

response seen in children with TD at the same age range (Shumway-Cook & Woollacott, 1985b). This atypical postural response often leads to loss of balance and may influence a child's ability to explore multiple environmental contexts. The use orthoses may help to stabilize the child by physically limiting the amount of sway around the ankles but if used too soon may inhibit the development of consistent and organized sway patterns.

Others have looked at dynamic postural control as it relates to learning to walk. It is apparent that the postural control learned in independent sitting and static stance, continue to improve with increased movement experience and contribute to toddlers' ability to walk independently. Bril and Breniere (1993) looked at multiple parameters to assess the postural control of their subjects with TD including step length, step width, relative double support time, and vertical acceleration of the center of mass. Bril and Breniere found that in the first 4 months of walking experience toddlers have a larger relative double support time. This is needed because toddlers are in a state of vertical fall at foot contact. Step width also decreases over this period. Bril and Breniere have called the first 4 months of walking experience the "integration phase." People with DS continue to show a larger step width throughout childhood (Parker, Bronks, & Snyder, 1986). When compared with children with TD, the gait of 7-year-old children with DS looks immature and displays less dynamic stability (Parker & Bronks, 1980). Dynamic postural control plays a role in the atypical gait pattern of children with DS. Orthoses may provide support at the foot and ankle to improve dynamic postural control. However, the first few months of walking experience is an important period of rapid change leading to the intergration of upright postural control and forward movement (Bril

& Breniere, 1993). Perhaps the use of orthoses should wait until after the period of rapid change though this period may be longer than 4 months in children with DS.

Thelen (1986) mentions visual flow sensitivity as another contributing subsystem to locomotor development. Visual flow sensitivity is part of a larger perception action coupling system. Perception action coupling is based on experience. As a person moves and experiences the environment, the sensory and motor experiences are coupled. This bond allows for sensation to modify motor action and also allows for motor experience to shape perception. Sveistrup and Woollacott (1993) point out that the perception action system is adaptable and changes over developmental time. They state that as the strength and type of perceptual input available changes, our internal reference changes leading to differences in movement responses. In other words, experience is important to the development of sensory systems. This is true even at a cellular level. In a review of the topic, Grubb and Thompson (2004) point out that timing of action potentials not only signals sensory information but also instructs the development of sensory systems. Experience shapes the development of the sensory systems and perception of movement leads to both long-term and short-term adaptations to our movement patterns. By limiting the movement and experience of movement at the ankle during skill acquisition, orthoses may limit the development of the sensory system, and lead to decreased adaptability of movement patterns. Once walking is a stable skill in children with DS, the use of orthoses may not have such a large affect on the perception-action system and may, in fact, positively contribute to the development of aspects of the perception-action system such as visual flow sensitivity and the development of affordances.

As implied by Thelen and colleagues's (1984) studies on anthropometric measurements in relation to newborn stepping, adequate strength to move the legs in an upright position is also a possible control parameter for gait. While strength was not directly measured, it appears that rapid growth leads to a disproportionate amount of weight for muscle strength and activities that were once achievable become impossible. Rapid growth in the first 2-4 weeks of life is associated with decreased stepping (Thelen Thelen, Fisher, & Ridley-Johnson, 1984). To determine whether this relationship was causal, Thelen and colleagues manipulated the mass of infants' limbs by adding weight or placing them in water to increase buoyancy. She found that weighted limbs showed less stepping and buoyant limbs increased their stepping. Thelen concluded that strength might limit behavior. Because strength measurements cannot be taken directly in infants, there are few studies that directly explore the role of strength in the development of gait. However, strength does appear to play a major role in the ability to perform upright locomotor tasks especially during periods of rapid growth.

Postural control provides insight into the importance strength in walking development. Breniere and Bril (1998) view the vertical acceleration of the center of mass as a measure of how muscular forces control external forces. They conclude that decreased muscular force or a poor use of muscular strength, as indicated by a lack of postural control, leads to a "rigid link" between vertical acceleration of the center of mass and velocity during the first year of walking experience. Strength does not only effect gait development during its very early phases but may also have an impact in newly independent walkers.

In the first year of walking, strength continues to shape walking kinetics and walking velocity. Early orthotic use may lead to a "learned non-use" situation in which the child relies on the brace for support instead of muscular activation. In this situation, the child with DS, who has decreased strength to begin with (Rast & Harris, 1985), does not develop an adequate amount of strength in the gastrocnemius. On the other hand, people with DS require more energy to walk than people with TD (Cioni et al., 2001). Orthotic use may provide an external source of stability and allow a child to perform an established motor task with greater ease because the muscles do not have to do as much work to support and position the foot and ankle.

Suggestions for intervention

Considering these contributing systems, perhaps it is possible to make recommendations about appropriate interventions for children with DS. In infancy, the primary focus of motor development and in turn intervention is learning to move (skill acquisition). Once the skills are learned, the focus shifts to learning about the surrounding world by moving through it. Early development that contributes to upright locomotion includes subsystems of pattern generation, joint differentiation and tonus control, and body constraints. Strength, postural control, and perception-action coupling begin to develop prior to walking onset and continue to develop with independent walking. The differences in motor development between infancy and early childhood lead to very divergent intervention styles at a various developmental levels.

As mentioned above, infancy is a period focused on developing motor skills and movement foundations. During this time, intervention should focus on the effect of movement on the developing perception action and neuromuscular systems. It should

include the use of motor skills in multiple setting and contexts that allow the infants to explore and refine their own movement characteristics. This would lead to increased movement adaptability and movement exploration. If we take the idea of contributing movement systems seriously, it becomes obvious that therapists can affect locomotor outcome long before we see signs of walking readiness. Specific early interventions that may influence walking include early upright positioning to influence postural control, perception action coupling, and extensor strength; motivational toys designed to increase supine kicking which will assist with joint differentiation and antigravity strength; and promoting early stepping through interventions such as treadmill training to improve pattern generation, joint differentiation, and strength. All of these activities are task specific but not overly structured. They allow for the exploration of movement within a contextual framework. The therapist's role is to provide a multitude of contexts for practice as opposed to providing feedback for quality of movement.

Movement in early and later childhood focuses on the exploration of the physical and social world surrounding a child. During this time, intervention should focus on efficiency of movement and solving motor problems that inhibit the child's participation. It should include quality of movement and task specific interventions to improve ability to participate in social play and environmental exploration. This would lead to specific changes in movement that affect the child's quality of life. In this context, it would be appropriate to provide biomechanical interventions, such as orthoses, to support the available movement patterns and affect contributing systems such as balance. In contrast to interventions in infancy, these interventions are also task specific but are much more

structured to make specific changes in movement that allow the child to participate more fully within their environment.

References

Adolph, K. E., Vereijken, B., & Shrout, P. E. (2003). What changes in infant walking and why. *Child Development, 74*(2), 475-497.

Almeida, G. L., Corcos, D. M., & Latash, M. L. (1994). Practice and transfer effects during fast single-joint elbow movements in individuals with Down syndrome. *Physical Therapy, 74*(11), 1000-1012.

American Academy of Pediatrics, Counsel on Genetics. (2001). American Academy of Pediatrics: Health supervision for children with Down syndrome. *Pediatrics, 107*(2), 442-449.

Aruin, A. S., Almeida, G. L., & Latash, M. L. (1996). Organization of a simple two-joint synergy in individuals with Down syndrome. *American Journal of Mental Retardation, 101*(3), 256-268.

Bernstein, N. (1967). *The co-ordination and regulationof movements*. Oxford: Pergamon Press.

Breniere, Y., & Bril, B. (1998). Development of postural control of gravity forces in children during the first 5 years of walking. *Experimental Brain Research, 121*, 255-262.

Bril, B., & Breniere, Y. (1993). Posture and independent locomotion in early childhood: Learning to walk or learning dynamic postural control? In G. J. P. Savelsbergh (Ed.), *The development of coordination in infancy* (Vol. 97, pp. 337-358). Amsterdam: Elsevier Science.

Cheron, G., Bengoetxea, A., Bouillot, E., Lacquaniti, F., & Dan, B. (2001). Early emergence of temporal co-ordination of lower limb segments elevation angles in human locomotion. *Neuroscience Letters, 308*(2), 123-127.

Cheron, G., Bouillot, E., Dan, B., Bengoetxea, A., Draye, J. P., & Lacquaniti, F. (2001). Development of a kinematic coordination pattern in toddler locomotion: planar covariation. *Experimental Brain Research, 137*(3-4), 455-466.

Cioni, M., Cocilovo, A., Rossi, F., Paci, D., & Valle, M. S. (2001). Analysis of ankle kinetics during walking in individuals with Down syndrome. *Americal Journal of Mental Retardation, 106*(5), 470-478.

Forssberg, H. (1985). Ontogeny of human locomotor control I. Infant stepping, supported locomotion and transition to independent locomotion. *Experimental Brain Research, 57*, 480-493.

Grubb, M. S., & Thompson, I. D. (2004). The influence of early experience on the development of sensory systems. *Current Opinion in Neurobiology*(14), 503-512.

Hadders-Algra, M. (1993). General movements in early infancy: what do they tell us about the nervous system? *Early Human Development, 34*, 29-37.

Haley, S. M. (1986). Postural reactions in infants with Down syndrome. *Physical Therapy, 66*(1), 17-22.

Heriza, C. B. (1991). Implications of a Dynamic-Systems Approach to Understanding Infant Kicking Behavior. *Physical therapy, 71*(3), 222-235.

Martin, K. (2004). Effects of supramalleolar orthoses on postural stability in children with Down syndrome. *Developmental Medicine and Child Neurology, 46*, 406-411.

Merzenich, M., Wright, B., Jenkins, W., Xerri, C., Byl, N., Miller, S., et al. (1996). Cortical plasticity underlying perceptual, motor, and cognitive skill development: implications for neurorehabiliation. *Cold spring harbor symposia on quantitative biology, LXI*, 1-8.

Newell, K. M. (1986). Constraints on the development of coordination. In W. M. G. & H. T. A. Whiting (Eds.), *Motor Development in Children: Aspects of Coordination & Control* (pp. 341-360). Dordrecht: Martinus Nijhoff.

Okamoto, T., Okamoto, K., & Andrew, P. D. (2003). Electromyographic developmental changes in one individual from newborn stepping to mature walking. *Gait & Posture, 17*(1), 18-27.

Palisano, R. J., Walter, S. D., Russell, D. J., Rosenbaum, P. L., Gemus, M., Galuppi, B. E., et al. (2001). Gross motor function of children with Down syndrome: creation of motor growth curves. *Archives of Physical Medicine & Rehabilitation, 82*, 494-500.

Parker, A. W., & Bronks, R. (1980). Gait of Children with Down Syndrome. *Archives of Physical Medicine & Rehabilitation, 61*, 345-351.

Parker, A. W., Bronks, R., & Snyder Jr, C. W. (1986). Walking patterns in Down's syndrome. *Journal of Mental Deficiency Research, 30*, 317-330.

Rast, M. M., & Harris, S. R. (1985). Motor control in infants with Down syndrome. *Developmental Medicine & Child Neurology, 27*(5), 682-685.

Sayers Menear, K. (2007). Parents' perceptions of health and physical activity needs of children with Down syndrome. *Down Syndrome Research and Practice, 12*(1), 60-68.

Scholz, J. P. (1990). Dynamic pattern theory--some implications for therapeutics. *Physical Therapy, 70*(12), 827-843.

Selby-Silverstein, L., Hillstrom, H. J., & Palisano, R. J. (2001). The effect of foot orthoses on standing foot posture and gait of young children with Down syndrome. *Neurorehabilitation, 16*(3), 183-193.

Shumway-Cook, A., & Woollacott, M. H. (1985a). Dynamics of postural control in the child with Down syndrome. *Physical Therapy, 65*(9), 1315-1322.

Shumway-Cook, A., & Woollacott, M. H. (1985b). The growth of stability: postural control from a development perspective. *Journal of Motor Behavior, 17*(2), 131-147.

Spencer, J. P., & Thelen, E. (2000). Spatially specific changes in infants' muscle coactivity as they learn to reach. *Infancy, 7*(5), 275-302.

Sporns, O., & Edelman, G. M. (1993). Solving Bernstein's problem: a proposal for the development of coordinated movement by selection. *Child Development, 64*(4), 960-981.

Sveistrup, H., & Woollacott, M. H. (1993). Systems contributing to the emergence and maturation of stability in postnatal development. In G. J. P. Savelsbergh (Ed.), *Development of coordination in infancy.* (pp. 319-336): North-Holland/Elsevier Science Publishers.

Thelen, E., & Corbetta, D. (1994). Exploration and selection in the early acquisition of skill. *International Review of Neurobiology, 37*, 75-102.

Thelen, E., & Fisher, D. M. (1982). Newborn stepping: An explanation for a "disappearing" reflex. *Developmental Psychology, 18*(5), 760-775.

Thelen, E., Fisher, D. M., & Ridley-Johnson, R. (1984). The relationship between physical growth and a newborn reflex. *Infant Behavior and Development, 7*, 479-493.

Thelen, E. (1992). Development of Locomotion from a Dynamic Systems Approach. In H. Forssberg & H. Hirschfeld, (Eds.), *Movement Disorders in Children* (Vol. 36, pp. 169-173). Basel: Karger.

Thelen, E. (1989). Self-organization in developmental processes: Can systems approaches work? In M. Gunnar & E. Thelen (Eds.), *Systems and Development* (Vol. 22, pp. 77-117). Hillsdale: Lawrence Erlbaum Associates.

Thelen, E., & Smith, L. B. (1994). Lessons from learning to walk. *A dynamic systems approach to the development of cognition and action* (pp. 3-20). Cambridge: MIT press.

Thelen, E., (1986). Development of coordinated movement: Implications for early human development. In M. D. Wade & H. T. A. Whiting (Eda.), *Motor Development in Children: Aspects of Coordination and Control* (pp. 107-124). Dordrecht: Martinus Nijhoff Publishers.

Ulrich, B. D (1997). Dynamic Systems Theory and Skill Development in Infants and Children. K. J. Connolly& H. Forssberg (Ed.), *Neurophysiology & Neuropsychology of Motor Development* (pp. 319-345). London: MacKeith Press.

Ulrich, B. D., Ulrich, D. A., Collier, D. H., & Cole, E. L. (1995). Developmental shifts in the ability of infants with Down syndrome to produce treadmill steps. *Physical Therapy, 75*(1), 14-23.

Ulrich, D. A., Ulrich, B. D., Angulo-Kinzler, R. M., & Yun, J. (2001). Treadmill training of infants with Down syndrome: evidence-based developmental outcomes. *Pediatrics, 108*(5), E84.

Yaguramaki, N., & Kimura, T. (2002). Acquirement of stability and mobility in infant gait. *Gait & Posture, 16*(1), 69-77.

Appendix 2. Behavior Coding Document

To code, use the Behavior Coding form:

Time	Upright	Leaning	# of Feet	Support Foot	# Hands	Support Hand	Hand Action	Crusing	Direction of Crusing	#of Steps	Pull to Stand

Definitions:
In the "time" box, write the time indicated on the screen (including all of the numbers in the time code) Fill out the line for that time

Check "upright" if the child is in an upright position using either his trunk or arms for support. If the child is moving into upright, upright starts when the child's hip is no longer moving upward. To determine this, the hip should not move upwards in the next 30 frames of video. If this is the case, mark upright at the first frame of the 30. If this is not the case, continue to watch for 30 consecutive frames when the hip is no longer moving upward. If the child is not upright, do not fill out the row

Check "leaning" if the child's midsection is touching the table for more than 30 frames. As with upright, mark leaning at the first frame of the 30. If the child's midsection does not touch the table for 30 frames or more, DO NOT mark leaning.

Record the "# of hands" that are on the support surface. For this to change, the child must raise or lower his hand for 30 frames or more. If the child is standing with 2 hands on the support surface and then lifts one one for 29 frames, do not consider this a change to 1 hand. If the child is standing with 2 hands on the surface and then lifts one hand for 30 frames or more, consider the first frame when the hand came off the surface as a change to 1 hand. Likewise, if the child has one hand off of the surface and lowers the hand for 29 frames or less do not consider this a change to 2 hands. If the child is has one hand off of the surface and lowers the hand for 30 or more frames consider this a change to 2 hands.

Mark which hand is the "support hand." If the child has one hand on the support surface, mark which hand is down. Every time the "# of hands" box says "1" this box must be marked. If the "# of hands" box say "0" or "2" do not mark this box.

Mark the "hand action" box. If the child has one hand on the support surface, mark what the other hand is doing. The choices are:

"S" playing with a stationary toy (a toy whose base does not move)
"M" playing with a moving toy (a toy whose base moves)
"R" reaching (not playing with a toy. The hand is in the air for 30 frames or more)

If the child has 2 hands on the surface, do not mark this box.

Record "# of feet" that are on the ground. For this to change, the child must raise or lower his foot for 30 frames or more. If the child is standing with 2 feet on the floor and then lifts one foot for 29 frames, do not consider this 1-foot standing. If the child is standing with 2 feet on the floor and then lifts one foot for 30 frames or more, consider the first frame when the foot came off the ground as 1-foot standing. Likewise, if the child is standing with one foot off the ground and lowers the foot for 29 frames or less do not consider this a change to 2 feet. If the child is standing with one foot off the ground and lowers the foot for 30 or more frames consider this a change to 2 feet.

Mark which foot is the "support foot." If the child is in 1 foot standing, mark which foot is on the ground. Every time the "# of feet" box says "1" this box must be marked. If the "# of feet" box say "2" do not mark this box.

If the child is stepping sideways while holding onto the surface mark the "Cruising" box. In order to be cruising, the child must pick up one foot and place it down in a different place and then move the other foot toward the first foot. Cruising time begins with the first step.

Mark the "direction of cruising" box in relation to the child facing the support surface.

"L" if the child is moving to his left

"R" if the child is moving to his right

If the child is not cruising leave this box blank

Mark the "number of steps" that the child is cruising. Each foot counts as one step. The child needs to take 2 steps to be "cruising"

Mark the "pulls to stand" box when the child pulls to stand from the floor. Just mark once for each episode.

Directions for filling out the form:

Code the first twenty minutes of play beginning when no one is touching the child. There will probably more than 20 minutes of activity taped. Make your last observation at the time point 20 minutes after the start.

The time code looks like this: hh:mm:ss.frames. There are 30 frames of video per second. Be sure to write all of the numbers and the **EXACT** frame that an activity begins.

Begin by writing the time code in the "Time" column when the researcher first releases the child. Then fill in the appropriate information in that row for that frame of video. When one of the conditions changes, write the time code in the "Time" column and fill in that row for that frame of video. Continue doing this until you have coded all twenty minutes of video.